Microscopic examination of rib heads: a useful adjunct in the investigation of infant deaths

Walter Loren Kemp
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MICROSCOPIC EXAMINATION OF RIB HEADS:
A USEFUL ADJUNCT IN THE INVESTIGATION OF INFANT DEATHS

By

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Dissertation
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The University of Montana, Missoula, MT

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Microscopic examination of ribs: A useful adjunct in the investigation of infant deaths

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The purpose of this study was to examine infant rib heads microscopically, document clefts in the anterior portion of the metaphysis and other findings including metaphyseal lesions, and statistically analyze these clefts and various features about the child, including circumstances of death (e.g., SIDS, bed sharing, or suspicious for abuse), whether or not CPR was performed, birth method, age in months at death, estimated gestational age at birth, and various socio-economic factors (e.g., married or unmarried mother, biological father or boyfriend involved with care of child). The glass slides with microscopic sections of rib head, neck and variable amounts of adjacent shaft previously obtained at autopsy from 90 children were used. The clefts identified by the author in two sentinel cases were found in a majority of the other 88 children, and were consistent morphologically with a fracture, indicating that children’s ribs are easier to break than previously thought. The number of clefts per child was strongly associated with age in months at death. The number of clefts greater than 1.00 mm in length per child was unequally distributed amongst groups differentiating the circumstances of death, and found in greater number in the group representing suspicious deaths, implying that this microscopic finding may indicate abuse. No statistical association between the clefts and birth method or use of cardiopulmonary resuscitation during the terminal event was identified; however, single cases suggested otherwise. A statistical association between estimated gestational age and number of clefts per child was identified via correlation using Spearman’s method, but not other statistical analyses (i.e., Chi-square test and Poisson regression analysis). This study supports the microscopic examination of infant rib heads obtained at autopsy, as potentially significant findings (including metaphyseal lesions) that are not identified via radiologic or gross examination at autopsy would otherwise be missed; however, further investigation is required to better understand the implications of the histologic findings.
DEDICATION

To my beautiful wife, Kelly, for the love, patience, tolerance, support, and understanding that she has shown me over the past 5 years.

To my good friend, Brian Kieffer, for always serving as an example to me that any trials and tribulations I face in my life are small compared to that faced by others, and I hope I can always face mine with the patience, acceptance, and humor that he has shown when facing his.

To my Aunt Lucille Bolyard, one of the most kind and generous people I have ever known, I sincerely regret not having shown and told you more how much I appreciated having you in my life and how you helped me. I only hope that I can be as kind and generous to others as you were to me.
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To my mom and dad, their support and encouragement of my reading as a child surely played a significant role in my life.

According to the State of Montana’s Office of Vital Statistics, the following statement must be included in this paper: “Data used in this study (report) were supplied by the Office of Vital Statistics, Montana Department of Public Health and Human Services. The Office of Vital Statistics is not responsible for the conclusions of the report or the analysis on which any conclusion is based.”
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CHAPTER 1: INTRODUCTION

Child abuse is an unfortunate, but real part of our society. The manifestations of child abuse are myriad and include bruises, fractures, intracranial hemorrhage, and injuries of internal organs (Wetli et al., 1999; DiMaio and DiMaio, 2001; Glass et al., 2002; Dolinak and Matshes, 2005). Multiple professions deal with child abuse through the diagnosis and treatment of abused children, investigation of the circumstances of the abuse, or prosecution of the abuser. Those professionals who perform these duties include pediatricians and other physicians, nurses and other health care workers, law enforcement officers, county and district attorneys, social workers, forensic pathologists, and forensic anthropologists. The physical identification of child abuse, as confessions from the perpetrator may be lacking, involves interpretation of patterns of injuries, which can include fractures of the skeletal system. Understanding the patterns of skeletal injury indicative of child abuse is especially important to forensic anthropologists, who are often consulted by forensic pathologists to aid in the interpretation of these findings.

Forensic anthropology represents a subgroup of both physical anthropology and applied anthropology. Forensic anthropologists use their knowledge of osteology, skeletal biology, and paleopathology to analyze human remains in the context of criminal investigation, both historical and current, or when a death is unexplained, or decomposition has rendered soft tissue analysis problematic (American Board of Forensic Anthropology, N.d.). Although the majority of work conducted by forensic anthropologists involves inspection of bones, either grossly or with the aid of a low power microscope, such as a dissecting microscope, in some circumstances, forensic anthropologists utilize higher power microscopic examination of bones to perform their duties (Kerley, 1965; Crowder and Stout, 2012). As forensic anthropologists are frequently consulted
by forensic pathologists to assist with the investigation of deceased individuals who are either skeletonized or have skeletal trauma, their knowledge of techniques to examine bone microscopically for injuries, which can be shared with forensic pathologists, is at the very least useful. As part of their duties, forensic anthropologists assist with the investigation of child abuse. Therefore, knowledge of microscopic features of the skeletal system indicative of child abuse would be of importance to a practicing forensic anthropologist.

Within the field of anthropology, knowledge of osteological features used to identify traumatic infant deaths and child abuse is not only important to forensic anthropologists, but also to other anthropologists, including other physical anthropologists, archaeologists, and cultural anthropologists, to allow for accurate interpretation of findings. While contrasting views of the presence or absence of child abuse in past populations are present in the literature, that parents can kill their children is well documented. For example, Freeman (1971) examined the adaptive significance of systematic infanticide in Eskimo society based upon an ecological and ethnographic perspective, concluding that female infanticide led to an increase in population stability. Green and Beckwith (1924), in a review article, discussed infanticide among Hawaiian populations, indicating it was practiced to preserve rank or because people needed to rid themselves of additional work (i.e., that of raising a child). However, regarding child abuse itself, Neves et al. (1999:257) reviewed skeletal remains from 244 individuals from three sites near San Pedro de Atacama, and, in interpreting the fractures, concluded that “the virtual absence of fractures in the large sample of infants and children is indicative of an absence of traumatic child abuse…”. In agreement, Walker (1997) states, “…child abuse resulting in severe skeletal trauma is primarily a modern phenomenon.” However, Lewis (1997) disagrees with
Walker (1997) and opines that child abuse is not a modern phenomenon. In agreement with Lewis (1997), Roberts and Manchester (2007:118) state, “The identification of child abuse in the archaeological record on the basis of injury to the skeleton is another area of investigation that is seeing some interest.” Given this discrepancy in opinions, more information is needed, or possibly, knowledge of a subtle marker of child abuse is required. Another example of the importance of knowledge of osteological features of injury outside of the practice of forensic anthropology is Larsen (1997), a text on bioarchaeology, which has a chapter on injury and violent death, and subsections on intentional injury, interpersonal violence, and the interpretation of skeletal trauma. Therefore, as anthropologists other than forensic anthropologists address skeletal features of child abuse in their publications, increased understanding of osteological features of child abuse would aid not only forensic anthropologists but also other anthropologists, including archaeologists. Although the amount of literature available to archaeologists to interpret skeletal trauma is large, the amount of literature available to help interpret injuries in the context of child abuse is less impressive, with Walker (1997) being the only source cited by Roberts and Manchester (2007) in the section of their review book describing child abuse.

In their duties, forensic anthropologists often collaborate with forensic pathologists in criminal investigations, assisting with, among other areas, the documentation of skeletal trauma (American Board of Forensic Anthropology, N.d.). However, while textbooks by forensic anthropologists may specifically address child abuse in a separate section (Galloway, 1999; Klepinger, 2006), the number of articles in the literature by forensic anthropologists specifically addressing child abuse is relatively small (Kerley, 1976; Kerley, 1978; Walker et al., 1997), and,
while knowledgeable of bone and its pathology, forensic pathologists do not have the expertise possessed by forensic anthropologists in this area; thus, collaboration between the two fields in the area of child abuse would be beneficial to both.

Forensic pathologists investigate sudden and unexpected deaths for the purpose of determining the cause and manner of such deaths. The cause of death is the condition that set in motion the chain of events that led to death (Hanzlick et al., 2002). For example, metastatic carcinoma, a gunshot wound of the head, and chronic alcoholism are all causes of death. In contrast, there are only five manners of death in most jurisdictions: natural, accident, homicide, suicide, and undetermined. In general, accident is certified when a death occurred due to an injury that resulted from an action with no intent to cause harm or death. Suicide is certified when a death occurred as the result of an intentional and self-inflicted action with the intent to do self-harm. Homicide is certified when a death resulted from an intentional act committed by another person with the purpose of causing fear, harm, or death—intent to cause death alone is not required. Natural is certified when the death is solely or almost solely due to a disease or the aging process, and a death is certified as undetermined manner when the information available does not reliably indicate a natural, accident, suicide, or homicide manner of death (Hanzlick et al., 2002).

In addition to the determination of cause and manner of death, forensic pathologists also perform autopsies to document natural disease and injuries, and, in the process, may collect evidence for later use. If the cause of death was an injury, forensic pathologists, if possible, help to identify the instrument used. Identification of an instrument used may involve collection of fragments of the instrument from the body (e.g., the tip of a knife lodged in a bone) or
interpretation of injuries and correlation with different weapons (e.g., matching patterned abrasions on the body to a telephone cord). However, instruments also include hands and feet and evidence can include fractures of a bone. Coordination between forensic anthropologists and forensic pathologists as to removal of bone likely to be of evidentiary value in identifying the instrument responsible for bone trauma is important.

In their capacity to determine the cause and manner of death, forensic pathologists employ a variety of techniques including gross examination, radiology, and chemical analysis (including toxicologic testing for the presence of medications and illicit drugs). Forensic pathologists also may examine tissue underneath the microscope to detect abnormalities that cannot be seen upon gross examination. In addition to the use of various techniques and methods, forensic pathologists also use a variety of consultants to help determine cause and manner of death, including neuropathologists, pediatric pathologists, and, when injuries involve the skeletal system, especially in the case of a homicide, either a child or an adult, a forensic pathologist will often consult with a forensic anthropologist to aid in the correct interpretation of the traumatic injury.

The state of Montana employs two forensic pathologists as state and deputy state medical examiner to assist the county coroners who are the primary investigating agents for sudden and unexpected deaths and who determine the cause and manner of death in such situations. The duties of the two medical examiners include providing assistance to coroners and law enforcement, stimulating and directing research in the field of forensic pathology, and performing autopsies as requested [Montana Forensic Science System Act, MCA 44-3-211 (1989)]. While Montana Code Annotated [MCA] 46-4-122 (1993), which defines deaths in
Montana requiring investigation by the coroner, does not specifically require the investigation of infant deaths (i.e., children under the age of 1 year), infant deaths fall under several other categories, including “a medically suspicious death, unusual death, or death of unknown circumstances” and, in many cases, a death that occurs “in a manner that was unattended or unwitnessed and the deceased was not attended by a physician at any time in the 30-day period prior to death.” Therefore, infant deaths in Montana quite often result in the performance of an autopsy to help determine the cause and manner of that death.

The two State of Montana medical examiners, between 2008 and 2012, conducted 1189 autopsies, of which, 83 involved infants, or 6.6% of the autopsies; however, of 43,979 deaths occurring in Montana from 2006 to 2011, only 375 deaths were of infants under the age of 1 year, or 0.85% of the deaths (Montana Department of Health and Human Services, 2011). While many factors, which need not be discussed here, determine which deaths do or do not result in the performance of an autopsy in the state of Montana, the above numbers at least allow for a rough appreciation that a much higher percentage of infants who die are autopsied than the combined percentage of other age groups that are autopsied. The reason for this is that, while infants do die from known congenital malformations and chromosomal abnormalities and conditions that originated in the perinatal period (99 and 123 infants, respectively, from 2006-2011 in Montana) and are thus most likely known by physicians to have an underlying potentially lethal condition at the time of death, other causes of death in infancy and early childhood are not obvious from the medical history or from investigation. These other causes of death include sudden infant death syndrome (SIDS), bed-sharing (indicating the possibility of accidental overlay), and inflicted trauma. In Montana, of 83 children aged 2 years and younger
autopsied by the two state medical examiners in a four year period, 20 died as the result of SIDS, 12 died as the result of other natural causes diagnosed at the time of autopsy (e.g., bronchopneumonia), and 28 died while bed-sharing with an adult; however, three died from inflicted trauma, and 10 died under suspicious circumstances, or had autopsy findings, which, while suspicious, did not reach a threshold to allow for a definitive diagnosis of inflicted trauma as a cause of death, and so, the cause and manner of death were certified as undetermined. Of note, eight children died under circumstances not suspicious for inflicted trauma, but with an undetermined cause of death among two or more choices, of which possible manners were other than just natural (e.g., bed-sharing), and therefore, the manner of death was undetermined as well. Of note, almost all deaths occurring while bed sharing are assigned an undetermined manner of death, as the cause of death may be either SIDS occurring while the infant was in bed with their parents, or an accidental overlay. Although the circumstance of bed sharing is known at the time of autopsy, an autopsy is required to rule out other possible causes of death. Therefore, although most infant deaths are either natural or probably accidental in origin (e.g., SIDS or accidental overlaying while bed-sharing), a small subset are due to inflicted injury.

In the description of injuries and circumstances associated with child abuse and fatalities, two recurring themes are skeletal injuries and shaking, squeezing, or compressing the infant. One form of skeletal injury that could result from compression of the chest is rib fractures; however, rib fractures can also result from direct blows to the chest wall (Cameron and Rae, 1975; Kleinman, 1990; Kleinman and Schlesinger, 1997). In general, rib fractures are strongly associated with child abuse in the medical and forensic literature and rib fractures in a posterior location are even more strongly associated with child abuse (O’Neill et al., 1973; Akbarnia et al.,
1974; Cameron and Rae, 1975; Thomas, 1977; Merten et al., 1983; Kleinman, 1987a; Zumwalt and Hirsch, 1987; Cohle and Byard, 1994; Strouse and Owings, 1995; Knight, 1996; Kleinman, 1998; Galloway, 1999; Parikh, 1999; Bulloch et al., 2000; Cadzow and Armstrong, 2000; DiMaio and DiMaio, 2001; Hymel and Spivack, 2001; Barsness et al., 2003; Dolinak and Matshes, 2005; Shkrum and Ramsay, 2007; Weber et al., 2009). Nevertheless, not all authors agree that posterior rib fractures are especially suggestive of child abuse. Knight (1996:462) said, “Whether or not such fractures [posterior rib fractures] can be caused by innocent handling, albeit rough or robust, as opposed to angry or impatient violence, is again a matter of dispute, beyond the competence of doctors to resolve—although this does not deter some from expressing strong opinions based upon weak facts.” Knight’s statement, however, is in the minority.

Rib fractures by themselves are not an outright cause of death unless so as to cause a flail chest, which requires three or more ribs to be broken in two or more places, or to later result in pneumonia. Although rib fractures are not by themselves directly a cause of death, if detected upon radiologic examination of infants and children or upon autopsy of deceased infants and children, rib fractures could serve as a marker of another action, such as intentional compression of an infant’s chest. Thus, the finding of rib fractures at autopsy combined with a knowledge that they are due to compression of the chest could provide investigators with more information with which to question caretakers about the care or death of an infant or child; however, the possibility that rib fractures are due to another mechanism must be considered.

Several years ago, I autopsied two deceased infants consecutively, both with grossly detectable rib fractures. In both cases, the autopsy, combined with the death investigation and
additional testing, failed to identify a definitive cause of death; however, some form of fatal abuse was suspected in both cases (note: the mother of one of the infants was subsequently convicted of child abuse). As part of the autopsy, numerous ribs in both cases were examined microscopically, and, at this time, I noted, in many ribs, a small, microscopic cleft, at the anterior aspect of the primary spongiosa in the rib head region (Figs. 1a and b). The clefts contained an amorphous, eosinophilic, acellular material and exhibited variable degrees of healing. The significance of these clefts was uncertain; however, the histologic features of the cleft, especially the features found in the larger clefts, were consistent with a fracture.

Figure 1a. Representative cleft, low power. The arrow indicates the cleft, which is filled with an amorphous eosinophilic material. Hematoxylin and eosin, 40x.

Figure 1b. Representative cleft, high power. The arrow indicates the cleft, which is filled with an amorphous eosinophilic material. Hematoxylin and eosin, 100x.

Following the identification of these clefts, and after further reading regarding the significance of posterior rib fractures, both I and my work partner began to include sections of the rib head as one of our standard areas for histologic examination in the evaluation of infant deaths. In deaths where the ultimate diagnosis was SIDS, these clefts were often present. So, did these clefts represent some form of birth injury, changes induced by resuscitation, or possibly a marker for past or current instances of chest compression by caretakers? If the microscopic rib
head clefts are indicative of intentional compression of the chest, their presence may provide law enforcement officers with useful information when questioning caretakers, or, if microscopic fractures of the rib heads can be shown to occur across a spectrum of infant deaths, they may indicate that handling alone of infants, and not necessarily inflicted trauma, has the potential to fracture ribs. However, only one study exists that has studied microscopic fractures of the rib head (Kleinman et al., 1992), and no studies are known to exist that have studied the incidence of these microscopic clefts of the rib head in association with causes of death in infants other than inflicted trauma. Understanding of the mechanism of formation of these microscopic rib head clefts could aid forensic pathologists and forensic anthropologists in their interpretation of skeletal trauma. As forensic anthropologists serve as consultants to forensic pathologists for the interpretation of skeletal trauma, and as examination of the rib heads is a time-consuming procedure, which many forensic pathologists may be unwilling to undertake, knowledge of their importance and of their possible associations (i.e., with abuse, if such an association exists), would be vital information for the forensic anthropologist consultant to be aware of and willing to help assess for.

These microscopic fractures of the rib head were found in many infants, and across a variety of causes of death (i.e., not solely associated with known child abuse homicides or suspicious deaths); therefore, what is the underlying etiology of the fractures? Are they associated with abuse, simple rough-handling, or with the delivery method of the infant (i.e., vaginal versus Cesarean), or with certain socio-economic factors under which the infant lives (e.g., married versus unmarried mothers)? If the rib head fractures can be found to be associated with abuse, they could serve as a essentially unknown and unexplored pattern of child abuse that
can be used by forensic anthropologists and forensic pathologists in the future when evaluating such deaths; however, to show they are indicative of abuse, other factors (e.g., CPR or delivery method) must be excluded. Also, if the rib fractures can be both associated with abuse and with certain socio-economic factors, they could serve as some validation of the use of certain social features to help identify live children potentially at risk for abuse. Through statistical analysis of various features associated with the microscopic rib head fractures, these questions may be answered.

In summary, several points must be highlighted: 1) during the investigation of all infant deaths, unless the cause of death is readily apparent from the medical history and scene investigation, the diagnosis of child abuse, though uncommon, must always be considered, 2) fatal child abuse, while often producing readily identifiable injuries at autopsy (e.g., subdural hemorrhage, lacerations of internal organs), can also be very subtle, and easily missed at autopsy, 3) common features of child abuse and fatal child abuse of infants often include skeletal fractures as a finding and a combination of squeezing or compression and shaking, 4) rib fractures are strongly associated with child abuse in the medical and forensic literature, 5) the author has identified microscopic clefts in the anterior portion of the rib head at the growth plate, which have features consistent with a fracture, and 6) microscopic studies of the rib head and neck are rare, and have essentially only been conducted on abused infants. Therefore, the purpose of this study is to review the features and associations of microscopic clefts of the rib head identified in autopsy specimens, and identify any associations with birth method, cardiopulmonary resuscitation, age at death, estimated gestational age at the time of birth, socio-economic
conditions under which the child lives, and the possibility of abuse to further our understanding of the importance and implications of the identification of rib head and neck fractures.
CHAPTER 2: REVIEW OF THE LITERATURE

Preview of literature review

To understand the implications of the presence of infant rib head fractures, a knowledge of several different, but related, topics is required. First, as autopsies on infants who have died frequently have no external or internal findings, and, because abuse can be subtle, and not present externally, but found internally, a discussion of SIDS, and its mimics, is important. Second, as infant rib head fractures may indicate abuse, a general discussion of child abuse, including its history, different criteria for its diagnosis, and social factors associated with abuse, including socio-economic conditions, mental status of parents and perpetrators of abuse is required. Third, a general review of bone structure and fracture formation is relevant background information. Fourth, a general discussion of the types of fractures found in abuse, including long bone fractures, spiral fractures, and metaphyseal fractures is necessary to appreciate the wide variety of skeletal findings in child abuse. However, as metaphyseal fractures occur at the same location as the infant rib head clefts studied, a more in-depth introduction to this type of fracture will be performed.

As the main topic of this study is infant rib head fractures, the review of the literature will contain a thorough discussion of the anthropological, medical, and forensic literature as it pertains to rib fractures. This discussion will include 1) association of rib fractures, especially posterior rib fractures, with child abuse, 2) possible causes of rib fractures other than abuse, including cardiopulmonary resuscitation, birth, metabolic disorders (especially osteogenesis imperfecta and prematurity), 3) how to identify rib fractures, 4) the mechanism of rib fractures,
5) the morphology of rib fractures, and 6) a brief review of how fractures in general, including rib fractures, may be dated. Finally, the extensive literature review will be summarized.

**Diagnosis of SIDS**

One common cause of death in infants is Sudden Infant Death Syndrome (SIDS). The definition of SIDS was first proposed in 1969 (Bergman et al., 1970:18) as “the sudden death of any infant or young child, which is unexpected by history, and in which a thorough post-mortem examination fails to demonstrate an adequate cause of death.” Recently, Krous et al. (2004:235), in light of additional information available regarding SIDS, proposed a revised general definition as “SIDS is defined as the sudden unexpected death of an infant <1 year of age, with onset of the fatal episode apparently occurring during sleep, that remains unexplained after a thorough investigation, including performance of a complete autopsy and review of the circumstances of death and the clinical history.” In addition, Krous et al. (2004:236) delineated three categories of SIDS, with each category in part based upon the degree of investigation into the death (e.g., whether or not the investigation included toxicologic, microbiologic, and radiologic components as well as metabolic screening and analysis of vitreous electrolytes), and other considerations (e.g., age outside the normal range for SIDS deaths of 3 weeks to 9 months, marked inflammatory changes not sufficient to cause death); however, in any of the three categories proposed, there could be “no evidence of unexplained trauma, abuse, neglect, or unintentional injury.” Although the exact underlying physiologic mechanism by which the condition causes death is still under investigation, SIDS represents an as yet undefined non-traumatic natural disease process or processes, and the manner of death is therefore natural. SIDS caused the
death of 63 infants between 2006-2011 in Montana, or 16.8% of all infant deaths (Montana Department of Health and Human Services, 2011).

As described above, the diagnosis of SIDS requires a thorough investigation of the circumstances of the death. Nevertheless, even when the cause of death is not SIDS, the investigation of infant deaths must be thorough and is both difficult and challenging, and, unfortunately, oftentimes does not provide a definitive answer for how the death occurred (i.e., the cause of death). While infants can die from cancer, motor vehicle accidents, known congenital malformations, complications of prematurity, and chromosomal abnormalities, most infants who are autopsied, especially in the state of Montana, do not die from these conditions. The above listed conditions result in either expected deaths or otherwise non-suspicious deaths (e.g., a motor vehicle accident witnessed by uninvolved individuals) and, while referral of the death to a coroner may be appropriate, an autopsy to investigate the death is not often required. In contrast, virtually all infants and young children are autopsied when they died under circumstances where the cause of death is not known at the time of death. Some of these infants will ultimately be diagnosed as having died from SIDS, but other causes of death, unexpected or unknown at the time of death, are sometimes identified (e.g., subtle inflicted trauma, undiagnosed congenital heart disease, unsuspected pneumonia).

In addition to SIDS, many infant deaths occur while bed-sharing with parents. Unfortunately, the definitive diagnosis of an accidental smothering due to overlaying the infant while bed sharing is difficult, and, even if the infant did die as the result of SIDS while sleeping in bed with his parents, the diagnosis of SIDS cannot be made because of the circumstances and the possibility of an overlay. The difficulty in diagnosing death due to overlay illustrates the
general problems encountered when investigating infant deaths. The lack of specific autopsy findings for many conditions, both atraumatic (such as SIDS) and traumatic (such as an accidental overlay) hinders the ability of a medical examiner to accurately determine the cause and manner of death. This fact is especially important in regards to infant deaths caused by intentional injury.

While inflicted injury in adults is often very visible, e.g., a gunshot wound, multiple stab wounds, or a severe beating (Cebelin and Hirsch, 1980), inflicted injury in children may be much more subtle, and not readily present on external examination, or even identifiable after autopsy (Zumwalt and Hirsch, 1980). This fact additionally contributes to the difficulties in the investigation of infant deaths--either (and most likely) parents have just lost their child, or, less likely, but still possible, parents or another caretaker have just killed a child, and, based only upon the external appearance of the child, the distinction is not apparent.

The diagnosis of SIDS is essentially a diagnosis of exclusion, meaning the diagnosis is not made based upon a set list of autopsy findings, but, instead on the lack of other more definitive findings (e.g., pneumonia or inflicted trauma). The fact that SIDS is essentially a diagnosis of exclusion and the fact that accidental overlay resulting in asphyxiation is a difficult diagnosis to make at autopsy explains why infants who die while bed sharing, and are usually of the age group in which SIDS occurs, are certified descriptively as “sudden unexpected death while bed-sharing with an adult”, neither confirming the diagnosis of an overlay nor refuting the possibility that the death is due to SIDS. Unfortunately, the fact that SIDS is essentially a diagnosis of exclusion has allowed death investigators to misuse the diagnosis. More than one Montana coroner has been told by an emergency room physician that the cause of death of an
infant is SIDS, because the infant had no external signs of injury and no other known cause of death—all before completion of a scene investigation or autopsy, vital components of a proper infant death investigation. And, unfortunately, many causes of infant death are not apparent externally, but do become apparent at the time of autopsy. This group includes inflicted trauma.

That inflicted injury can masquerade as SIDS is well known. Before the identification of SIDS, the sudden death of an infant was often referred to as "crib" or "cot" death; however, the fact that these deaths may result from inflicted injury was not ignored. McRae et al. (1973:863) stated, "Obviously abuse must be considered as one cause of 'crib death'." Perrott and Nawojczyk (1988) reported eight cases referred to a medical examiner where the coroner had initially made a diagnosis of SIDS, but the diagnosis of SIDS was proven incorrect after autopsy and subsequent evaluation. The eight cases represented one each: overdose, overlay, drowning, suspected inflicted head trauma, battered baby syndrome, gastric perforation, marked dehydration, and pneumonia associated with malnutrition and possible neglect. The authors further described that, of 170 cases classified by the coroner as SIDS, only 101 were classified as SIDS after an autopsy. This study highlighted the importance of autopsy in the diagnosis of SIDS and the evaluation of inflicted trauma—at least two of the eight infants died from inflicted injuries, yet externally and by circumstances, must have appeared as natural deaths to warrant the diagnosis of SIDS by the coroner. Meadow (1999) reviewed 81 infants thought to have died of natural causes, including 42 of SIDS, who were later deemed by the courts to have been killed by their parents. Meadow (1999:10) said

The likelihood that the court verdicts about parental responsibility for death were correct is very high indeed. Even in family courts, where the judgement is made on the balance of the probabilities, all experts, particularly the judge, are aware of
the dire implications of the verdict in terms of a parent being allowed to care for a future child.

However, this statement implies the courts are infallible in their judgement, which they are not. In addition to Perrott and Nawojczyk (1988), other authors have demonstrated the subtle nature of inflicted trauma. Williamson and Perrot (1990) reviewed radiographs of 108 children clinically thought to have died from SIDS; however, multiple fractures were found in three infants, and these three were later determined to have died from child abuse. Therefore, the diagnosis of SIDS must be made carefully as deaths where the cause is actually inflicted trauma may appear as SIDS-like upon initial investigation and external examination (i.e., with characteristic investigative and external examination features of the SIDS diagnosis) but have the trauma identified at autopsy.

O’Halloran et al. (1998), apparently in response to the notion that a significant number of infants diagnosed as dying from SIDS may actually have died from negligence, abuse, or intentional suffocation, reviewed 157 SIDS deaths and 150 controls and found no indication that infants dying of SIDS were more likely than the control group to have been referred to child protective services. The conclusion is that, because SIDS deaths occurred in no greater number in families who were referred to child protective agencies, that the likelihood that these deaths were actually concealed homicides is unlikely. This conclusion assumes that parents who are referred to child protective services are more likely to kill their child than parents who are not referred to a child protective service.

While its incidence may be debated, that smothering of an infant can masquerade as SIDS because of the paucity of diagnostic findings at autopsy is well-described in the medical
literature (Reece, 1993; Byard and Krous, 1999; Meadow, 1999; Oehmichen et al., 2000; DiMaio and DiMaio, 2001; Dolinak and Matshes, 2005; Hymel, 2006). A disagreement on the number of infant smotherings, in combination with a lack of definitive autopsy findings in SIDS and a less-than-thorough approach to death investigation employed by some investigators, may have led Meadows (1999:13) to say that “it is sad that the term SIDS has become a barrier to the sensible and sensitive investigation of infant deaths.” However, the failure of some death investigators to correctly evaluate infant deaths does not nullify the utility of the diagnosis of SIDS when carefully and appropriately made. Nashelsky and Pinckard (2011) have proposed abandoning the use of the diagnosis of SIDS in the ICD-10 (International Statistical Classification of Diseases and Related Health Problems) in favor of the more general cause and manner of death of undetermined and undetermined. Unfortunately, their use of an undetermined manner seems to provide parents with even less information (e.g., the diagnosis of SIDS at least provides for a natural manner of death), and to open questions that may not be necessary, as an undetermined manner of death can imply that trauma is one possible cause. The authors address that the use of undetermined as the cause and manner does not necessarily imply that a death was “suspicious”, but that it is simply an honest conclusion to the death investigation. Whether or not SIDS is an appropriate autopsy diagnosis is not the topic of this dissertation; however, the importance of the potentially subtle nature of inflicted injury in infants is.

General discussion of child abuse

There are three forms of child abuse: neglect, physical abuse, and sexual abuse. In the United States, neglect accounted for 78.3% of cases of abuse in 2010 and physical abuse a much
Both neglect and physical abuse can result in the death of a child. While the overall rate of child abuse, especially in the 1 year and under age category, is high, at 20.6/1000 children in the United States, the rate of child fatalities from abuse is much lower, at 2.07 deaths per 100,000 children (US Department of Health and Human Services, 2011). In comparison, the overall mortality rate for children under the age of 1 year was 6.7/1000 births in 2006 (Centers for Disease Control and Prevention, 2009). So, fatal abuse accounts for a relatively small number of the infant deaths in the United States. In Montana in 2010, of 12,838 children less than 1 year of age, 1,383 sustained an episode of abuse, and of these, 167 sustained physical abuse. Infant deaths due to fatal abuse are relatively rare in Montana. According to the 2005-2006 Montana State FICMR (Fetal, Infant, Child Mortality Review) review (Montana State Department of Public Health & Human Services, 2009), there were 70 deaths of infants (i.e., children less than 1 year of age) in Montana in 2006, and from 2005-2006, four infant deaths were the result of a homicide.

As described above, child abuse is not a modern phenomenon. In his review, Lynch (1985) reported mention of intentionally struck children in Rhaze’s Practica Puerorum, written around 900 AD and a description by the Greek Soranus in 200 AD that angry women were like maniacs and may let a crying newborn drop from their hands. Lewis (1997) cites an author who reported that medieval physicians were against the common practice of vigorously rocking children to make them fall asleep, and that this practice could render the infant unconscious. Although these mentions of child abuse, or suggestion of child abuse, are in literature dating far back, a more organized approach to the documentation and understanding of child abuse did not occur until relatively recently. While Ambrois Tardieu in 1860 reportedly discussed 32 children
with features now recognized as abuse, Caffey’s 1946 paper is recognized as the first association between multiple fractures and subdural hemorrhages (Lynch 1985; Lonergan et al., 2003), findings that are now associated with child abuse, specifically, what is sometimes termed, “shaken baby syndrome”.

The coverage of child abuse in forensic pathology and forensic anthropology texts has varied through the years and corresponds to the development of the understanding of child abuse. Semple (1892) has an entire chapter on infanticide (referring to deaths of children immediately around the time of conception), but no chapter on child abuse. The author devotes 26 pages to sterility, rape, pregnancy, delivery, criminal abortion, infanticide, evidence of live birth, and causes of death of the fetus, but only 8 pages to blunt force injuries, sharp force injuries, and gunshot wounds. Moritz (1941) reported no specific information regarding infanticide or child abuse. Gonzales et al. (1954) devote one chapter to infanticide, but no specific chapter to child abuse. Therefore, in the past, an understanding of deaths around the time of birth (e.g., liveborn versus stillborn, and deaths due to abortion) was apparently, based upon the page counts devoted to the topic, of more concern that child abuse. In contrast, Moritz and Morris (1970) has a separate chapter on the battered child syndrome, and Dolinak et al. (2005) devote one chapter (45 pages) to child abuse, and only 1 page to the fetus and determination of live birth or stillbirth. Therefore, while child abuse has long been known to medical practitioners and others, its importance in the medical literature has definitely changed, most significantly after Caffey (1946) recognized and published the association between long bone fractures and subdural hemorrhage, and after Kempe et al. (1962) defined the term, “battered-child syndrome”.

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Caffey (1946) presented six children, each with a subdural hemorrhage and fractures of long bones. He summarized his findings, indicating that a history of injury to the head or extremities was lacking in each case and that there was no radiologic evidence of bone pathology that would predispose the infants to fracture. Later, Caffey (1972) introduced the concept that the subdural hemorrhage and long bone fractures, often metaphyseal fractures, were the result of whiplash shaking of the infant. Although Caffey’s work is generally listed as the starting point for our understanding of shaken baby syndrome (SBS), 120 years before Caffey, Cutter (1852:376) stated, “Concussion of the brain, and the results above mentioned, may be produced by the sudden motion attendant on the violent shaking of a scholar. Consequently, a child should never be seized by the arm and shaken violently as a method of chastisement.” Caffey (1972) did appear to support the idea that the injuries were not necessarily intentional, but could occur during times of stress or rough-handling of the infant.

Although Kempe (1962) introduced the term and concept of battered child syndrome, which was apparently the first diagnostic term used that implicated abusive injuries, other authors before him and after Caffey (1946) discussed child abuse. Altman and Smith (1960:413) indirectly linked abuse to skeletal injuries, in stating, “The early recognition of the condition [fracture] may be lifesaving, for removal of the child, if at all possible, from the environment in which these occur should result in complete cure.” Adelson (1961) described, in an article titled, "Slaughter of the Innocents", among other causes of inflicted death in infants, infants and children being squeezed by an irate father, causing multiple rib fractures and lacerations of internal organs. Building upon the work and ideas of these early pioneers, the understanding of child abuse has increased. Because of this increasing knowledge about and increasing
recognition of child abuse, the incidence of the diagnosis of child abuse has increased. For example, Lauer et al. (1974) in reporting 130 child abuse cases, saw 5-10 cases per year in 1965, and 16-36 cases per year in 1971. And, the diagnosis of child abuse based upon physical changes is not merely within the domain of pediatricians and forensic pathologists, Kerley (1976), “Forensic anthropology and crimes involving children” and Kerley (1978), “The identification of battered-infant skeletons”, early and solidly established the role of forensic anthropologists in the identification of child abuse.

In the recognition of child abuse, Gradwohl’s Legal Medicine (Camps, 1968:436-437) states

The story of each case is so uniform as to be almost a carbon copy of all. A child is taken to the doctor or the hospital and is found to be seriously injured or dead. In the latter event, there is usually a subdural haematoma with or without a fracture of the skull; otherwise there may be a fracture of a long bone or of the ribs and often multiple bruises on the lower/upper trunk and head. An explanation, which is always forthcoming, is that the child sustained the injuries as a result of some accident, such as falling from a table, from its cot, or even from the parent’s arms.

And, later, “If the doctor is alert he will arrange for an X-ray examination of the whole child. This may show other injuries such as fractured ribs or long bones, or epiphyseal separations, some of which may be of different ages and united.” While the authors may have described a classic case of fatal child abuse, and one that should be readily diagnosed by autopsy and investigation of circumstances, many infant deaths, suspicious for fatal abuse, are not so easily discovered. Camps (1968:444) also discusses smothering of infants, stating that “in order to prove death from smothering it is essential to show evidence of the act itself and this, as in the cases of alleged ‘overlying’ or ‘cot deaths’, is by no means easy to demonstrate if associated with
a soft surface.” In these two statements above, the authors essentially describe two ends of the spectrum of fatal child abuse, the first which is relatively easily diagnosed and the second, which is much more difficult, if not impossible in most cases, to diagnose. Of course, other cases of fatal child abuse can fall between the two ends of the spectrum.

**Shaken baby syndrome**

The first scenario described above by Camps (1968) essentially describes the condition sometimes referred to as shaken baby syndrome (SBS). This condition, having origins with Caffey (1946), with an association between subdural hemorrhage and long bone fractures, and receiving its first name by Caffey (1972), has evolved. Wetli et al. (1999) describe the features of whiplash shaken infant syndrome, or SBS, as intracranial hemorrhage (subdural and subarachnoid), retinal hemorrhages, absence of scalp injuries, and possible fingertip bruises on the back. The authors also discuss how a diagnosis of fatal child abuse can be readily made when a child has multiple injuries in various stages of healing, with fresh, recent, and old rib fractures being classic signs and can be combined with abrasions, burns, contusions and other repetitive injuries. Thus, from an initial syndrome characterized mainly by subdural hemorrhage and long bone fractures, the variety of injuries identified in cases of child abuse, including SBS, is now numerous.

Shaken baby syndrome (SBS), abusive head trauma (AHT), non-accidental injury (NAI), or inflicted head injury are terms variably used to describe an infant who has sustained head injuries at the hands of an adult. In infants, the SBS is identified by a subdural hemorrhage, retinal hemorrhages, and encephalopathy, with no other reasonable medical explanation for the
findings. However, whether or not shaking alone can cause the findings, or whether impact of
the head against a surface is required to produce the pathologic findings is debated (Case and
DiMaio, 2011). The issue of whether or not shaking alone can cause these findings in children is
not vital to this discussion; however, whenever some component of shaking or slamming of a
child as the mechanism of injury is involved, it would seem that an association with rib fractures
is distinctly possible, as compression of the chest could occur during the shaking.

In some publications, inflicted head injury is associated with rib fractures, and, in others,
there may be no mention. For example, Duhaime et al. (1987) reviewed 48 cases of SBS, but
made no mention of rib fractures. Similarly, Hadley et al. (1989) reviewed 36 infants with non-
accidental head trauma, and made no mention of rib fractures. However, other authors do
describe rib fractures. Gilliland and Folberg (1996) used rib fractures as one of three criteria
(requiring two of three of their criteria) to classify the mechanism of death as shaking in their
study regarding retinal hemorrhages. Lancon et al. (1998), Tzioumi and Oates (1998), Kivlin
(2001), Case et al. (2003), Oehmichen et al. (2005), and Bennett (2008) all support the
association of inflicted head trauma and rib fractures. Feldman et al. (2001) reviewed 39 infants
with a subdural hemorrhage due to abuse, and found that eight of the infants had rib fractures,
two with acute rib fractures, five with remote rib fractures, and one with both acute and remote
rib fractures. Carty and Pierce (2002) found rib fractures in 154 of 435 children with confirmed
non-accidental head trauma, and, in 31 children, only a single fracture each was present.
Matschke et al. (2009) reviewed 17 infants with non-accidental head injury, and found that three
infants had rib fractures. Adamsbaum et al. (2010) reviewed 112 infants diagnosed with abuse
head trauma and found that five had healing rib fractures. Sieswerda-Hoogendoorn et al. (2012)
discussed that the presence of rib fractures can make the diagnosis of abusive head trauma easier to identify. Thus, SBS, a common cause of death in an abused infant, which can easily have a component of chest compression, is associated with rib fractures.

While the features of SBS (i.e., subdural hemorrhage, retinal hemorrhages, and cerebral edema and encephalopathy) are readily diagnosed at autopsy, other injuries to infants and children can be subtle. Zumwalt and Hirsch (1980) described six homicides in children, where the cause of death was not necessarily available from autopsy alone, instead requiring investigation or additional testing to confirm. The deaths involved excessive salt intake, dehydration combined with bruising and sickle cell trait, smothering, neglected burns, heat stroke, and hypothermia. The infant, who was smothered, had two healing rib fractures. Cohle and Byard (1994) describe a 7-month-old boy with recent and healing fractures of the ribs who died from parentally-induced asphyxia. Therefore, inflicted trauma and other forms of non-accidental injury can be difficult to identify at the time of autopsy, and rib fractures can be a component of these subtle deaths.

Criteria for diagnosis of abuse

The identification of an abused infant or child is, unfortunately, not an easy task, and both overlooking abusive injuries in a child as well as identification of abusive injuries when there are in fact none have serious repercussions. In the first case, the child returns to a hostile home environment that may lead to their death, and in the second, innocent parents or caretakers may be prosecuted for a crime that they did not commit. Banaszkiewicz et al. (2002) describe that the diagnosis of child abuse is essentially a clinical diagnosis that is difficult to make and requires
much information. A comprehensive assessment of the case by a team composed of doctors, social workers, and police, among others, is advocated. In agreement, Morris et al. (2000:553) state, “The type of in-depth investigation required to unravel a complex case is not within the reach of individual doctors regardless of their expertise in child abuse. A multidisciplinary child protection team however brings together a wide range of resources, which can aid in the medical diagnosis of child maltreatment.” Kemp et al. (2008) also prefer a multiagency child protection group or legal panel reviewing all available information to make a determination of child abuse, or, if possible, perpetrator admission, or an independent witness to the abuse. Barsness et al. (2003) used a multifactorial determination of child abuse (using two pediatricians, two social workers, and one physician assistant). Therefore, although the recommended exact composition of the group may vary, many authors stress that the identification of child abuse requires cooperation and communication between multiple agencies.

Some authors have attempted to establish set criteria for the diagnosis of child abuse. These criteria can be heavily based upon historical information (Thomas, 1991) or radiologic findings (Radkowski et al., 1983). Thomas (1991) listed six categories for the causation of injuries in children: definite abuse, likely abuse, questionable abuse, questionable accident, likely accident, and definite accident, and the features of each category. The classification scheme was based upon the author’s review of injuries initially recognized by other clinicians and then submitted for consultation. Radkowski et al. (1983) listed 14 radiologic findings to be used for the diagnosis of child abuse.

According to Thomas (1991), signs of definite abuse include a positive skeletal survey (with multiple recent fractures or with fractures of various ages), an eyewitness to the episode,
multiple internal injuries, physical findings including bruises consistent with hands, electric cord, or teeth, or suspicious or unexplained burns or scars, history of a sibling abused at the same time or history of a definite intentional act causing physical harm to the child or a parental fight with injury not directed at the child, or a suspicious injury followed by definite abuse later. Signs of likely abuse include an initial suspicion by referring clinicians coupled with a history that is not sufficient for the injury, and/or the history regarding the incident where the injury occurred changes, and/or family members or caretakers present a different version of the history of the injury, and/or there was inappropriate delay in seeking care for the injury. Signs of questionable abuse were the same as those for likely abuse, except that the referring clinicians did not have a suspicion for abuse, but review by consultants indicated the possibility of abuse. These criteria defined by Thomas (1991) indicate the importance of multiple injuries (either skeletal or internal) as well as the importance of discrepancy between the history reported by caretakers as to how an injury occurred and evaluation of the injury itself (e.g., the injury identified by the treating physician could not have resulted, or most likely would not have resulted, from the scenario described by the caretakers). In addition to the importance of correlation of the injuries and the history given by caretakers as to how those injuries occurred, additional clues from the history as described by other authors that may indicate child abuse as the cause of identified injuries in children include delay in seeking treatment, poor child-parent interactions, and a malnourished child (Galleno and Oppenheim, 1982).

According to Thomas (1991), signs of a questionable accident included an isolated accident, where social workers or physicians had no suspicion of abuse, but the history provided was somewhat inconsistent although the injury was consistent with the history, or the history
provided was inconsistent with the extent of the injury, but the referring clinician or social worker had no suspicion of abuse, or the injury was the result of an isolated incident, where there was no suspicion of abuse, but the history regarding how the injury occurred was unknown. Signs of a likely accident included a consistent history, with an isolated injury and no suspicion of abuse, or a consistent history with no suspicion of abuse, but with neglect involved, or a consistent history (although with minimal information provided) with no suspicion of abuse and an isolated injury, or a history consistent with the injury, but with aggressive or irresponsible behavior involved, but no directed at the child. Signs of a definite accident included a substantiated motor vehicle accident, an accident witnessed by several uninvolved witnesses, or a pedestrian struck by a motor vehicle.

The criteria used by Thomas (1991) lean heavily on historical data and the evaluation of the injuries in the context of the scenario reported to have caused them, and also on correlation between an initial impression formed by the primary doctors involved as well as a secondary impression formed by the consultant. In contrast, Radkowski et al. (1983) listed criteria for enabling a radiologist to make a diagnosis of child abuse, based upon the presence of specific radiologic findings: 1) unsuspected lesions, 2) extent of trauma more severe than indicated by history provided by caretakers, 3) transverse diaphyseal fractures, 4) metaphyseal-epiphyseal injuries, 5) multiple fractures, 6) fractures of different ages, 7) rib fractures, 8) follow-up radiographs, 9) injuries to small long bones of the hands and feet, 10) compression fractures of vertebral bodies or fractures of spinous processes, 11) focal bone lesions with medullary fat necrosis, 12) head injuries associated with rib fractures and/or multiple long bone fractures, 13) visceral injuries, and 14) other imaging studies.
Obviously, the diagnosis of abuse in living children is not simple--physicians must carefully consider the nature of the injuries that they identify the infant or child to have, and compare them with the history provided by the parents or caretaker. And, the preferred method for establishing a diagnosis of child abuse would require the input of multiple people, spanning a variety of disciplines, each with an understanding of a general or a specific area with reference to the abuse of children. Nevertheless, clearly the radiologic evaluation of a child is of critical importance in the diagnosis of child abuse.

**Radiologic criteria for diagnosis of child abuse**

Although the most common injuries seen in battered children are those of the soft tissue, such as abrasions, contusions, or lacerations (O’Neill et al., 1973; McRae et al., 1973), fractures as a prominent component of child abuse are well-described in the medical literature and different fracture types have variable association with child abuse. Recurring features consistent with child abuse include metaphyseal fractures, multiple fractures, varying stages of healing and repair indicating fractures of varying ages, and rib fractures (Silverman, 1974; Silverman, 1987; Cameron and Rae, 1975; Galleno, 1982; Cohle and Byard, 1994; Knight, 1996).

Several authors have described the association of specific fracture types with child abuse. Kleinman (1987b) describes fractures with a high, moderate, or low specificity for abuse. Fractures with a high specificity for abuse include metaphyseal fractures, posterior rib fractures, scapular fractures, spinous process fractures, and sternal fractures. Fractures with a moderate degree of specificity include multiple fractures (especially bilateral), fractures of varying ages, epiphyseal separations, vertebral body fractures and subluxations, digital fractures and complex
skull fractures. Thus, in contrasting opinions, Thomas (1991) would classify children with multiple fractures or fractures of varying ages as definite abuse, whereas Kleinman (1987b) indicates these findings have a moderate specificity for abuse. Hobbs (1989) listed metaphyseal or epiphyseal fractures, rib fractures, wide complex skull fractures, scapular or sternal fractures, multiple fractures, fractures of different ages, and unpresented fractures (i.e., old fractures with no history of having been taken to the emergency room or other health care facility at the time of the incident causing the fracture) as having a higher specificity for abuse than other fractures, and described six patterns of fractures seen in child abuse: 1) single fracture with multiple bruises, 2) multiple fractures in various stages of healing, 3) metaphyseal-epiphyseal fractures, 4) rib fractures, 5) formation of new periosteal bone, and 6) skull fractures in association with intracranial injury. Hart et al. (2006) in their review list multiple fractures in various stages of healing, femur fractures in pre-ambulatory children, "chip" fractures of the metaphysis, skull or rib fractures, scapula fractures, and ulna midshaft fractures as suspicious for abuse. Jenny et al. (2006) in their review, highlight metaphyseal fractures and rib fractures. Thus, while differences exist between authors as to what skeletal injuries are most suspicious for abuse, certain fracture types are relatively commonly listed and among these are both metaphyseal fractures and rib fractures.

Regarding the specificity of radiologic findings for child abuse, Silverman (1974:51) stated that "in general it can be said that the skeletal manifestations of the battered child syndrome are so characteristic as scarcely to be confused with anything else. Nevertheless, from time to time there is reluctance to accept the specificity of these lesions." Carty (1997:1365) stated, "While the diagnosis [of child abuse] is most often based on clinical, social, and
radiological features taken together, the radiological abnormalities may on occasion be the strongest evidence of abuse,” agreeing with Silverman (1974), but not to the same degree of certainty. In support of Carty (1997) and Silverman (1974), Mandelstam et al. (2003:388), citing Kleinman (1998), stated that "it is well recognized that certain patterns of injury are sufficiently characteristic to permit a firm diagnosis of inflicted injury in the absence of clinical information." However, while the above authors appear to strongly favor the use of radiology and specific radiologic features in the diagnosis of child abuse, not all authors agree. For example, Kemp et al. (2008:1) state, "No fracture, on its own, can distinguish an abusive from a non-abusive cause.”

In addition to specific bones being fractured as indicative of abuse, other features about the fractures may help diagnose abuse. Specifically, multiple fractures and fractures showing a variable degree of healing are common components in the discussion of skeletal injuries associated with child abuse (Fatteh, 1973; Silverman, 1974). However, multiple fractures are not necessary for child abuse to have occurred. Loder and Bookout (1991:432) based upon a review of 493 children with fractures, 75% of which were sustained due to abuse, determined that "...an isolated acute fracture without signs of other trauma was the most common orthopedic occurrence in this series." And, as described above, a history that is incompatible with the injury is also another commonly cited indicator of the possibility of abuse (Kogutt et al., 1974).

In summary, in the evaluation of skeletal trauma to assess for the possibility of child abuse Kleinman (1987b:10) states

For those physicians and other professionals dealing with cases of child abuse, there is a proverbial question that inevitably arises. Is there a radiologic alteration that regardless of history in an otherwise normal patient can be viewed as ‘diagnostic’ of non-accidental injury? The classically described metaphyseal
lesions of the long bones satisfy this definition more closely than any other skeletal or visceral abnormality occurring in cases of child abuse.

The author agrees with numerous other authorities that the classic metaphyseal ‘corner’ fracture and ‘bucket-handle’ fracture lesions are virtually pathognomonic of infant abuse (Lonergan et al. 2003).

**Socio-economic factors associated with child abuse**

That social class plays a role in the development of disease was promoted by Dr. Rudolf Virchow, a German pathologist and anthropologist, most memorably with his report documenting the typhus epidemic in Upper Silesia in 1848 (Baer, 1982; Sy and Brown, 2006; Brown and Fee, 2006). In this regard, Virchow, along with Friedrich Engels with his study of the condition of the working class in Manchester, was reportedly one of the ancestors of the political economy of health theory (Baer, 1982; Singer, 1998).

The political economy of health theory is “an attempt to understand health-related issues within the context of the class and imperialist relations inherent in the capitalist world-system.” (Baer, 1982:1) and “…may be employed to understand and address such issues as: the social causes of preventable disease and injury…” (Minkler et al., 1994). The political economy of health is not a single theory, but incorporates elements of other theories, including “orthodox Marxist approaches, cultural critiques of medicine, and dependency theories” (Morgan, 1987; Minkler et al., 1994:131), and can be divided into two major areas: political economy of illness and political economy of health care (Baer, 1982), and is important to many disciplines, including, but not inclusive to, anthropology, political science, and epidemiology (Baer, 1982; Wilkerson, 2003; Szreter and Woolcock, 2004). Although the utility of the political economy of
health is much more broad and encompassing, its guidance in understanding the social origins of preventable disease and injury is of utility in the discussion of microscopic fractures of the rib heads in infants, as child abuse is a preventable injury. Numerous authors, to be discussed below, have linked various socio-economic factors (e.g. age of mother of infant) to risks of child abuse. If the finding of these microscopic rib head fractures can be linked to both suspicious circumstances of death and certain socio-economic factors, they could serve as a marker of subtle abuse, and serve to support the theory that social circumstances impact the health of an individual, in this case, the presence or absence of child abuse. In an applied context, although markers of subtle abuse may not necessarily allow a forensic anthropologist examining skeletal remains or a forensic pathologist performing an autopsy to outright determine child abuse, this information can be important to other investigators, and, as described above, the best investigation of child abuse is one performed in concert by a variety of individuals covering a range of specialties.

*General social factors associated with child abuse*

Schnitzer and Ewigman (2005) in their study of 149 fatally-injured children found that, compared to controls, the fatally-injured children were more likely to be black, reside in households with young siblings, and have been born to a young, unmarried, Medicaid-eligible mother with less than a high school education, who had late or no prenatal care or who had a history of a prior report to child protective services. Zhou et al. (2006) found that maternal smoking during pregnancy, families with three or more siblings, maternal age of less than 20 years, births to unmarried mothers, Medicaid beneficiaries, and inadequate prenatal care were the
best prenatal predictors of child abuse. Kotch et al. (1999) found that depression, complaints of psychosomatic symptoms, lack of high school graduation, alcohol use, public income support, caring for more than one dependent child, and history of maternal separation from her own mother before the age of 14 years were associated with a higher incidence of child abuse. Altemeier et al. (1982) studied 1400 low-income mothers, seeing abuse of their infant by 23 of the mothers. In comparing the mothers of abused children to the mothers of non-abused children, the mother of the abused child was more likely to have lived in foster care (p-value of .0001) and did not get along with her own mother (p-value of .001). Murphey and Braner (2000) identified that there was a higher incidence of abuse (compared to the rate among the entire population) among children who received no prenatal care, or prenatal care that began after the first trimester or to those whose mothers were less than 20 years of age, smoked during pregnancy, or had a less than high school education level.

Murry et al. (2000) offer a protocol for the identification of families with children under the age of 3 years at risk for abuse, which includes a screening list, with factors described in the literature as associated with child abuse. The authors’s checklist includes parental age of less than 18 years, parental education of less than 12 years, an unmarried mother, poor or delayed bonding, deficits in parenting skills, unstable socioeconomic status, insufficient support, parental history of substance abuse, parental history of abuse or neglect, parental history of depression or emotional illness during pregnancy, four or fewer prenatal visits prior to 34 weeks gestational age, no well-child check before two months, child’s height or weight less than 5% without medical reason, history of atypical accidents, poor hygiene for child, preterm or low-birth weight child, and child with chronic disease. However, Jaudes and Mackey-Bilaver (2008) found that, in
a study of 101,189 children, that chronic physical disease conditions only slightly increased the risk of abuse. In agreement with the criteria listed by Murry et al. (2000), Lauer et al. (1974), in a review of 130 child abuse cases, saw that parents of battered children were more commonly younger than control parents.

In conclusion, although there is not complete agreement among various authors as to social factors constituting a risk for future child abuse, recurring factors include unmarried mothers, teenage parents, parents with less than a high school education, and history of tobacco use during pregnancy and substance abuse, and little or late prenatal care. And, although at least some of the factors described by Schnitzer and Ewigman (2005) and others could be associated with lower income classes, it must not be forgotten that Kempe (1971) described that battering parents came from all classes (including upper and lower), and importantly that parents from the lower classes were much more likely to be reported by others to authorities, accused of abuse, and convicted by the courts. This information can help investigators understand that even rich and influential families can kill their child and such deaths must be properly investigated and, in comparison, that poor families are not necessarily any more likely to kill their children, and should not be subjected to unfair treatment in the investigation process or unwarranted suspicions merely because of their financial status. However, in apparent contrast to Kempe (1971), O'Neill (1971) found that all perpetrators were from poor families.

*Mental status of parents and child abuse*

Although Adelson (1961:1346), who studied 41 victims of fatal child abuse (with 17 perpetrators being fathers and 11 being mothers), said, “Frank psychosis in the assailant was the
single most common factor in precipitating the fatal incident”, Anderson and Lauderdale (1982) opined that their research confirmed previous reports that said that less than 10% of abused children had psychotic or aggressively psychopathic parents, but that instead the parents may be less able to cope with stress. Brown (1976) agreed with this premise, as do other authors. Kleinman (1990:704) said

> As most child abusers are persons poorly equipped to deal with stress, the occurrence of infant abuse is a product of a delicate balance between the severity of the stimulus of crying and the threshold for violent action by potential abusers. The effects of drugs, alcohol, and environment conditions may potentiate this interaction.

In support of these findings and statements, Salmon (1971) described how Kempe preferred non-accidental injury to wilful damage because child abuse can occur under stress of adverse social and personal conflicts.

> Stress can manifest in many ways, and many different circumstances can cause stress; however, financial stress is important when discussing child abuse. In support of Kleinman (1990), Cadzow and Armstrong (1999) studied 151 mothers whose infants had reached 7 months of age. The authors found that variables associated with financial stress were the most significantly associated with an elevated child abuse potential, with the child abuse potential determined through a questionnaire designed to serve as a screening tool to assess for the potential for child abuse. Financial stress included concerns about no housing and no food. Cadzow and Armstrong (1999) also found significant associations between an education level of less than 10 years and elevated child abuse potential, and between various aspects of domestic abuse. The authors did not find any significant association with single parent, ambivalence
regarding the pregnancy, young maternal age, history of psychiatric illness, or drug or alcohol use and an elevated child abuse potential.

**Perpetrators of abuse**

An understanding of which caretakers are most likely to be instigators of child abuse can be helpful in the investigation of a fatality. Of 400 battering parents, Kempe (1971:29) said, “All social classes, all races, creeds, religions, and levels of education and income are proportionally represented,” but included that stepchildren and premature infants were more at risk. Salmon (1971) agreed that stepparents are common instigators of child abuse. Daly and Wilson (1985), based upon the results of their study of Canadian children using a phone survey and review of 99 abuse cases, opined that preschoolers in a household composed of a natural parent and a stepparent are 40 times more likely to sustain abuse than a child living in a house with two natural parents.

Numerous authors have described the actual incidence of various caretakers involved in abuse. Loder and Bookout (1991) reviewed 75 infants who sustained fractures as the result of child abuse, and found that 72 infants sustained injuries in their own home or a relative's home, two sustained injuries in a foster home, and one sustained injuries at daycare. Lazoritz et al. (1997) found in a review of 71 infants and Caffey's case reports that fathers were 33.3% of the perpetrators and mothers 6.7%. Price et al. (2000) listed 33 cases of child abuse, 13 due to boyfriends, eight due to the father, four due to the mother, one due to a babysitter, two due to step-parents, and in five cases, the abuser was unknown. Starling et al. (2007) reviewed 194 abused children, and found that biological fathers were 45.1% of the 155 known perpetrators,
3.3% were due to stepfathers, and a specific category for foster parents was not listed. O’Neill (1973) found, of 110 cases of child abuse diagnosed based upon the finding of an injury with investigation revealing circumstances that did not fit the injury, that in 55 cases the mother was the perpetrator, in 24 cases the father, in five cases siblings, and in 14 cases babysitters. Brown (1976) also said that mothers were the worst offenders. Starling et al. (2007) specifically addressed the types of perpetrators involved in inflicted skeletal trauma in their study of 194 children with fractures. Biological fathers and mother’s boyfriends accounted for 58.2% of fractures (with 45.1% being biological fathers). Of interest, the median age of children with skeletal injuries inflicted by a male was less than the median age of children with skeletal injuries inflicted by a female (4.5 versus 10 months). Related to Daly and Wilson (1985), Schnitzer and Ewigman (2005), in a study of 149 fatally-injured children less than 5 years of age, found that children who lived in a household with an unrelated adult(s) (e.g., mother’s boyfriend) were 47.6 (95% CI of 10.4-218) times more likely to die of inflicted injuries than those living with their two biological parents. McRae et al. (1973) studied 132 battered children, 16 were premature, 68 perpetrators were the mother or father, 25 were a common-law parent, 12 were a foster parent, 20 were single parents, two were adoptive parents, and three were relatives.

As can be seen, the frequency of different caretakers (e.g., mother versus father versus boyfriend) involved in child abuse varies between the studies. To make an accurate comparison of the different caretakers involvement in abuse, the authors would have had to compare the frequency of the different caretakers in their study to the frequency of the different caretakers in the population from which their study sample was drawn. For example, McRae (1973) found that while 68 perpetrators were the mother or the father, 12 were a foster parent; however, the
authors did not indicate the frequency of foster parents in their population from which their sample was drawn.

Regarding the role of foster parents in fatal child abuse, about 500,000 children live in some form of foster care in the United States, and the US child population in 2010 was 68,986,423 (American Academy of Child & Adolescent Psychiatry, 2005; US Department of Health and Human Services, 2011), meaning that approximately 0.7% of children live in a foster care environment. In 2010, 1262 children in the United States died as the result of abuse. Of the caretakers causing the death of the child, 12 were day care providers, three were non-relative foster parents, and one was a foster parent of unknown relationship. Therefore, foster parents were responsible for about 0.3% of deaths, which would seem to indicate that children living with foster parents were less likely to sustain fatal abuse than those in other relationships. In agreement, Schnitzer and Ewigman (2005), in their study of 149 fatally-injured children, identified six children fatally injured while living in a household with foster parents, and six children living with foster parents in the control group. Compared to the reference sample of fatally-injured children living in a household of two biological parents, the odds ratio for fatally-injured children living in a household with stepparents or foster parents was 1.1 (with a 95% CI of 0.3-4.7), with the results for step-parents contrasting with Daly and Wilson (1985). In contrast, Zuravin et al. (1993) described that of 296 supervised foster homes, 62 had at least one confirmed report of maltreatment during their five-year study period; however, 51% received public financial aid, 38% were single parents, and 58% of mothers and 70% of fathers has less than a high school education. Hobbs et al. (1999) described that foster children were 7-8 times more likely to be assessed by a pediatrician for abuse than a child in the general population;
however, Ainsworth (2000) stated the study had inadequate methodology and incorrect presentation of the statistical data.

Normal morphology of bone

A brief review of the anatomy of the rib head and neck is required to appreciate the changes to be described later. Three general terms used to describe the structure of a long bone are epiphysis, metaphysis, and diaphysis. Stedman’s medical dictionary (Hensyl and Flescher, 1990:525, 954, 430) defines epiphysis as “a part of a long bone developed from a center of ossification distinct from that of the shaft and separated at first from the latter by a layer of cartilage,” metaphysis as “growth zone between the epiphysis and diaphysis during development of a bone,” and diaphysis as “the shaft of a long bone, as distinguished from the epiphysis, or extremities, and apophyses, or outgrowths.” The term, physis, is sometimes used to describe the cartilaginous portion of the epiphysis (Hensyl and Flescher, 1990).

Long bones are formed through the ossification of a cartilage model. Ossification occurs at a primary center as well as two or more secondary centers. The primary center of ossification forms the diaphysis, while the secondary centers of ossification form the epiphysis and other structures of the bone. Although the rib is not classified as a long bone, it has a primary center of ossification and multiple secondary centers of ossification, including for the tubercle and the rib head. Scheuer and Black (2007) summarize that the epiphyses for the rib heads are the last to form, with complete fusion occurring around 17 years of age. Therefore, in the infant and young child population (less than 3 years of age), the rib head epiphyses should not be present.

As predicted, in the sections of infant and young child rib head and neck examined, the cartilage at the vertebral end does not harbor a separate epiphysis. The vertebral end and the
sternal end of the ribs examined have a growth plate though. So, although the actual rib head in
the ribs examined is not yet formed, or even in the first stages of formation as a secondary
ossification center, the region studied is where the rib head ultimately will form, and it represents
the current junction between the rib and the vertebral body. As there is a growth plate present,
the cartilage in the sections examined may be viewed as the physis and the section of forming
bone immediately distal to it is the metaphysis. Therefore, the microscopic structure of the area
of the ribs examined in this study will be considered equivalent to the rib head and neck,
including a zone of cartilage (physis) and immature and mature bone (metaphysis), with the
growth plate the junction between these two areas.

An additional histologic feature that must be mentioned is the perichondrial ossification
groove of Ranvier that completely encircles the growth plate (Shapiro et al., 1977). Oestrich and
Ahmad (1992) proposed the term periphysis for this structure, as it encircles the physis and the
most adjacent portion of the metaphysis, with the zone of Ranvier adjacent to the physis (the
cartilaginous portion of the epiphysis), and the ring of LaCroix adjacent to the metaphysis. Both
the zone of Ranvier and the ring of LaCroix are histologically a single structure, and both
produce bone bark (i.e., membranous bone; Figs. 2 and 3). However, terminology for these
histologic features is not consistent. Calmar and Vinci (2002) use the terms groove of Ranvier
and perichondral ring of LaCroix. Gurley and Roth (1996) use the term ring of Ranvier for the
collar of membrane bone located at this site. The bone bark produced in this area can be divided
into a metaphyseal collar adjacent to the metaphysis, and a spur adjacent to the physis (Oestreich
and Ahmad, 1993). Knowledge of this structure and the appearance of the bone bark is
important so that this normal anatomic structure is not confused with healing traumatic lesions.
No reference to clefts at the anterior edge of the growth plate, as identified by the author in his autopsy specimens, was found in the medical literature.

Figure 2 and 3. Bone bark. In both Figure 2 (left) and Figure 3 (right), the arrow indicates the bone bark, providing examples of this normal anatomic finding in two different ribs. Hematoxylin and eosin, 100x.

The layers of cartilage prior to the metaphysis are zones of resting (or reserve), proliferation, hypertrophy, and calcified cartilage (Figs. 4 and 5). However, the exact definition of zones and number of zones described varies between authors, and the descriptions include overlap between the physis and metaphysis. Burkitt et al. (1993) describes a zone of maturation between the zone of proliferation and the zone of hypertrophy, partially combines the zone of hypertrophy and zone of calcification, and describes a zone of cartilage degeneration (with capillary invasion), and the osteogenic zone (where osteoblasts congregate on the surface of spicules and commence bone formation), and this osteogenic zone is described as the metaphysis. Rosenberg (1994) describes that the physis is composed of a reserve zone, a zone of
proliferation, a zone of hypertrophy, a zone of mineralization, and the primary spongiosa, which has spicules with a cartilage center, bone on the outside, and is associated with mesenchyme and vessels between. Gurley and Roth (1996) define the physis (or epiphyseal growth plate) with the same zones as Rosenberg (1994), except the primary spongiosa is defined as the zone of provisional ossification. Gurley and Roth (1996) discuss the inability to distinguish the zone of hypertrophy and the zone of mineralization in decalcified sections and that the metaphysis is separated from the epiphysis by the epiphyseal growth plate.

Regardless of the delineation of specific zones, the metaphysis is best described as including the primary and secondary spongiosa (see Figs. 4 and 5), and is the junction between the cartilage and the mature bone. The primary spongiosa is where new bone is being deposited around central calcified cartilage cores, producing spicules (Kleinman, 1987b; Marks, 1998). Describing this process in more detail, osteoblasts produce osteoid and deposit it around the cartilage framework and this osteoid mineralizes into the trabeculae of woven bone (i.e., endochondral modeling). Osteoclasts then absorb the primary spongiosa, and with the aid of
osteoblasts, the woven bone is replaced with lamellar bone, which is the secondary spongiosa (Laor and Jaramillo, 1993; Xian and Foster, 2006). Importantly, disorders of the mineralization can lead to retention of cartilage in the metaphysis, which can occur in rickets, hypophosphatasia, vascular injuries, and trauma (Laor and Jaramillo, 1993). Kleinman et al. (1991) have described extensions of the growth plate cartilage into the metaphysis in association with child abuse. As the primary spongiosa represents immature bone, the idea that it may be more prone to fracture is not invalid.

**Review of fracture mechanisms**

A basic knowledge of the process of fracture development is useful for understanding the information contained in this dissertation. Carter (1985), Turner and Burr (1993), and Lucas et al. (1999) provide good reviews of important concepts in bone strength and fracture development.

Bone is a composite material composed of both organic materials (collagen and other proteins) and inorganic materials (hydroxyapatite crystals composed of calcium and phosphorus). As a composite material, bone contains both brittle material (the minerals) and ductile material (the collagen). The specific organization of these substances gives bone its material properties that then determine fracture characteristics. However, the mechanical properties of bone (i.e., how bone responds when force is applied to it) cannot be explained individually by either component alone (Burstein et al., 1975), and, thus, it is the interaction of the organic and inorganic substances that determine the mechanical properties of bone. The organization of these inorganic and organic substances and the architecture of bone, gross, microscopic, and
ultrastructural, is neither uniform nor simple, and this organization affects the mechanical properties. Martin (1991), in a review article, listed some of these organizational and architectural categories affecting bone structure: porosity, mineralization, density, architecture (compact or trabecular), and organization of collagen fibers, each of which can vary depending upon the type of bone being studied (e.g., compact versus trabecular, adult versus child) and each of which can potentially affect the mechanical properties of bone.

Numerous studies have been performed with bone to assess its mechanical properties when stressed and thereby, how fractures are produced; however, most of these studies involve the use of small segments of machined bone (a few centimeters in size) and not the entire bone (Laird and Kingsbury, 1973; Reilly et al., 1974; Saha and Hayes, 1976; Norman et al., 1995; Zioupos and Currey, 1998; Bayraktar et al., 2004). The reason for this use of bone segments instead of whole bones when testing is that values for the mechanical properties of bone derived from the study of whole bones are only useful for comparison and not for determining mechanical properties of the bone tissue itself (Reilly and Burstein, 1974). The use of machined segments of bone allows for much greater control of an experiment and serves to eliminate or minimize the effects of other structural variables associated with a whole bone that may affect the test results. In other words, throughout the whole bone, the composition and architecture varies; whereas in small machined segments, the composition and architecture is more uniform. Alms (1961) said that quantitative examination of whole bone strength is not possible because of this variation in composition and architecture. For example, the relative amounts of cortical bone (a high density form of bone) and trabecular bone (a low density form of bone) vary by region in long bones, and each can have different mechanical properties (Porta, 2005).
Therefore, while much information has been obtained regarding the mechanical properties of select segments of bone, application of these results to fracture patterns in whole bone is more difficult because of an inability to control for numerous variables that each can affect the bone being studied; however, a review of the information known about the mechanical properties of bone is still warranted.

To understand the mechanics of bone fracture, the definitions of several terms is necessary: force, load, stress, strain, elastic deformation, Young's modulus, stiffness, yield point, plastic deformation, toughness, and strength. Load is the amount of force applied to a bone. To provide for a more uniform measurement (i.e., across all sizes of bones), load is related to the area. Stress is load (or force) applied per unit of area and strain is the ratio of change in length to original length (Reilly and Burstein, 1974; Rogers, 1982; Porta, 2005). Load can be plotted versus degree of deformation to produce the load-deformation curve, or stress can be plotted versus strain to produce a stress-strain curve. Examination of the stress-strain curve can reveal other mechanical properties of the bone (Fig. 6).

**Stress-strain curve**

When stress is applied to bone, strain results. Initially, the stress (y-axis) and strain (x-axis) increase proportionally. In other words, as force is applied, the bone deforms in proportion to the amount of force; however, the deformation is not permanent and will reverse if the force is removed (Crowe and Swischuk, 1977; Rogers, 1982; Porta, 2005). This segment of the stress-strain curve is termed the elastic phase, the strain produced in the bone is termed elastic deformation, and the slope of the line is the elastic modulus, or Young’s modulus (Porta, 2005).
The elastic modulus is a measure of the stiffness of the bone (Porta, 2005). Turner and Burr (1993) describe bone as having both an extrinsic stiffness and an intrinsic stiffness. In the load-deformation curve, as with the stress-strain curve, increasing load initially causes reversible deformation of the bone (elastic deformation) and then irreversible deformation of the bone (plastic deformation). The slope of the elastic deformation portion of the load-deformation curve represents the extrinsic stiffness of the bone, and the size of the bone is a factor in the assessment of this mechanical property (Turner and Burr, 1993). The elastic modulus is a measure of the intrinsic stiffness of the bone, with the more stress required to deform the bone, the greater the stiffness of the bone (Reilly and Burstein, 1974; Rogers, 1982; Turner and Burr, 1993). Another
elastic constant, like the elastic modulus, is the Poisson’s ratio, which is a measure of the bone’s ability to conserve volume when loaded in one direction (e.g., expansion of the sides of bone when the bone is under compression and contraction of the sides of bone when the bone is under tension) (Reilly and Burstein, 1974).

At a certain amount of stress, the bone reaches the yield point. At this point, the bone enters the plastic phase. The yield point is also referred to as the yield strength, or structural strength (Daegling et al., 2008). In the plastic phase, the increase in strain is no longer proportional to the increase in stress. Instead, for the increase in stress, there is a disproportionately greater increase in strain (Crowe and Swischuk, 1977; Pierce et al., 2004; Porta, 2005). The strain produced in the bone is termed plastic deformation. Plastic deformation, unlike elastic deformation, will not reverse when the stress is removed. Plastic deformation of the bone will occur until the level of stress reaches the ultimate strength of the bone and fracture occurs (Porta, 2005; Daegling et al., 2008). The amount of strain that can occur in a material after the yield point and before fracture occurs is a measure of the ductility of the material (Turner and Burr, 1993).

The area under the stress-strain curve is the amount of energy required to cause fracture of the bone and is referred to as the toughness (Rogers, 1982; Turner and Burr, 1993). Toughness occurs in three stages, diffuse damage (energy absorbed prior to development of a major crack), initiation of the fracture crack, and progression of the fracture through the bone (Zioupos and Currey, 1998). Saha and Hayes (1976) described the total energy required to fracture bone as divided into the elastic deformation stage, the plastic deformation stage, and energy absorbed during fracturing.
As described above, bone has both brittle and ductile properties. On the stress-strain curve, a pure brittle substance such as glass has a line that parallels the y-axis. For a brittle substance, minimal change in strain is produced with increasing stress, until failure is reached; whereas, a pure ductile substance such as rubber has a line that parallels the x-axis. For a ductile substance, a small amount of stress produces a large degree of strain prior to fracture (Porta, 2005). Thus, brittle material breaks inside or just beyond the zone of elastic deformation, while ductile material can undergo much plastic deformation before fracture occurs (Reilly and Burstein, 1974; Rogers 1982; Turner and Burr 1993). However, with viscoelastic material such as bone, the rate of application of the stress also affects the stress-strain curve shape, and thus, the mechanical properties of the bone (Porta, 2005). A rapidly-loaded bone has increased stiffness and greater ultimate strength, and thus failure of the bone will more represent failure of a brittle substance; whereas a slowly loaded bone has decreased stiffness and lower ultimate strength and thus, failure will more represent failure of a ductile substance (Reilly and Burstein, 1974). This difference in response to rapid loading and slow loading is in part reflective of the fracture at the microscopic level. At lower strain rates, the fracture moves longitudinal to fibers and then breaks across the fiber, forming a series of steps in the bone. At higher strain rates, the fracture moves indiscriminately across all components of the bone (Pope and Outwater, 1972). This viscoelastic nature of the bone can affect the fracture morphology with slow loading more likely to produce linear fractures and rapid loading more likely to produce comminuted fractures (Smith and Peters, 1996).

As stated, bone is a composite material, and each material (organic and inorganic) contributes differently to the overall biomechanical behavior of the bone. Conducting
experiments on wet unembalmed bone tissue decalcified to various degrees with hydrochloric acid, Burstein et al. (1975) found that as the mineral content of the bone decreased, the elastic modulus decreased, the amount of stress required to enter the zone of plastic deformation decreased, and the ultimate strength (or point of fracture) decreased. Therefore, bone obtains most of its elastic stiffness from its mineral content, and the mineral content of bone is the source of its tensile strength (Burstein et al., 1975). The slope of the stress/strain curve after the yield point in non-decalcified bone, and incompletely calcified bone, was the same as the slope of the stress/strain curve for completely decalcified bone (Burstein et al., 1975). This finding would indicate that the collagen content of the bone is responsible for its behavior in the zone of plasticity. Burstein et al. (1975) appears to be the first work modeling the zones of elasticity and plasticity in bone.

Testing of whole bones

The application of information obtained via study of segments of bone to whole bones is difficult. Factors that can be controlled for in the study of a segment of bone cannot be controlled for in a study of a whole bone. In addition to that described above, Saha and Hayes (1976) discussed the difficulty in calculating stress and strain for whole bone because of changes in cross-sectional area through the bone and different proportions of compact and trabecular bone, among other factors. Simkin and Robin (1973:37) summarized the difficulty in correlating experimental work using controlled specimens (e.g., machined segments of compact bone from the tibia), with observations in living individuals as “the cortex may show different types of histologic structure with large morphological variations in organization, direction of fibers, and
amount of calcification. Any of these variations might affect mechanical behavior under stress.”

So, while much work has been done attempting to understand the mechanical properties of bone, and how force may produce fractures in those bones, the interaction of multiple variables would appear to complicate interpretation outside of the confines of a controlled laboratory experiment.

Although much of the experimental work with bone has been conducted on small segments of bone, several authors have carried out tests with whole and intact human bones. Messerer tested whole bones with hydraulic equipment and under various loads, such as tension, compression, bending and torsion, and identified the characteristic fracture patterns associated with each of these load types (Roesler, 1987). Rabl et al. (1996) illustrated the variable nature of fracture experiments conducted on whole bone through testing of artificially-induced fractures of human tibia. Depending upon the direction of loading (all loading was in the transverse plane but was varied in approach from the ventral, dorsal, medial, or lateral aspect of the bone), the authors saw different frequencies of direct and indirect fractures. So, the direction in which force is applied is important. Although they used small segments of bone as compared to whole bones, Reilly and Burstein (1975), using cortical bone from humans and bovines, also found lower ultimate stress and strain when force was applied in a transverse manner than when applied in a longitudinal manner. Schmidt (1979) conducted tests using cadavers protected by seat belts and evaluated the fracture patterns of the rib and sternum. Also, apparently, much work regarding experimental fractures induced in whole bone has been published by the Society for Automotive Engineers (Porta, 2005).
Different types of stress cause fracture of the bone: tension, compression, and spiral and different fracture patterns are seen in the bone due to the different types of stress based upon how bone responds to that specific type of stress. Alms (1961) and Rogers (1982) described that bone fails first in tension, then compression, and did not distinguish between adult bone and child bone. Worthwhile of mention, Rogers (1982) contained a chapter entitled, “Special consideration in children”. However, Pierce et al. (2004) describe that child bone is weaker in compression and that adult bone is weaker in tension and cite Ogden (2000) and Hall (1999); but, in review of those two references, the validity of the Pierce et al. (2004) statement is uncertain. Ogden (2000:48) states, “Adult bone usually fails initially in tension, whereas a child’s bone may fail in either tension, compression, or both.” Unfortunately, the author provides no study or citation to support this statement. Hall (1999:91) describes that

These minerals [calcium carbonate and calcium phosphate] give bone its stiffness and are the primary determiners of its compressive strength…Collagen is a protein that provides bone with flexibility and contributes to its tensile strength. There is progressive loss of collagen and increase bone brittleness with aging. Thus, the bones of children are more pliable than the bones of adults.

The final statement by Ogden (2000) would appear to be based upon the fact that child bone is less mineralized than adult bone, and therefore weaker in compression, and has more collagen than adult bone, and is therefore, more pliable and stronger in tension. In confirmation, in his review, Roesler (1987) stated that resistance to tension was due to collagen fibers and that resistance to compression was due to hydroxyapatite crystals. Currey and Butler (1975) describe that younger bone has less bending strength and is less stiff, but can absorb more energy before
fracture, and thus has greater ability [than adult bone] to undergo plastic deformation. Calmar and Vinci (2002) in their review describe that pediatric bone has a lower bending strength and lower mineral content. In addition to mineral content, other differences between adult and pediatric bone may play a role in fracture development. Nimityonskul et al. (1991) described that the bones of children are more porous and flexible than adults due to the presence of larger haversian canals, although Barer (1967), using ribs aged 11 to 88 years, described that the haversian canal size does not change. In conclusion, if resistance to tension is due to collagen, and if resistance to compression is due to the inorganic matrix, the possibility that adult bones are less resistant to tension and more resistant to compression, while the reverse may be seen in children, is plausible.

The variation in composition of adult and infant/child bone, and its effects on bone properties, is reflected in the presence of greenstick and bowing (or, bending) fractures, conditions that occur almost exclusively in children (Alpar et al., 1981; Mabrey and Fitch, 1989; Casey and Moed, 1996). Stedman's Medical Dictionary (Hensyl and Flescher, 1990:616-617) defines a greenstick fracture as "the bending of a bone with incomplete fracture involving the convex side of the curve only," and a bending fracture as "an injury in which a long bone or bones, usually the radius and ulna, are bent due to multiple microfractures, none of which can be seen by x-ray imaging." The presence of these certain fractures types commonly in children but uncommonly in adults implies that children’s bones respond in a different manner to stress than do adult bones; however, the possibility that children sustain fractures in circumstances different from adults (e.g., falling in a different manner) cannot be entirely excluded as a possibility; however, it would seem must less likely.
Bowing (i.e., bending) fractures of the forearm occur in children due to falls onto outstretched arms (Crowe and Swishchuk, 1977) causing longitudinal force to be applied at each end of a tubular bone, and result from enough force being applied to the bone to cause entrance into the plastic deformation region, but not enough to cause fracture (Borden, 1974). The greenstick fracture is another fracture type commonly associated with children that Rogers (1982:67) described as

the result of bending or angulation forces that place the convex side of the bone in tension and the opposite cortex or concave side in compression. These forces result in a fracture that is analogous to the break resulting from bending a green stick or twig. An incomplete transverse fracture is produced in the convex cortex by tension forces and usually extends to the middle of the shaft, involving about one-half of the circumference of the bone.

Therefore, the greenstick fracture results from failure first at tension and not compression. If the greenstick fracture is more common in children than adults, and if the mechanism of the greenstick fracture is failure first at the point of tension (rather than failure first at the point of compression), it would follow that children's bones are also susceptible to tension before compression in at least some circumstances, and therefore, similar in some circumstances to adults.

So, the issue of whether or not children’s bones fail first in compression or in tension as compared to adults is not simple. Also, the failure of adult bone in tension first is not an absolute. Love and Symes (2004) described failure of ribs due to compression before tension. Rohl et al. (1991:1148) also contradicted the idea that adult bone is stronger in compression than tension. Using tibial cancellous (i.e., trabecular) bone from donors aged 42-76 years, they found higher values for tensile strength than compressive strength, and concluded that “in testing to
failure, the values of strength, ultimate strain, and work to failure were significantly higher in
tensile than in compressive testing, indicating a different failure mechanism.” In contrast,
Keaveny et al. (1994) used bovine tibial trabecular (i.e., cancellous) bone and found that ultimate
strength and yield strength were greater in compression than tension. However, they noted that
the tensile and compressive strength was dependent upon the orientation of vertical, transverse,
and oblique struts, and that, based upon this orientation, tensile strength could be lower than,
equal to, or even greater than compressive strength. In agreement, Ebacher et al. (2007) studied
19 un-embalmed human tibiae and femora and determined that the bones failed in tension first,
followed by compression. And, Bayraktar et al. (2004), using 12 human trabecular bone
specimens from cadavers aged 51-85 years also found that yield strain was consistently higher in
compression than tension. The above three authors used trabecular bone, and, given that the
structure of trabecular bone (more lattice like) is different than compact (i.e., lamellar) bone, the
possibility that the two different structures respond differently to tension and compression is not
unreasonable.

The number of studies specifically addressing the properties of child bone tissue is small,
being about four (Currey and Butler, 1975; Hymel and Spivack, 2001; Ohman et al., 2011).
Ohman et al. (2011) determined that the ultimate compressive stress of children (aged 4-15
years, with a small fragment of the diaphysis of the femur or tibia as the test specimen) was 33%
less than adults. The ultimate compressive stress varies with the strain rate (Reilly and Burstein,
1975), and Ohman et al. (2011) used a strain rate of 0.1 s\(^{-1}\). Reilly and Burstein (1975)
determined the ultimate compressive and tension stress of adults, aged 21-63, using a strain rate
of 0.2-0.5 s\(^{-1}\). With both forms of force (i.e., tension and compression), the ultimate stress was
higher in longitudinal than transverse application of the force. The ultimate tension stress for longitudinally delivered force varied between 125-137 x10⁶ N/m², and for transversely delivered force was 40-62 x10⁶ N/m². However, the ultimate compressive stress for longitudinal varied between 198-211 x10⁶ N/m², and for transverse was 118-151 x10⁶ N/m².

If Ohman et al. (2011) results are applied to Reilly and Burstein (1975) results, the ultimate compressive stress for children's bone (130x10⁶ N/m²), when that force is applied in a longitudinal manner, would be similar to the ultimate tension stress of adult's bone (125x10⁶ N/m²), when that force is applied in a longitudinal manner. This comparison would indicate that the same amount of force that could cause adults bone's to fail in tension, but not compression, could cause children's bones to fail in compression; however, Ohman et al. (2011) only studied the effects of compressive force and not tension, and it is unknown how children's bones would respond to tension. Also, the strain rate used was different between the two studies.

The type of force applied may also be important in the morphology of the fracture. Moen and Pelker (1984), testing bovine femur and tibia from skeletally immature animals, found that failure in compression was through the zone of calcification and the distal metaphysis, but through the upper level of columnation in tension and between the upper columnar zone and the lower hypertrophic zone in shear, and with a variable pattern in torsion. In the metaphysis, the outer portion is compact (i.e., lamellar) bone, and the inner portion is cancellous (i.e., spongy, or trabecular) bone (Pope and Outwater, 1972).

While the above discussion concerns bones in general, there also exist specific considerations for the ribs when discussing mechanical properties and the response to an applied force. For example, Barer (1967) found a decrease in the thickness of the cortex of the ribs with
age, using 74 rib specimens from individuals aged 11 to 88 years. Such a decrease in thickness of the cortex could affect a response to tension and compression. Another consideration is that loading of the chest does not necessarily apply an equal amount of force to all skeletal elements. Roberts and Chen (1970) found that with application of force to the sternum, the maximum tensile force varied in location between different ribs (e.g., between the angle and tubercle in ribs 1-2, 4, 6-7, and 10, and between the tubercle and neck in ribs 3, and 8-9) and in intensity between different ribs (e.g., 2290 psi for rib 8 and 8700 psi for rib 1). Tsai et al. (2012) developed a finite element model to study the mechanism of rib fractures in infants, and determined that any predictions would depend on 1) the elastic modulus of the bone, in reference to the age of the child, 2) asymmetry in infant’s ribs, and 3) the amount of force applied by the individual who is squeezing the child’s chest. The authors reported that, at this time, little data is available on those three important features.

Battered child syndrome and child abuse

Along with Caffey 1946 and Caffey 1972, Kempe 1962 is considered, as of the year 2000, to be one of the top 25 classic papers of child abuse (Donnelly and Oates, 2000). Kempe (1962) introduced the first diagnostic term used to categorize injuries in children as abuse, the name, "battered child syndrome." As described by Kempe (1962), the condition "battered child syndrome" may occur at any age, but in general is found in children <3 years of age, and the clinical manifestations can occur as a result of a single episode of trauma, but that more often the child shows signs of neglect and multiple injuries. Not all authors adhere to this description that battered child syndrome can include children who are the victim of a single episode of abuse. In
fact, Kempe (1971) himself later described child abuse as a spectrum of non-accidental injury and deprivation, and that battered child syndrome was at one end of the spectrum. Zumwalt and Hirsch (1987) indicate that, in a pathologic sense, battered child syndrome only includes those children who have sustained repeated episodes of abuse. Loquvam (1977) and DiMaio and DiMaio (2001) also excluded deaths due to a single episode of trauma from the definition of battered child syndrome. Thus, although the term "battered child syndrome" may have been the first specific syndrome attributable to abusive trauma in children, the syndrome is currently only used to describe a small spectrum of child abuse, indicating a condition in which repetitive injuries are inflicted on a child, and, at clinical or autopsy diagnosis or anthropologic evaluation of the skeletal remains, evidence of multiple episodes of inflicted injury are evident. Of course, both single episodes of abuse as well as repetitive trauma can occur in the context of the abuse of children. Weston (1974) described 36 children killed by an adult, only 13 of whom had no evidence of previous injury and died from a single episode of injury. Of these, three had minimal evidence of injury to the external surface of the body, and one had no evidence of injury to the external surface of the body. Of the 23 infants with evidence of repetitive trauma, three had acute fractures of the ribs.

Although child abuse can present with a variety of injuries, skeletal trauma is a common component of those injuries. O'Neill et al. (1973) reviewed 110 abused children, aged 3 weeks to 11 years (with the majority between 6 months and 1 year of age), and found fractures in 35 patients (multiple fractures in 29 patients and a single fracture in six patients), with 20 patients having old fractures in various stages of repair. McRae et al. (1973) found, of 132 battered infants, that 31 had fractures of the long bones, 12 had fractures of the skull, and 7 had other
fractures. Akbarnia et al. (1974) reviewed 231 children with battered child syndrome, and 74 had recent or remote fractures. Of these children, 25 were under the age of 6 months and 13 were 6-12 months of age. Each child had an average of 3.6 fractures, with 1-15 fractures present per child. The most common long bone fractured was the humerus (n=42), followed by the femur (n=32). There were 72 rib fractures total in the 74 children with 264 total fractures; however, rib fractures should not be considered on a one-to-one basis with fractures of the long bones—there are 24 ribs to break and only two humeri, and rib fractures can more easily occur in multiples. Worlock et al. (1986) also listed the total number of rib fractures. Once again, this practice can make the overall number of fractures identified appear high, when, in actuality, the force used to produce those fractures as well as the number of abusive episodes causing the total number of fractures may have been less. For example, the force and scenario required to break six left-sided ribs would be quite different than the force and scenario required to break two femurs, yet, in the first case, six fractures are recorded, while in the second case, two fractures are recorded. Kogutt et al. (1974) reviewed 100 children aged 6 weeks to 8 years with a diagnosis of abuse, and found that 52 had skeletal fractures (with 34 children having long bone fractures and 8 children having rib fractures). Merten et al. (1983) reviewed 904 children with a strong clinical evidence for abuse, aged 3 weeks to 16 years, with a mean of 2.2 years, and, of 494 children with a complete radiological evaluation, found that 124 patients had 155 fractures, with the most frequent bones injured being the long bones. King (1988) reviewed 750 children ruled by a social services team or the courts to have been battered, and found that 189 had fractures. The age range of children in the study was less than 1 month to 13 years, with a median age of 7 months. Of 189 children with fractures, 71 had fractures of the humerus, 66 of
the femur, 50 of the tibia, 36 of the skull, and 34 of the ribs. Multiple fractures within a given child were common. Loder and Bookout (1991) identified a total of 154 fractures in their study, with 45% being of long bones, 32% of the skull, and 20% of ribs. Therefore, it can be seen that the fracture types frequently associated with battered child syndrome and child abuse are long bone fractures, but fractures of the skull and ribs are also frequently cited. Also, the common age for presentation of battered child syndrome and child abuse is around 6 months to 1 year of age.

One important point is that the actual incidence of child abuse resulting in long bone fractures or other fractures can be difficult to determine when compared to the incidence of accidents resulting in long bone fractures and other fractures, as the incidence depends upon the population being studied and the organization of the study. For example, MacGregor (2003) reviewed 434 children presenting to an accident and emergency department due to accidents. Of the infants, 30 had fractures. Although child abuse was always considered as a possible cause of injuries, in only six cases were the injuries determined to be non-accidental in origin, and only two of these children had a fracture (one parietal skull fracture and one humeral fracture). Similarly, Rennie et al. (2007) reviewed 2198 fractures found in 2168 patients (average age was 9.7 years), and, in the entire cohort, only two confirmed cases of child abuse were identified. In contrast, Leventhal et al. (2008:603) reviewed 15,143 fractures and found that 50.4% were due to a fall and 12.08% were due to abuse, and concluded that, regarding infants, “In the first year of life, 25% of children hospitalized with fractures were abused and the incidence of fractures attributable to abuse was 36.1 cases per 100,000.” Worlock et al. (1986) reviewed 35 children who sustained fractures due to abuse, and 826 children who sustained fractures due to an accident. Of note, in children <18 months of age, 28 infants had abusive fractures and 19 had
accidental fractures. Stewart et al. (1993) reviewed 5500 infants less than 3 months of age presenting to the emergency room with traumatic injuries. In 80 infants the underlying cause was an accident and in 31 infants the underlying cause was abuse. Thus, in the five studies cited, the authors found a range of number of injuries caused by accidents versus those caused by abuse. One consideration is that the diagnostic work-up may be more thorough when abuse is suspected. For example, DiScala et al. (2000) reviewed 1997 abused children and 16,831 children who sustained accidental injuries, with all children being aged 1 day to 4 years. The children in the abused group were more likely to have undergone a CT scan and skeletal survey (in both cases, the p-value was <0.001).

**Long bone fractures**

*Incidence of long bone fractures in abuse*

Fractures of the long bones, both the diaphysis and metaphysis, are commonly reported in child abuse. Smith and Hanson (1974) studied 134 infants and children under the age of 5 years diagnosed as abused (but, the criteria to diagnosis abuse was unclear). They found 42 children had fractures, with the children having 37 skull fractures, 19 fractures of the humerus, 18 fractures of the radius and ulna, 17 fractures of the femur, 17 fractures of the tibia and fibula, and 28 fractures at other sites. Obviously and not unexpected given the sample population, many children had multiple fractures. In relative agreement as to the distribution of fractures in abused children, O’Neill et al. (1973) in a review of 110 abused infants, found that fractures of the humerus (n=20) were the most common, followed by fractures of the femur (n=17). However, other authors have identified a different distribution of long bone fractures associated with child
abuse. Loder and Bookout (1991) found 18 fractures of the humerus and 15 fractures of the femur due to abuse, but 25 fractures of the tibia. Therefore, although the relative incidence of fractures of the various long bones depends upon the author cited; the humerus, femur, and tibia rank as the most common long bones fractured in abuse.

Although the above authors describe the relative incidence of fractures of various long bones in infants who were diagnosed as abused, other authors have compared the incidence of long bone fractures between children who have been abused and those who have sustained accidental trauma. For example, Thomas et al. (1991) reviewed fractures in 215 children younger than 3 years of age who sustained injuries due to both abuse and accidents and found 11 of 14 humeral fractures were abusive in origin and 9 of 25 femoral fractures were abusive in origin. Abusive fractures were of the transverse, oblique, or greenstick varieties. Gross and Stranger (1983) reviewed 74 children, aged 0-5 years with femoral fractures, with confirmed or suspected abuse determined by a Child Protection Committee, and in children less than 1 year of age, 18 of 26 fractures were the result of abuse. Accordingly, Banaszkiewicz and Scotland (2002) said that in children younger than 1 year of age, humeral and femoral fractures were not diagnostic of abuse, but that abuse should be considered in the differential diagnosis. So, while the long bones most likely to be fractured in abuse are the humerus, the femur, and the tibia, the exact frequency reported would likely vary depending upon the patient population studied as well as other aspects of the study (e.g., type of doctor conducting the study). And, importantly, not all long bone fractures in infants and children are abusive in origin, and distinguishing abusive from accidental in origin is crucial. In this regard, the type of fracture may help distinguish between an abusive origin or an accidental origin.
Spiral fractures and abuse

In some publications, diaphyseal spiral fractures of the long bones are strongly associated with abuse. Gross and Stranger (1983:343) stated, “Infants cannot generate the rotational forces required to produce spiral fractures with straight falls.” Worlock et al. (1986) reviewed 28 infants with abusive fractures and 19 infants with accidental fractures who were <18 months of age, of seven abusive humeral shaft fractures, six were spiral, and of three abusive femoral shaft fractures, two were spiral. Worlock et al. (1986:101) stated, “Long bone fractures resulting from child abuse are mainly indirect injuries: spiral fractures and periosteal new bone formation as a result of gripping or twisting injuries or metaphyseal chip fractures from traction injuries.” Cohle and Byard (1994) describe a spiral fracture of a long bone as suspicious for child abuse. Knight (1996:462) states, “A spiral fracture of the diaphysis of the long bone must be considered a suspicious injury in infants, as such a lesion is likely to be the result of a twisting strain, unlikely to occur in accidental circumstances.” Walker (1997:203) citing Worlock et al. (1986) stated, “Spiral fractures and metaphyseal injuries are commonly seen in the long bones of abused infants.” DiMaio and DiMaio (2001:356) stated, with no citation to another source, that “spiral fractures are caused by twisting of an extremity. Especially in the non-walking child, they are highly suggestive of abuse.” In support of a non-accidental origin of spiral fractures, Tredwell et al. (1984) examined 500 consecutive fractures in the forearm of children who fell onto an outstretched arm, and found the majority of the fractures to be transverse (356 were transverse, 114 were oblique, and 30 were comminuted), and none were spiral. Thus, many authors would indicate that the finding of a spiral fracture is highly suspicious for abuse.
However, Cameron and Rae (1975:45) stated, “The spiral fracture is more common than the transverse in cases of accidental trauma.” And, Silverman (1987:223) partially agreed in that “transverse, rather than the usual oblique fractures of childhood, should be viewed with suspicion”. Neither the statement by Cameron and Rae (1975) nor Silverman (1987) was supported by a citation. However, Amir et al. (1988) reviewed 973 premature infants, 12 of whom developed fractures during their hospitalization and not from the birth process. Five infants had spiral fractures. Also, Galleno and Openheim (1982) reviewed 24 diaphyseal fractures in abused infants, and found that 17 were transverse and five were spiral. Loder and Bookout (1991) reviewed fractures in abused children, and found, of 50 shaft fractures of long bones, 28 were transverse, 18 were oblique, and four were spiral. Also, Scherl et al. (2000) reviewed 207 children aged 0-6 years with a total of 214 closed diaphyseal femur fractures. In their study population, the authors saw that children with spiral fractures were more likely to be investigated for abuse, but that transverse fractures were more common than spiral fractures in the abused children. In accordance, Scherl et al. (2000:102) indicated that considering spiral fractures as abusive in origin was a misconception and that “there are no fractures truly pathognomonic for child abuse.” However, that transverse fractures are more common in abuse than spiral fractures does not, by itself, negate the concern that spiral fractures are highly suspicious for abuse. Although Cameron and Rae (1975) opined that spiral fractures were more common in accidents than transverse fractures and while the work of Galleno and Openheim (1982) and Loder and Bookout (1991) indicated that abused infants were more likely to sustain a transverse than a spiral fracture, Cameron and Rae (1975) had no citation nor study to support their opinion, and the work of Tredwell (1984) indicates that spiral fractures are very rare, if
occurring at all, in accidental trauma. While the morphology of the fracture, whether transverse, oblique, spiral, or other, is not necessarily specific for an abusive versus an accidental mechanism, DiMaio and DiMaio (2001) are definitely correct in that fractures in non-mobile infants are always suspicious and must be adequately explained by caretakers.

**Metaphyseal fractures**

Described above in the context of long bone fractures were fractures of the diaphysis, both spiral and transverse; however, of importance in the discussion of child abuse and skeletal injuries, are injuries of the metaphysis. Kleinman et al. (2011) credit John Caffey with the first description of the metaphyseal lesion in 1957. However, Caffey (1957) was not the first person to describe lesions of the metaphysis in children. Moritz (1941) described that lacerations of the epiphyseal plate and fractures of the epiphysis were the most common skeletal injuries sustained by children. Moritz (1941:349) also said, “One of the most common types of epiphyseal injury is transverse laceration through the primary trabeculae on one side of the cartilaginous plate. Such an injury represents the shearing effect of a twisting force...” The primary trabeculae on one side of the cartilaginous plate refer to the primary spongiosa, which is considered a part of the metaphysis. A fracture at this site is a metaphyseal lesion. Thus, prior to Caffey (1957), the lesion and its mechanism of formation had already been described. However, the term, “classic metaphyseal lesion” (CML), a term that is now essentially synonymous with abuse, first appeared in the literature in 1996 (Kleinman, 2008).

Kleinman (1987b:11) described a metaphyseal lesion as “a series of microfractures occurring in a planar fashion through the most immature portion of the metaphyseal primary...
spongiosa.” The zone of calcified cartilage and a thin metaphyseal zone of primary spongiosa is on one side of the fracture line and on the other side is the remainder of the metaphysis. The lesion can be complete, extending across the entire metaphysis, or incomplete. Although the terms, “corner fracture” or “chip fracture” and “bucket-handle fracture” have been used to describe what were thought to be separate versions of metaphyseal fractures, Kleinman (1987b) describes that these are not necessarily different fracture types, but are instead different appearances of the same fracture due to different planes of orientation on the radiograph. Importantly, metaphyseal lesions may heal without subperiosteal new bone formation and can become inconspicuous in 4 weeks (O’Conner and Cohen, 1987; Kleinman, 2008) and early histologic features can include decreased cellularity (Kleinman, 2008). And, although hemorrhage in the soft tissue adjacent to bone can provide gross evidence of an underlying fracture, Kleinman (1987b:11) states that “hemorrhage is conspicuously absent” in association with metaphyseal lesions. The fact that metaphyseal lesions do not often have obvious hemorrhage associated with them and can heal without subperiosteal new bone formation both contribute to an inability to identify them grossly, and support removal of the suspected region for histologic examination.

*Mechanism of formation*

In regards to the mechanism of formation of the metaphyseal fracture and its association with child abuse, Caffey (1974:396) said

it became conclusively clear that a reasonable explanation for the pathogenesis of these common lesions--metaphyseal avulsions and subperiosteal hemorrhages--was traction-stretching stresses on the periosteum, induced by grabbing the
infants by the extremities or by the thorax, and then shaking them, which in turn induced whiplashing of the head onto the thorax.

Similar to Caffey (1974), Silverman (1974:43) discussed the mechanism of metaphyseal fractures as “lesions of the metaphyses are a common observation [in child abuse] and the most typical. Their frequency is probably related to the fact that most of the injuries are incurred not as much by direct blows as by vigorous handling, as in shaking the child. The extremities are the ‘handles’ for the ‘mishandling’.” Cameron and Rae (1975:25) supported this mechanism, saying that “these [metaphyseal lesions] occur in the region of the joint of the limbs and are caused by the baby being held by the arms or legs and jerked violently upwards or forwards, or by the baby being swung by the arms or legs and then thrown down. This sudden jerk, or wrench, of the limb causes an avulsion injury of the metaphysis and the epiphysis.” In agreement with other authors, Knight (1996) describes the mechanism of metaphyseal lesions as avulsion or chipping due to swinging, wrenching, or twisting. However, while Kleinman (1990) and Kleinman and Marks (1996b; 1998) agree that the cause of metaphyseal fractures is violent shaking by the thorax, or being twisted or swung by the extremities, instead of an avulsion, they believe the mechanism of the bone changes is a shearing of the primary spongiosa.

Association of metaphyseal fractures with child abuse

Multiple authors describe the association between metaphyseal fractures and child abuse. Silverman (1974) indicates that metaphyseal lesions, being more typical of child abuse, strengthen the diagnosis of child abuse when rib fractures are present, or in other words, in a child with rib fractures only, the diagnosis of child abuse may not necessarily be made, but when
the rib fractures are combined with the more typical metaphyseal lesion, the diagnosis of child abuse can more readily be made. Cameron and Rae (1975) discuss how a fracture of a long bone may be difficult to distinguish as the result of an accident versus as the result of an inflicted injury; however, the authors opine that the concomitant presence of a metaphyseal fracture means the injury should be interpreted as non-accidental trauma. Unfortunately, Cameron and Rae provide few studies or citations to support their interpretations. DiMaio and DiMaio (2001) indicate that epiphyseal-metaphyseal fractures of the long bones of the arms and legs exclusive of the newborn period are specific for child abuse. Cooperman and Merten (2001:130-131), in their review of the literature, state, “Metaphyseal fractures require biomechanical forces that are not produced by the usual accidental trauma of infancy. Rather rotational forces are generated as the shaken infant is held by the trunk or when the extremities are used as convenient handles for violent shaking.” Arkader et al. (2007) reviewed 117 children with distal femoral fractures, 29 of whom had a fracture through the distal femoral metaphysis. 20 of the 29 fractures occurred in children under the age of 1 year, with an average age of 6 months (one of the 20 children also had a rib fracture). Arkader et al. (2007) concluded that 75% of complete metaphyseal fractures of the distal femur in children younger than 1 year of age were highly associated with abuse. Their diagnosis of abuse was based upon a child protective services law.

Although metaphyseal fractures are associated with abuse, they are not necessarily the most frequent fractures found in abuse victims. Kogutt et al. (1974) described, of 34 children with long bone fractures, 30 had transverse or spiral fractures of the shaft, and 14 had epiphyseal-metaphyseal fractures. Merten et al. (1983) found that spiral and transverse fractures of the shaft (grouped together) were four times more common than epiphyseal-metaphyseal
fractures. Silverman (1987) described that fractures of the diaphysis of the long bone were more common than metaphyseal lesions. However, in contrast with the above three authors, Kleinman et al. (1995) identified 72 long bone fractures in 31 infants (of which not all were abused), and 64 of the 72 fractures were metaphyseal fractures.

_Other causes of metaphyseal lesions_

While Merten et al. (1983), Ellerstein and Norris (1984), Knight (1996), Hymel and Spivack (2001), DiMaio and DiMaio (2001) and Kleinman et al. (2011) indicate that metaphyseal fractures have long been recognized as highly specific for child abuse, there is a differential diagnosis when bony changes in the metaphysis of infants and children are identified that must be considered before a diagnosis of child abuse is made. Brill and Winchester (1987), Rasool and Govender (1989), Lee and Hunter (2004), Kleinman (2008), Kleinman et al. (2009), and Gabaeff (2011) describe that metaphyseal fractures or lesions similar to metaphyseal fractures can be seen in osteogenesis imperfecta (OI), congenital syphilis, rickets, Menke’s syndrome, physiologic bowing, congenital indifference to pain, and possibly scurvy. In physiologic bowing, the metaphyseal fragmentation is always incomplete, and would only appear as a corner fracture and not a bucket-handle fracture. Silverman (1974:51) described radiographs of children with epiphyseal separations of known cause (i.e., witnessed accidents), and who had radiographs obtained at least two weeks after the incident, but less than six weeks after the accident, and almost all had “metaphyseal irregularities and subperiosteal new bone formation, which was radiologically indistinguishable from that seen in the battered child.” Metaphyseal lesions have also been reported as a form of birth trauma. Weston (1957) described
metaphyseal fractures due to obstetrical trauma in two infants delivered vaginally. Lysack and Soboleski (2003) report the occurrence of a classic metaphyseal lesion (CML) of the proximal tibia following an external cephalic version of a frank breech fetus and subsequent urgent Cesarean section. The authors added that absence of a periosteal reaction and healing response in a CML in a neonate over the age of 14 days is suggestive of abuse. O’Connell and Donoghue (2007) reported three neonates with distal femur CMLs who were born by an uncomplicated Cesarean section and with no prior external cephalic version (based upon 22 years experience and 8500 babies delivered per year).

In addition to pathologic conditions and birth trauma, normal anatomic variants and accidental trauma can mimic a metaphyseal fracture. Kleinman et al. (1991) studied 78 infants who died from SIDS and found four variants that could be confused with metaphyseal lesions: a step-off (acute angulation), a beak (medial projection), a proximal tibial cortical irregularity, and a spur (projection of bone from cortex that extends past the metaphyseal margin).

Because metaphyseal lesions can occur under situations other than inflicted trauma, a determination of the incidence of these metaphyseal lesions in situations other than abusive trauma would be useful. Kleinman et al. (2011), in an effort to compare the relative likelihood of encountering a CML in an infant deemed at high risk for abuse versus in an infant deemed at low risk for abuse, reviewed radiology reports and hospital protection team consultations to identify high-risk infants. The criteria for inclusion as a high-risk infant were subdural hemorrhage, retinal hemorrhages, and skeletal injuries other than metaphyseal fractures, and skull fractures. In the high-risk group of 18 infants, there were 30 rib fractures, 18 CMLs, and five long bone fractures, while in the low-risk group, there were 42 infants who had sustained an injury either in
a short fall (31), a long fall (8), or in a complex fall (3), and none had a CML. The p-value was <.0001. This study would indicate that the likelihood of a CML occurring due to accidental trauma is significantly less likely than one occurring due to abuse.

Location of metaphyseal fractures

Common locations for metaphyseal lesions are the proximal humerus, proximal tibia, distal tibia, and distal femur (Kleinman and Marks, 1996a-c; Kleinman and Marks, 1998). However, identification of a metaphyseal lesion at the time of autopsy can be difficult. Love, a forensic anthropologist, and Sanchez, a forensic pathologist, (2009:1443) advocate gross direct visual inspection of long bones at the time of autopsy to check for the presence of CMLs, as “these injuries [CMLs] are often occult to standard autopsy and radiograph techniques.” Kleinman et al. (1986) indicate that metaphyseal lesions may have no gross abnormality (e.g., hemorrhage), and that resection, combined with high-detail radiology and histology is warranted, and, that even with no radiographic abnormality, resection of high-risk areas (proximal humerus, distal femur, proximal tibia, and distal tibia) is warranted. Kleinman (1987b) again recommend resection of the proximal humerus, distal femur, proximal tibia, and distal tibia metaphyses (for radiologic and histologic assessment) in cases where scene investigation suggests abuse, or in cases where scene investigation suggests an accident or SIDS, but skeletal survey or autopsy suggest abuse. Ellerstein and Norris (1984:1077) in a review of 460 skeletal surveys, with 331 performed to evaluated for abuse, concluded that “metaphyseal fracture, which is a common skeletal injury in abused children, may not consistently produce clinically noticeable signs and should be one of the abnormalities specifically looked for in the skeletal survey.” This statement
highlights the potential difficulty with identifying this important finding, and that extra care must be taken to conduct a thorough examination for such an important finding. Cooperman and Merten (2011) discuss the necessity of removal of the proximal humerus, distal femur, proximal tibia, and distal tibia at autopsy to search for metaphyseal lesions. Love and Sanchez (2009) and Kleinman (1986, 1987b) also advocate removal and detailed inspection of areas of the bone that are high-yield for the possible diagnosis of injuries that can help confirm abuse, but are otherwise very difficult to examine, either because of the subtle nature of radiographic analysis, or the lack of gross findings.

Occurring in the same general location as the metaphyseal lesion (at the epiphyseal-metaphyseal junction) is the Salter-Harris type fracture. Salter and Harris (1963) outlined five types of epiphyseal fractures, Types I to V. Of the five types, the only one that may be confused with a metaphyseal fracture as described above is the Salter-Harris Type II, in which the line of separation is along the cartilage, but there is one oblique branch at the end that extends to the periosteal surface at an angle through the metaphysis. Based upon this description by Salter and Harris (1963), no confusion should exist between a metaphyseal lesion and a Salter-Harris fracture; however, a review of the medical literature and accompanying images implies these two lesions may occasionally be misinterpreted as the same. And, while the metaphyseal lesion is most commonly associated with abuse, Salter-Harris fractures, including Type II, are associated with accidental injury. Kleinman (1987b) distinguished between metaphyseal lesions and Salter-Harris fractures, in that Salter-Harris occur beyond infancy and are accidental in nature. The image shown of a Salter-Harris type II indicates the fracture plane through the zone of calcified cartilage (at the end of the hypertrophic zone).
However, the images and descriptions of Salter-Harris fractures and metaphyseal lesions, and their respective associations with accidents and abuse are not consistent in the literature. Hymel and Spivack (2001) describe a metaphyseal lesion as a shearing fracture across the calcified chondrocyte column of the metaphysis and the underlying uncalcified chondrocyte column of the epiphysis. The calcified chondrocyte column can be interpreted as the final portion of the physis, just prior to the primary spongiosa, and as thus described, indicates a Salter-Harris type II fracture. Stutz and Mencio (2010), in a review article, discuss that the most common cause of distal forearm injuries is a fall on the outstretched hand, and that distal radius fractures account for 20-35% of all childhood fractures. In their diagram, listed as a Salter-Harris II, the fracture line is just distal to the physis, and has a metaphyseal fracture fragment. If the fracture line is just distal to the physis, the image is more consistent with a metaphyseal lesion.

Also, Salter-Harris fractures are not unanimously associated with accidental trauma in the medical literature. Horan and Beighton (1980) reviewed 15 children with metaphyseal irregularities. Their reassessment caused seven of the children to be relabeled as battered child syndrome. Of note, they said that the children had metaphyseal fractures, and adjacent epiphyseal damage usually of the Salter-Harris II variety. Merten et al. (1983) indicate that epiphyseal-metaphyseal fractures due to sudden violent torsion or traction of the limb in a child, can occur within the metaphysis, but can also involve the physeal plate and epiphysis, producing a Salter-Harris type fracture.
Rib fracture incidence in child abuse

Rib fractures are commonly listed with long bone fractures (including metaphyseal lesions) and skull fractures as occurring due to abusive trauma. One reason for the association of rib fractures and child abuse is that infant ribs are generally considered to be highly pliable, most likely due to the variation in composition of organic versus inorganic materials as compared to adult ribs, and therefore, relatively resistant to fracture. Thus, rib fractures are believed to more likely occur only in high-energy situations, such as that found in abusive trauma.

Schmidt (1979:105) stated that “a child’s rib cage may be pressed as far back as the spine without fracture occurring,” thus, it is not surprising that rib fractures are often listed in forensic textbooks, textbooks regarding child abuse, edited chapters, and journal articles as a feature of child abuse, or, when an infant or older child with child abuse is presented, rib fractures are frequently a member of the associated findings, because of the perceived extreme force needed to cause such injuries (Adelson, 1961; Camps, 1968; Moritz and Morris, 1970; Salmon, 1971; Fatteh, 1973; O’Neill et al. 1973; Kogutt et al. 1974; Cameron and Rae, 1975; Merten et al., 1983; Schweich and Fleisher, 1985; Zumwalt and Hirsch, 1987, Kleinman, 1987a; Silverman, 1987; Kleinman, 1998; Rivara et al., 1988; Cohle and Byard, 1994; Knight, 1996; Galloway, 1999; Parikh, 1999; DiMaio and DiMaio, 2001; Hymel and Spivack, 2001; Glass et al., 2002; Dolinak and Matshes, 2005; Shkrum and Ramsay, 2007; Worn and Jones, 2007; Dedouit, 2008). Also, some features of rib fractures are considered especially suspicious for child abuse. Thomas (1977:120) said, “The site of fracture is of considerable help. The posterior rib ends and the lower ribs favor NAI [non-accidental injury]. The lateral arcs are usually fractured in anteroposterior compression, as in resuscitation.” Fatteh (1973) and Weber et al. (2009) indicate that
ribs fractures, especially when healed or healing, multiple in number, and/or of different ages are particularly indicative of non-accidental injury.

*Importance of posterior rib fractures*

Multiple authors have opined or published studies describing the association of posterior rib fractures with child abuse. Kogutt et al. (1974) described that rib fractures associated with child abuse are most commonly posterior or lateral. Cameron and Rae (1975) described that a common site of rib fractures in the battered babies is in the posterior aspect, and that they are caused by lateral compression. Thomas (1977) in a review of 25 infants with rib fractures (out of 10,000 with chest radiographs) indicated that most infants with non-accidental injury had posterior rib fractures. Smith et al. (1980) describe four case reports with costovertebral rib fractures occurring in association with abuse. Merten et al. (1983) in a review of 904 children aged 3 weeks to 16 years (with a mean of 2.2 years) found posterior rib fractures. Carter and McCormick (1983) presented four cases of child abuse, one of which was identified by four posterior rib fractures on the left side of the chest. Smeets et al. (1990) present one abused child with posterior rib fractures. Ng and Hall (1998) identified posterior rib fractures in two infants who sustained non-accidental injury. Dolinak and Matshes (2005), Breysem et al. (2002), and Galloway (1999) discuss the association of posterior rib fractures with abuse. The fact that Dr. Galloway, a forensic anthropologist, addresses rib fractures and their association with child abuse in her edited book, indicates the importance of knowledge in this area to that field of specialists. Barsness et al. (2003) found 130 posterior fractures, 107 lateral fractures, and 66 anterior fractures in their non-accidental injury group, and, in comparison, two posterior fractures, 30
lateral fractures, and one anterior fracture in their accidental group. Hansen et al. (2008) reviewed 21 cases of child abuse, with a mean age of 4 months, and between all 21 cases, identified a total of 85 rib fractures. The mean number of rib fractures per child was 4 (with a range of 1-15), and 38% of the fractures were posterior, 48% were lateral, and only 8% were anterolateral or anterior in location.

However, not all studies or reviews link posterior rib fractures so highly with abuse. Bulloch et al. (2000) found that 5 of 7 non-abused infants had posterior rib fractures and 20 of 32 abused infants had posterior rib fractures. The difference was not significant, with a p-value of >0.05. Cadzow and Armstrong (2000) found 9 rib fractures that were accidentally inflicted, and 92 that occurred secondary to abuse. In the accidental group, 5 of the 9 fractures were posterior, and in the abused group, 39 of 92 fractures were posterior. The difference was not statistically significant.

Specificity of rib fractures for child abuse

Carty (1997:1367) said, "Rib fractures are regarded as being virtually diagnostic of child abuse, being seldom seen in infants even in response to trauma as violent as road traffic accidents", and "the concern upon their discovery is that rib fractures are commonly associated with shaking of the baby." Worlock et al. (1986) found 82 rib fractures in 35 children, all were due to abuse, and none were due to accidents, although, no abused children presented with rib fractures alone (i.e., rib fractures were always present in association with another fracture type, such as of the humerus or femur). Kleinman et al. (1996) examined 31 infants, 18 of which died from abusive head trauma and 9 of which died from SIDS. The authors identified rib fractures in
11 of the infants (not differentiating between the causes of death, e.g., SIDS versus abusive head trauma for presence of fractures), 40 fractures were found at the rib head and 10 at the costochondral junction. Kleinman et al. (1996:650), based upon their study and accompanying literature review, opined that “rib fractures are also strong indicators of infant abuse.” Walker (1997) described that pelvic and rib fractures were hard to explain in a small child other than being due to abuse. Strouse and Owings (1995) reviewed 35 children (age 1 day to 24 months). Excluding one infant with osteogenesis imperfecta (OI), the other 34 children had a total of 121 rib fractures (average of 3.9 per child), 12 were victims of child abuse and 13 had osteopenia (9 due to prematurity). Cadzow and Armstrong (2000:325) reviewed the literature regarding rib fractures and concluded that “cases of isolated rib fractures should be considered to be indicative of life-threatening physical abuse”, and “accidental rib fractures are extremely rare in infants and are accompanied by a history of massive trauma.” Bulloch et al. (2000), in a 2-1/2 year study, identified 39 infants with rib fractures. In 32 infants, the fractures were due to abuse. In three infants, rib fractures were due to accidents, in one, the fractures were due to birth trauma, and in three infants, the rib fractures were due to fragility (OI, rickets and prematurity). The authors concluded that although rib fractures in infants are uncommon that they are frequently indicative of abuse. Barsness et al. (2003) reviewed 3758 trauma evaluations of children, and identified 336 rib fractures in 78 children age 3 weeks to 15 years. Of the 78 children with rib fractures, 62 were younger than 3 years. Of the 62 children, 51 were identified as having been abused. Of the 51 abused infants, rib fractures were the only skeletal injury in 15. When the rib fractures were not due to non-accidental injury, they were more likely to be isolated. Barsness et al. (2003) determined that a rib fracture has a positive predictive value of
95% for the diagnosis of non-accidental trauma, and after the exclusion of an accident or bone
disease, the positive predictive value increased to 100%. Similarly, Kemp et al. (2008) based
upon their review of the literature (32 articles (of 439) fulfilling criteria for inclusion),
determined that, among various types of fractured bones, rib fractures had the highest probability
for abuse (0.71, with a 95% confidence interval of 0.42 to 0.91), followed by humerus fractures,
then femur fractures, then skull fractures, with skull fractures having a probability of 0.30 for
abuse (with a 95% confidence interval of 0.19 to 0.46). Each probability was based upon the
definition of abuse used by the original author of the article. Therefore, based upon review of the
literature, rib fractures are strongly associated with abuse.

Other causes of rib fractures

Rib fractures are not solely due to abusive trauma, and can occur in other situations, such
as a result of accidental trauma. Meller et al. (1984) reviewed 68 children and found rib
fractures in 19 children; however, only five had sustained abuse. Schweich and Fleisher (1985)
reviewed 21 children with rib fractures on radiographs, in 16 children the fractures were the
result of a motor vehicle accident and in five the fractures were due to abuse. The number of rib
fractures per infant was less in accidents (1-8, with a mean of 2.5) and more in abuse (3-23, with
a mean of 7). Nakayama et al. (1989) reviewed 105 children with chest injuries and found rib
fractures in 49.5%. In children aged 0-4 years, 19 of 33 had rib fractures, five children were
victims of abuse, ten sustained injuries in a motor vehicle accident, six were a pedestrian hit by a
car, one was a child who fell, one was a child in a sled accident, and nine others sustained
accidental fractures. Roux and Fisher (1992) reviewed 100 children involved in a motor vehicle
accident, and 62 had radiologically visible rib fractures (14 had one fracture, 18 had two fractures, 11 had three fractures, and 19 had four or more fractures). Bixby et al. (2011) reported a 13-month-old who sustained fractures of the right 4th-8th ribs near the costotransverse process and lateral fractures of the same ribs in a motor vehicle accident.

In addition to abusive and accidental trauma, other conditions are associated with rib fractures. Although the cause of a fracture is essentially always trauma, numerous conditions can predispose to the development of a fracture with more minor trauma than what may normally be required to produce one. Along this line, numerous review articles have been published regarding causes of fractures (Hobbs, 1989; Glass et al., 2002; van Rijn and Sieswerda-Hoogendoorn, 2012). These other causes include birth trauma, OI, rickets, prematurity, disuse osteoporosis, copper deficiency and Menke’s syndrome, Caffey’s disease, osteomyelitis, and others. While the articles may not specifically address rib fractures, in general, they serve as a reference for conditions predisposing to bone fractures, and, in the right situation, could predispose to rib fractures. The major categories will be discussed below. It must be remembered though that accidents and abuse are the two most common causes of infant and child fractures. Leventhal et al. (2008) reviewed 15,143 fractures and found that 50.4% were due to accidental falls, 12.08% were due to abuse, 0.85% were due to a bone abnormality, and 0.12% were due to a metabolic abnormality.

Identification of rib fractures

Before discussing causes of rib fractures, it is important to discuss the ability of different medical techniques to identify them, especially acute rib fractures. The identification of rib
fractures, either radiographically, in either the living or the dead, or at autopsy, is difficult. Kleinman (1987a) indicated that acute rib fractures are often overlooked radiologically. Rizzolo and Coleman (1989), Chapman (1990), Carty (1993), Kleinman et al. (1996), Ng and Hall (1998), Glass et al. (2002), Klotzbach et al. (2003); Maguire et al. (2006), Bishop et al. (2007), and Weber et al. (2009) all agree with the decreased ability of radiography to detect acute rib fractures. Ng and Hall (1998) list features contributing to this decreased ability: overlapping structures, obliquity of the fracture line in relation to the x-ray beam, poor radiographic penetration, and inadequate film-screen control. Apparently highlighting this fact, Kleinman et al. (1988) reviewed 16 infants who had sustained abuse and were examined either radiographically (n=12) or by autopsy (n=4) and found 103 posterior rib fractures, with each infant having between 2 and 24 rib fractures. Of the 12 infants examined radiologically, 87 old fractures and only one acute fracture were identified; however, in the four infants examined via autopsy, seven old rib fractures and eight acute rib fractures were identified. Kleinman et al. (1996) found, in a study of 31 infants, that 40 fractures of the rib head were very difficult to identify radiographically, even if the fractures were healing. Only 30 (of 84 total) rib fractures were identified on antemortem or postmortem radiography in 8 of 11 cases in which rib fractures were ultimately identified. Kleinman et al. (1995) in a review of 84 fractures in 11 infants, found that only 30 fractures were visible on skeletal survey. McGraw et al. (2002) reviewed 106 postmortem skeletal surveys and compared them with the autopsy findings. No rib fractures were identified by postmortem radiography, but 19 acute fractures (12 being located anterior) were identified by autopsy. Klotzbach et al. (2003) reviewed three children with a total of 16 rib fractures, all of which were old. Five paravertebral rib fractures (2-4 weeks old) were diagnosed
by skeletal survey, but six ventrolateral and six paravertebral fractures, (between 4 and 30 days of age) were diagnosed at autopsy and not identified by skeletal survey. Thus, radiography would appear to be a relatively poor tool to identify acute rib fractures.

In addition to radiographs, CT scans can be used to help identify rib fractures and thusly, Berdon and Feldman (2012) have recommended the use of computed tomography (CT) scans in the evaluation of child abuse. But, in the identification of rib fractures, CT scan, like radiography, is imperfect. Hong et al. (2011) reviewed 56 cases, with some children having a postmortem x-ray and others having a postmortem CT. Of 83 fractures identified at autopsy in children with postmortem x-rays, only 24 were found by postmortem radiography, and 49 of the 59 fractures missed by radiography were anterior. Of 101 fractures found at autopsy in children who had a postmortem CT for comparison, only 52 were found by CT, and 28 of the 49 missed fractures were anterior. Thus, CT, like radiography, appears to have difficulty with the identification of acute anterior rib fractures. In contrast, Wootton-Gorges et al. (2008) reviewed 12 infants who had sustained abusive injuries and had both radiography and CT scan evaluations, with both chest x-ray and CT scan being performed within one week of the other. No fracture was identified by chest x-ray that was not identified by CT scan. Oberladstaetter et al. (2012) compared identification of fractures via CT and autopsy in female cadavers who underwent 1 minute of CPR, and found that the CT scan missed both sternal and rib fractures. Yen et al. (2007) found that CT and MRI failed to identify lesions that were under 3 mm in size. So, while CT scans appear, in general, more effective at identifying rib fractures, they too are imperfect. However, even autopsy is not perfect for identification of rib fractures. Catteneo et al. (2006) found, that of 62 rib fractures in four piglets, radiology found 47%, CT found 34%, and autopsy
found 65%. Thus, a more extensive evaluation of the skeletal system may be necessary to identify all fractures.

Schmidt (1979), in their study, indicated that not all rib fractures were identified on radiograph and that maceration of the ribs and sternum was the best way to document all fractures. In agreement, Kemp et al. (2013) illustrated the ability of removal of soft tissue to identify help skeletal fractures not found via postmortem radiography or physical examination of the body in a decomposed individual. However, removal of soft tissue from the bones via maceration may impair identification of subtle fractures in infant ribs, and therefore, histologic examination would be more appropriate, although Love et al. (2014) opine that removal of the soft tissue is more appropriate. Similarly to Kleinman et al. (1986) advocating removal of high-risk areas to better examine the bone for metaphyseal lesions, Dolinak and Matshes (2005) advocate en-bloc removal of the left and right ribs when numerous recent or remote fractures are identified at autopsy to facilitate the search for other fractures. In agreement with Kleinman et al. (1986) and Dolinak and Matshes (2005), Catteneo et al. (2006) opined that osteologic analysis of high-risk areas may be required to properly identify fractures, and, in the case of rib fractures, this analysis may include removal of parts of the rib cage and preparation of the bone for osteologic analysis. Both McGraw et al. (2002) and Klotzbach et al. (2003) also indicate that histology is the gold standard for identification of rib fractures. As histologic examination of bones is within the realm of work conducted by forensic anthropologists, and as forensic pathologists often consult with forensic anthropologist regarding skeletal findings, forensic anthropologists knowledgeable about various techniques to examine the skeletal system to the best degree possible in the search for obvious and subtle injuries would benefit from an
understanding of possible fractures patterns that might be identified by such examination, and would in turn provide for better consultation to requesting forensic pathologists.

Cardiopulmonary resuscitation (CPR) and rib fractures

A common disagreement in the literature concerns the role of CPR in the production of rib fractures in infants. Silverman (1974) state, “Rib fractures [in abuse], recent or healing, are comparable to those seen after vigorous resuscitation activities” which would imply that CPR can produce rib fractures in infants; however, it is not certain whether or not the author was just comparing the location and nature of abusive infant rib fractures to those seen in adults after CPR. Thomas (1977) indicates that rib fractures can occur with resuscitation. Cameron and Rae (1975) also described fractures in the mid-axillary line as developing due to compression used in resuscitation attempts. However, while these early authors described rib fractures of children in association with resuscitative attempts, later authors did not so readily agree that resuscitative efforts could produce rib fractures in infants and children.

Feldman et al. (1984) reviewed radiographs from 113 living children, 41 victims of child abuse, 50 who had received CPR, and 22 who had incidental rib fractures (i.e., found during investigation for another medical condition), and 29 infants had rib fractures, 14 of these infants were victims of abuse. Other causes of rib fractures were motor vehicle accidents, rickets or osteoporosis, surgery, and OI. Feldman et al. (1984) opined that “children’s ribs are rarely, if ever, fractured by resuscitation, but frequently fractured by child abuse.” One difficulty is that their study involved living children, and, as described above, it is known that identification of acute rib fractures via radiography is difficult (Schmidt, 1979; Kleinman, 1987a; Rizzolo and
Chapman, 1989; Conway et al., 1993; Ng and Hall, 1998, Glass et al., 2002). In a review article, Hobbs (1989) stated, “Cardiopulmonary resuscitation does not cause rib fractures in this age group [infants].” Spevak et al. (1994) reviewed 91 deceased infants, age 26 hours to 8.5 months who had undergone CPR, and found no rib fractures on skeletal survey or autopsy. Spevak et al. (1994:618) did concede that “there is a small possibility that a rib fracture might elude detection by both radiography and direct inspection.” Betz and Liebhardt (1994) reviewed 233 children age 5 days to 7 years who were autopsied, and of 94 children who sustained a non-traumatic death and received CPR, none had rib fractures, in contrast to rib fractures being found in 15 of 43 children who died as the result of trauma. Bush et al. (1996) reviewed 211 deceased children who had undergone CPR, and found that, while 15 had injuries from CPR, including pulmonary hemorrhage and gastric perforation, only one had rib fractures. Ryan et al. (2003) reviewed 153 infants who died of non-traumatic causes and had undergone CPR, with none having rib fractures detected at autopsy. Thus, although radiographic examination may miss acute rib fractures due to CPR in living patients, accounting for a lack of rib fractures identified in such studies, autopsy examination also apparently fails to identify rib fractures due to CPR. However, in contrast to the above articles, Knight (1996:463), agreeing with earlier authors, describes that acute rib fractures in infants could be the result of chest compression during CPR, “even though some paediatricians and radiologists will strenuously deny the possibility of this happening.” In this discrepancy in the literature regarding CPR and the production of infant rib fractures, authors will cite the same article and report different conclusions. Galloway (1999) cites Betz (1994) and Feldman (1984) saying that rib fractures due to resuscitative efforts are a very rare
occurrence, while Shkrum and Ramsay (2007) cite Feldman (1984) to indicate that rib fractures in children may be due to CPR and not abuse.

To summarize the incidence of rib fractures in infants found associated with CPR, in a review of the literature from 1950 to 2005, Maguire et al. (2006) studied rib fractures and CPR in infants less than 18 months of age. Of 427 studies identified, they included only six in their article, excluding review articles, expert opinion, consensus guidelines and studies that were significantly methodologically flawed based upon criteria defined by the NHS Centre for Reviews and Dissemination. In the six acceptable studies, 923 children were represented, three of which had anterior rib fractures due to CPR. Importantly, Maguire et al. (2006:747) stated

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\text{there is thus always the possibility that subtle fractures may have been missed in the included studies relying on postmortems with only standard AP radiography. It is also unclear within autopsy protocols whether the parietal pleura have been reflected off the ventral surface of the posterior ribs to ensure full visualization in all cases. If this is not performed, a rib fracture is less likely to be detected, again raising the possibility of subtle fractures being missed in these studies.}
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This statement is vital to the understanding of the other studies. As previously described, radiologic techniques are not accurate in identifying acute rib fractures and accurate examination at autopsy is dependent upon the techniques and care used by the examiner. Matshes and Lew (2010b) reviewed 382 infants who died from non-traumatic causes and had CPR, and found no rib fractures. Matshes and Lew (2010b:181) report that “dissection and visual inspection, rib cage palpation, stripping of the parietal pleura, and liberal use of radiography” were used to identify rib fractures, and that “it is conceivable that small, undisplaced perimortem fractures without localized hemorrhage were, on rare occasions, overlooked.” However, Dolinak (2007) reviewed 70 consecutive autopsies on infants age 2 weeks to 8 months who had undergone CPR,
and found recent anterolateral rib fractures in 8 of the 70, and in 7 of the 8 children, there were multiple rib fractures, from 2 to 10 per infant. In reference to other articles citing a lack of rib fractures due to CPR identified at autopsy, Dolinak 2007:109) stated, “In these articles, it is not clear how actively the presence of rib fractures was pursued and whether or not the parietal pleura was stripped to optimize the detection of more subtle rib fractures.” Other recent publications have also identified rib fractures in infants associated with CPR. Clouse and Lantz (2008) described posterior rib fractures due to CPR in four hospitalized neonates or infants. Weber et al. (2009) identified 25 infants (from a sample of 546) who were found at autopsy to have rib fractures. In seven infants, with recent anterolateral fractures, the cause was determined to be CPR. In agreement with other authors, Weber et al. (2009) discussed the importance of removal of the pleura prior to final examination of the ribs. So, the presence or absence of acute rib fractures identified as reported in the medical literature is dependent upon the techniques employed. The inability of radiographs to detect acute rib fractures lends less credence to those reports based upon post-resuscitation radiographs performed on living children; whereas, autopsy technique is vitally important to the identification of acute rib fractures, and if proper technique is not followed, acute rib fractures will be missed.

The method by which resuscitation is conducted on an infant can impact whether or not rib fractures may occur. In the past, infant CPR was performed exclusively by placing the infant on the responders forearm, or a hard surface, and compressing the chest with two-fingers; however, recent changes involve holding the infant with both hands and compressing the chest with two-thumbs. Matshes and Lew (2010a) reviewed infant autopsies where two-thumb CPR was performed. In their initial page, Matshes and Lew (2010a:303) state that “cardiopulmonary
resuscitation (CPR) efforts have been offered as an explanation for numerous soft tissue, bony, and visceral injuries in young children—a possibility supported by a review of the adult literature, but not supported by a careful review of the pediatric forensic pathology literature.” The authors cited 18 papers, including several on resuscitation in adults, but not Dolinak (2007), which was available at the time they wrote their paper. The five infants in their study were determined to have sustained rib fractures due to the two-thumb CPR technique. Although Matshes and Lew (2010a) did not identify any posterior rib fractures, Worn and Jones (2007:207) stated, “The TT [two-thumb] method bears a striking resemblance to the method commonly believed by authors to be attributable to abusive compression or shaking of an infant.” Menegazzi (2011) discuss the two-thumb technique and that without lateral chest support, there is an increased risk of fracture. Reyes et al. (2011) reviewed 571 autopsied infants, age newborn to 6 months, and found rib fractures in 19, all of them of the anterior or lateral segment of the rib. Of the 19 who had rib fractures, 15 had postmortem radiography, and fractures were only seen in four infants, reinforcing the inadequacy of radiography in detecting acute rib fractures. Reyes et al. (2011) noted that the frequency of acute rib fractures identified at autopsy had increased since the mid-2006s (around the time when two-thumb CPR was first instituted), and that their study reinforced the idea that anterior and antero-lateral rib fractures can occur as a result of CPR and are not necessarily indicative of abuse. Of course, although two-thumb CPR may contribute to infant rib fractures, poorly performed two-finger CPR, or adult-type CPR (with the hands) may also subject the ribs to abnormal stress.
Birth method and skeletal injuries

Although the birthing process is associated with fractures, rib fractures are not commonly mentioned. O’Brien et al. (1966) reviewed 10,995 live births and found that the overall risk of birth trauma was 0.45%, increasing to 3.6% in infants with a midforceps delivery, 4.6% in babies weighing greater than 4500 grams, and to 22% in infants with shoulder dystocia. Valman (1979), in a review article, discussed fractures of the clavicle and humerus associated with birth, but did not mention rib fractures. Pappas (1984) described bowing of the leg at birth in 33 infants. Nadas et al. (1993) reviewed 29 infants who were referred to the pediatric surgical unit for observation or treatment after sustaining a fracture at birth. The neonates had seven depressed fractures of the skull, six fractures of the femoral shaft, six fractures of the humerus, and ten fractures of the clavicle. All six femoral fractures were associated with Cesarean section deliveries. Nadas et al. (1993) used no statistical analyses of their data and concluded that

This study now shows that the common risk factors [for fracture] are obstetrical manoeuvres during delivery, prolonged labour, cephalic presentation, and breech delivery. Cesarean section does not prevent obstetrical fractures, and, in fact, was associated with all femoral lesions. Weight, size, gestational age, age of mother, parity, gravidity, and length of delivery cannot be considered as risk factors for obstetrical fractures.

The authors did not describe rib fractures. Bhat et al. (1994) reviewed 34,946 live births and found 35 injuries, with no mention of rib fractures. The injuries were most common in neonates with an abnormal presentation, Cesarean section deliveries, and low Apgar scores. Differences in birth weight, term versus preterm, and parity were not significant. Morris et al. (2002) reviewed 55,296 live births and found seven infants with a total of eight femoral fractures. Of the seven infants, five had undergone Cesarean section, but all but one infant were less than 3050
grams, and four were breech deliveries. In a review, Pressler (2008) described the 20 most common injuries associated with birth, and rib fractures were not among them. In contrast to Nadas et al. (1993), Pressler (2008) listed risk factors for birth injuries as fetal weight of greater than 4500 grams (in mothers with diabetes) or greater than 5000 grams (in mothers without diabetes), large fetal head and very low birth weight infants, maternal age less than 16 years or greater than 35 years, primigravida, and prolonged or rapid labor; however, the author also listed, in agreement with Nadas et al. (1993), abnormal presentation and use of midcavity forceps or vacuum extraction. Sauber-Schatz et al. (2010), in a discussion of birth-related injuries, made no mention of rib fractures. Therefore, although fractures do occur as a result of the birthing process, authors do not agree on the risk factors associated with fracture development and rib fractures are not usually listed among such fractures.

Rib fractures due to birth

Even though rib fractures are not apparently a significant or commonly identified complication of birth, some reports describe rib fractures as a result of birth. Thomas (1977) described three posterior rib fractures in an infant born at 40 weeks, weighing 5896 grams and with the use of midcavity forceps. Thomas (1977) attributed the rib fractures to lateral compression during birth. Rizzolo and Coleman (1989) describe posterolateral rib fractures in a 38 weeks estimated gestational age neonate who required mechanical assistance during delivery, developed difficulty breathing nine hours after birth, and was found to have five rib fractures. Although the mother had multiple risk factors for abuse, and could conceivably have inflicted the injuries, follow-up 12 months later with the child revealed no evidence of abuse, and thus, the rib
fractures were attributed to the birth. Barry and Hocking (1993) reported a 5020-gram infant with shoulder dystocia complicating a vaginal delivery, who had five posterior rib fractures. Barry and Hocking (1993:250) stated, “When healing rib fractures are found unexpectedly in infancy, inquiries into the birth history should be made. They are not all due to NAI [non-accidental injury].”

Metabolic disorders and other natural disease processes associated with fractures

Laor and Jaramillo (1993:1035) state, “Fractures through the most immature portion of the bone, the primary spongiosa, can be found in normal bone that is under abnormal stress or in abnormal bone that is under normal stress.” Thus, the differential diagnosis of fractures in infants, as compared to inflicted or accidental causes, includes numerous metabolic disorders, as well as other natural disease processes that can result in the formation of abnormal bone. This differential diagnosis includes scurvy, syphilis, osteogenesis imperfect (OI), infantile cortical hyperostosis, severe rickets associated with low-birth-weight or prematurity, hypophosphatasia, leukemia, myelodysplasia, metastatic neuroblastoma, osteomyelitis, congenital indifference to pain, methotrexate toxicity, Menke’s syndrome, prostaglandin E1-induced cortical proliferation, secondary hyperparathyroidism, vitamin A toxicity, tuberculosis, copper deficiency, temporary brittle bone disease, vitamin K deficiency, infantile spinal muscular atrophy, calcium wasting due to renal and metabolic disorders, glucocorticoids, osteopenia due to immobility, and vitamin D deficiency (Weston, 1957; Silverman, 1974; Kerley, 1976; Grunebaum, 1980; Koo et al., 1982; Radkowski et al., 1983; Brill and Winchester, 1987; Amir et al., 1988; Paterson, 1990; Vermeer et al., 1998; Miller and Hangartner, 1999; Courtens et al., 2002; Torwalt et al., 2002; Pawley and Bishop, 2004; Jenny et al. 2006). Silverman (1974:57) said that “individuals afflicted with the
so-called congenital indifference to pain also fail to react normally to skeletal injuries and metaphyseal rarefaction...develop as a consequence just as in children with unrecognized trauma.” However, why is it that skeletal injuries and metaphyseal defects can develop as a result of an accident in those who cannot feel pain, yet in those who can feel pain, they are considered to be inflicted injury? In some cases, authors have reported fractures or osteoporosis in infants and children with no specific underlying cause identified (Fulkerson and Ozonoff, 1977; Smith, 1980; Nicol et al., 1984). In their review, Bishop et al. (2007) indicated that the two most frequent underlying diseases associated with fractures in children were metabolic bone disease of prematurity and OI. Pandya et al. (2010) in their review of the literature determined that only metaphyseal dysplasia and OI present possible diagnoses that can be confused with child abuse and neglect. Of importance in the discussion of metabolic disease of the bone and risk for fracture is the ability of radiologists to detect a decreased bone mineral density on radiograph. Finsen and Anda (1988) supported the common conception that a deficit of 30% or greater is required before decreased bone mineral density can be detected by radiography.

Prematurity and fractures

Prematurity and low-birth-weight are associated with decreased bone mineral content. Thomas (1977) found that of greater than 10,000 infants, only 25 had rib fractures, 16 of these infants were 2-4 months of age, and 13 were premature. Two of the premature infants had fractures of the posterior end of the rib and the posterior axillary region. Minton et al. (1979) reviewed 42 term infants (estimated gestational age of 38 to 40 weeks) and 30 preterm infants (estimated gestational age of 31 to 36 weeks) and found that the bone mineral content correlated
significantly with gestational age (with a p-value of <.001). Koo et al. (1982) reviewed 19 children with a birth weight of less than 1500 grams, and while 13 had no radiologic evidence of skeletal demineralization, six did, and the birth weight between the two groups was significantly different (p-value of <.001). Dabenzies and Warren (1997) reviewed 247 premature infants with birth weights between 352 and 1500 grams and with estimated gestational ages of 21 to 36 weeks, and diagnosed rickets in 96. The reason for lower bone mineral content in premature and low birth weight infants may be a variety of reasons, including inadequate intake or absorption of calcium, phosphorus or vitamin D, improper vitamin D metabolism, or an inadequate end-organ response to vitamin D (Roberts and Badger, 1984). Calcium and phosphorus deficiency in premature infants may be due to prolonged use of hyperalimentation (Dabenzies and Warren, 1997).

However, while prematurity and low-birth-weight are associated with decreased bone mineral content, the association of a decreased bone mineral content with fractures is debated. Cook et al. (1987) reviewed 17 children, aged 3-14 years, with fractures and 17 control children, and made bone mineral content determinations. They found no difference (with p-values of >.10) in age, height, weight, bone mineral content, trabecular bone density, or bone mineral density between the 17 children with fractures and those without. Cook et al. (1987:) concluded that “the results of this study indicate that a reduced bone mass is likely not to be a factor in children sustaining acute traumatic fractures.” And, “thus, it must be concluded that there is probably not a generalized mineral reduction in pediatric patients sustaining acute fractures.” In contrast, Landin and Nilsson (1983) measured the mineral content in the forearms of 90 children who had recent fractures. They defined low energy falls as falls at the same level, as from skis or
a skateboard, and high energy falls as falls from a higher level, as from a bicycle, or less than 3 meters. Landin and Nilsson (1983) found that the bone mineral content in children who sustained fractures in low-energy situations was reduced by 8% when compared to controls (a p-value of <.01). They did not specifically discuss premature infants. In contrast to Cook et al. (1987), Landin and Nilsson (1983:296) conclude that “the data indicate, rather, that there is a difference in bone mass between children who sustain a fracture due to minor trauma and control subjects without a fracture, similar to findings in the elderly.” As Koo et al. (1989) found that fractures and rickets diagnosed in premature and low-birth-weight infants had complete resolution beyond 6 months after birth, the above authors reporting on the association of decreased mineral content and fractures in older children is likely less relative to the discussion of fractures in infants. However, Dahlenburg et al. (1989) reviewed 362 infants with fractures and 362 control cases, and found that 6.8% of children were born at less than 37 weeks estimated gestational age and 1.1% were born less than 33 weeks, and that there was no statistically significant difference in the incidence of prematurity between children presenting with fractures and those used as controls. In contrast to Dahlenburg et al. (1989), Dabenzies and Warren (1997) identified 96 infants of 247 premature infants to have rickets, and of this 96 infants, 26 had fractures. Amir et al. (1988) identified eight infants of 973 premature infants who developed rib fractures during hospitalization. Callus was always present and the rib fractures did not have clinical signs. Therefore, among infants, the increased risk of fractures associated with prematurity is debated.
OI and other conditions

OI, also described as brittle bone disease, is a hereditary condition characterized by the defective synthesis of type I collagen and a resultant decreased amount of bone (Kumar et al., 2007). The incidence of OI is 1/20,000 (Taitz, 1987; Patterson and McAllion, 1989). Sillence et al. (1979) defined the most current classification scheme for OI as follows: Type I OI, the largest group, had an autosomal dominant inheritance pattern, and was associated with fractures, blue sclerae, and presenile deafness. Type II OI was autosomal recessive in most, if not all cases, with newborns presenting with neonatal fractures and dying during or soon after birth. Type III OI was sporadic, with cases having either autosomal dominant or autosomal recessive inheritance, 2/3s of newborns having fractures, and developing severe and progressive deformity; however, the sclerae were less blue than in Type I, and the blue discoloration decreased with age. Type IV OI is autosomal dominant, with variable deformity of long bones and with patients having normal sclerae. Sillence (1981) proposed two subgroups of Type IV OI, with IVA having normal teeth, and IVB having dentinogenesis imperfecta. Byers (1990), in a review article, said that dentinogenesis imperfecta was a common feature of OI Types III and IV.

Other than the above listed features, another characteristic of OI is the presence of wormian bones. In a study by Cremin et al. (1982), of 81 patients with proven OI, all but 10 had greater than 10 significant wormian bones (defined as greater than 6 x 4 mm in size and having a mosaic instead of linear architecture). Of the 10 OI patients without greater than 10 significant wormian bones, the radiographs used for analysis were of poor quality or there was insufficient ossification to properly evaluate the skull. Taitz (1987) agrees that the absence of wormian
bones is strong evidence against a child having OI, although Paterson and McAllion (1989) cite that, based upon their experience, as few as 1/3 of children with OI have multiple wormian bones. And, wormian bones are not specific to OI as children with Menke’s syndrome can have multiple wormian bones (Kleinman, 1987b). Menke’s syndrome, like OI, can be differentiated from child abuse on clinical grounds, with children having thin and coarse hair with decreased pigmentation, seizures, psychomotor retardation, failure to thrive, low serum copper, and osteopenia after 6 months of age (Kleinman, 1987b; Bacopoulou et al., 2006).

**OI versus abuse**

Of the four types of OI, Type II and III should not be confused with non-accidental injury, since both forms have recurrent multiple fractures, often present at birth, and have severe skeletal deformity, and Type I, with the blue sclerae, in most cases, should also not be confused in child abuse (Taitz, 1987; Patterson and McAllion, 1989; Chapman and Hall, 1997; vanRijn et al., 2009). Ablin et al. (1990) argue that Type IV and rare forms of Type III may be confused with abuse. In agreement, Pandya et al. (2010) in their review of the literature describe that Type IV OI is the most common type that may be confused with child abuse. Lamptey et al. (2009) reported one case of OI Type IV initially thought to be child abuse. In addition to the gross features used to distinguish Types III and IV OI from normal children with fractures due to other causes, Jones et al. (1999) described microscopic differences, with OI Types III and IV having markedly sparse and very cellular cortical and trabecular bone when compared to age-matched controls, and that primary osteonal systems continued to be formed later than expected. Sanguinetti (1990) described increased thickness of the hypertrophic zone and reduced
cellularity and shorter columns in the proliferative zone in Type I and III OI. While external examination and review of history is important in the diagnosis of OI, Marlowe et al. (2002) indicate that clinical examination alone may not be enough to detect some children with OI, and that laboratory testing is an important supplement; however, in their case list, of 138 children in which clinical information was available for review, nine had OI, and all but one had blue sclerae.

OI is commonly offered as the underlying cause for fractures that are identified in infants and children, as opposed to the possibility of child abuse; however, the frequency of each cause of fractures (i.e., OI versus abuse) in infants and children varies between authors, with some authors seemingly more likely to diagnosis OI and others more likely to diagnose abuse. Paterson (1990) reported investigating 86 children with unexpected fractures and suspected child abuse, and that most cases were due to OI or another entity, temporary brittle bone disease. In contrast, Taitz (1991) reviewed 22 infants and children (mean age of 6 months) diagnosed by another physician with OI, prematurity, or copper deficiency as the underlying cause of the fractures. Only two ultimately had OI, but also evidence of abuse, and in the other 20 infants, no evidence of an underlying metabolic disorder was identified on review, and the cause of the fractures were identified to be abuse, missed by the referring physician. In an interesting article, Paterson and Monk (2011) presented 85 cases of infants with fractures due to suspected abuse, but no subdural hemorrhage or retinal hemorrhages. Of the 85 cases, in 33 cases there was a judicial determination of abuse, in 24 cases the parents were exonerated, and in 28 cases, there was resolution without formal judicial findings. The authors, who unfortunately did not use a statistical analysis with calculated p-values, compared the birthweight, age at which the first
fracture was found, total number of fractures, number of rib fractures, and number of
metaphyseal abnormalities for the three groups, and the numbers looked similar. Paterson and
Monk (2011:98) opined that “the great differences in judicial outcome must therefore reflect
non-clinical factors”, indicating that the three groups could not be separated based upon the
above described factors. On a related note, this study highlights that factors other than physical
or pathologic anatomical findings, such as socio-economic conditions, are apparently important
in distinguishing between abuse and non-abuse.

Taitz (1987) describes that the chance for a child to have no blue sclerae, no progressive
deformity, and no family history, and a diagnosis of OI, would be 1 in 3 million. His calculation
was based upon the idea that only sporadic cases of type IV OI (those without dentinogenesis
imperfecta, progressive deformity, or other features such as wormian bones) should pose a
difficult distinction between OI and inflicted abuse as the cause of fractures in infants, combined
with the incidence data of Sillence et al. (1979), where only 9 of 180 patients with OI were type
IV and only one had no family history. Using another study, with a higher incidence of Type IV
OI, Taitz (1987) calculated the chance as 1 in 1 million. To give an example using the
calculations, in a city of 500,000 people with 6000 births per year, the chance of a less than 12
month old infant with OI, but no diagnostic features of OI, with a chance of 1-3/million, there
would be one case every 100-300 years, contrast that with about 15 cases of non-accidental
injury per year in the same city.
Other metabolic diseases predisposing to a risk for fractures

In addition to OI, other conditions already listed previously, are offered as an underlying cause of fractures in infants and children. One of these is copper deficiency. Of note, Menke’s syndrome, another potential underlying cause of fractures, is due to an inherited defect in the metabolism of copper. In their review article, Chapman and Hall (1997) describe that a full-term infant who is less than 6 months of age and breast or formula-fed should have normal copper stores and that fetal copper stores are sufficient to allow for up to 2.5 months of prematurity; however, Grunebaum et al. (1980) described four cases of clinically-proven copper deficiency, and all were full term infants. They described the features as increased density of the provisional zone of calcification, metaphyseal or diaphyseal callus formation, and sickle-shaped spurs in the metaphyseal region, among others.

Miller and Hangartner (1999), Miller (1999), and Paterson (2009) promote an entity they describe as temporary brittle bone disease as a cause of infant fractures; however, other authors refute the existence of this condition (Jenny, 2010). Miller (1999) reviewed 26 cases of temporary brittle bone disease, with 13 of 26 infants having metaphyseal fractures, 22 of 26 infants having ribs fractures, and 16 of 26 infants having posterior rib fractures. Of 17 infants with temporary brittle bone disease with multiple rib fractures, none had internal thoracic or intracranial injury. Miller (1999) critiques 1) the general acceptance of metaphyseal fractures and posterior rib fractures as being essentially considered pathognomonic for child abuse, and 2) that the finding of apparent normal bone density on plain radiographs is considered to exclude an underlying intrinsic bone disease. Miller (1999:180) states that “however, if the infant has an
intrinsic bone disease with low bone strength, a metaphyseal fracture of an extremity could result from routine handling of the infant such as in the changing of diapers or clothing.”

**Mechanism of rib fracture**

In the medical literature opinions range regarding the mechanism by which rib fractures in infants occur. As with fractures in other portions of the body, rib fractures may be due to both direct and indirect forces. One point of contention in the medical literature is the mechanism of force causing posterior rib fractures, and this point of contention is especially important given the frequent association of posterior rib fractures with child abuse. Early authors opined that lateral compression of the chest caused posterior rib fractures; however, more recent authors believe the mechanism of formation of posterior rib fractures is anterior-to-posterior compression (AP) of the chest.

O’Neill (1973) describe that rib fractures are most frequently due to crushing forces, and when the child is struck with a flat object, essentially indicating that rib fractures are due to direct forces; however, Cameron and Rae (1975) described posterior rib fractures, often bilateral, and said they were caused by lateral compression of the chest (or indirect forces), and that the infant might have associated fractures of the costo-chondral junctions. The authors also described fractures occurring as a result of AP compression being in the mid-axillary line, and that this site is the least common location for rib fractures to be seen in the battered baby syndrome. Merten et al. (1983) described that posterior rib fractures result from lateral chest compression and that AP compression results in lateral fractures. Gunther et al. (2000) examined cases to distinguish between fractures caused by CPR and those caused by abuse.
Based upon review of the medical literature, Dr. Kleinman appears to be the source of change regarding the opinion as to how posterior rib fractures are formed. Kleinman (1987a), based upon a case where 30 rib fractures were inflicted over a 6-week period on a 6-month-old child, with the abuser providing a history describing multiple episodes of shaking with AP compression of the chest, developed a hypothesis as to how posterior rib fractures can occur. The abuser provided precise information, describing palms along the lateral surface of the chest, the thumbs anterior at the midline, and the fingers posterior and that compression was front to back. Some of the posterior rib fractures were located anterior to the transverse process, and would have been shielded from blows to the back, and the periosteal reaction was only along the ventral surface of the rib. Kleinman (1987a:68) proposed (Fig. 7)

On the basis of these facts, a plausible mechanism of injury consistent with AP thoracic compression can be formulated. As the chest is compressed front to back, the rib is levered over the fulcrum of its transverse process. Stress is applied to the ventral cortex at the costovertebral junction along the posterior arch of the rib. If sufficient force is developed, the ventral cortex and periosteum are disrupted. If greater force is applied, a fracture fragment may arise from the dorsal surface of the rib.

The author also indicates that fractures along the lateral aspect are also due to AP compression. Most current sources agree with Kleinman (1987a, 1990) as to the interpretation of how posterior rib fractures occur (Knight, 1996; Parikh, 1999; Dolinak and Matshes, 2005); however, others apparently disagree. Zumwalt and Hirsch (1987), citing an earlier author, describe that AP compression produces lateral fractures, and side-to-side compression produces posterior fractures; albeit, the authors paper was published around the same time as Kleinman’s hypothesis, and the information may not have been available to them. But, DiMaio and DiMaio
(2001) indicates that posterior rib fractures are due to squeezing or direct trauma, and that lateral fractures are due to AP compression of the chest.

Malcolm (2008) also indicates that fractures of the rib neck are the result of backward and inward bowing due to lateral compression of the chest, yet the author cites Kleinman’s articles suggesting anterior-to-posterior compression of the chest as the cause.
To test his hypothesis regarding the mechanism of formation of rib fractures in infants, Kleinman and Schlesinger (1997) reviewed 10 human cadavers after sternotomy and two children with accidentally inflicted posterior rib fractures and performed experiments on three rabbits. The rabbits were compressed anterior to posterior on a firm surface (simulating CPR), following which a CT scan revealed no rib fractures. After this CT scan, the same rabbits were then compressed anterior to posterior in an abusive manner until fractures were audible or palpable. CT scans then revealed 13 fractures of the rib head or neck in the three rabbits. Kleinman and Schlesinger (1997:91) opined that this study showed how posterior rib fractures would not occur as a result of CPR (as there was no dorsal migration of the ribs, with the CPR being performed on a firm surface), and that, if there is no history of massive anteroposterior compression of the chest in an accident, that, with posterior rib fractures, “abuse should be presumed.” In summary of the above discrepancies, Worn and Jones (2007:200) in their review...
article opine “there is still no clear understanding of what forces and mechanisms of injury are involved in the production of rib fractures.”

**Morphology of rib head fracture**

The number of studies examining fractures of the rib head is limited; however, applicable statements regarding the morphology of rib fractures have appeared in other references. Kleinman (1998) describes a fracture of the rib head as disruption of the ventral bony cortex adjacent to the chondro-osseous junction, with the fracture line extending posteriorly into the region of the rib head cartilage, undercutting an osseous fragment. Kleinman et al. (1992), in their study of seven infants with rib fractures, identified 29 rib head fractures, but the author cautioned that isolated fractures of the rib head should be interpreted with caution. In their study, all infants with rib head fractures also had fractures of the rib neck. However, Malcolm (2008), under the subheading, “Non-accidental rib fractures”, presents a photomicrograph of an incomplete fracture of the posterior neck of an infant, and the fracture line extends immediately adjacent to the cartilaginous plate.

Based upon its location and description, fractures of the anterior surface of the rib head at the growth plate could be considered a form of metaphyseal lesion. Therefore, important in the identification of rib head fractures was Kleinman et al. (1986), who described that periosteal disruption does not necessarily accompany a metaphyseal lesion, and thus, there may be no periosteal reaction. If a periosteal reaction is absent, identification of the rib fracture grossly will be much more difficult.
In the clefts identified by the author, the defect was partially or completely filled with an amorphous eosinophilic material of unknown origin. Only Dolinak and Matshes (2005:395) apparently offer a description of this cleft material, as “amorphous eosinophilic fibrinous material”.

**Microscopic examination of fractures, including dating**

Histologic examination of the rib is important not just to identify fractures but also to assess the age of the fracture. Fatteh (1973) describes the importance of microscopic examination of fractures as a method to assist in the determination of the time of infliction of the injuries. Zumwalt and Fanizza-Orphanos (1990) indicate that, when examining rib fractures, that each fracture should be excised and examined.

*Radiographic dating of fractures*

Some findings to assist in the determination of the age of a fracture have been published, with the literature providing both radiologic and histologic criteria. Cameron and Rae (1975:50) describe that, in regards to radiologic but not histologic review, that callus is not visible for 7-10 days following the injury, but the authors do highlight that estimation of the age of a fractures is "...a matter of experience." O’Conner and Cohen (1987) also list features that can be used to date a fracture radiologically: periosteal new bone formation as early as 4-10 days, but peaking at 10-14 days; loss of fracture line definition as early as 10-14 days, but peaking at 14-21 days; soft callus formation as early as 10-14 days, but peaking at 14-21 days; and hard callus formation as
early as 14-21 days, but peaking at 21-42 days. O’Conner and Cohen (1987) describe soft callus as proliferation of osteoblasts, cartilage and woven bone; while a hard callus occurs when lamellar bone begins to bridge the gap. In agreement with Cameron and Rae (1975), O’Conner and Cohen (1987:111) caution that "the dating of skeletal injury and its chronologic relation to the history of injury or exposure to a suspected abuser is, at best, inexact." And, Prosser et al. (2005:1285) conclude “our analysis showed that the evidence base for current methods of radiologic dating [of fractures] is sparse. Dating of fractures in children is an inexact science.”

**Histologic dating of fractures**

Zumwalt and Fanizza-Orphanos (1990) list features used to date a fracture microscopically: periosteal thickening as early as 24 hours, but usual at 2-3 days; medullary cell proliferation as early as 24 hours, but usual at 2-3 days; microscopic appearance of cartilage and new bone as early as 4-5 days, but usual at 7-14 days; palpable calcification in the callus as early as 10 days, but usual at 2-3 weeks; microscopic bony union of the fracture as early as 18 days, but usual at 3-6 weeks; and solid uniting of fracture as early as 4-6 weeks, but usual at 6-10 weeks. Frost (1989a,b) described various stages of healing: 1) fracture formation, 2) granulation tissue (at 2 weeks), 3) callus formation (replacement of the granulation tissue with hard tissue) at 1-4 months, 4) conversion of callus to lamellar bone at 1-4 years, and 5) remodeling at 1-2 years or longer. Related to healing of fractures, and occurring due to injury to the periosteum through elevation of the periosteum, with resultant hemorrhage underneath, but without fracture of the bone, and associated with child abuse, is periosteal thickening (Kleinman et al., 1986). Kleinman et al. (1991) described how a healing fracture can be associated with cartilage
extension. In a review of radiographs from abused and non-abused infants, Tufts et al. (1982) found periosteal thickening more commonly in abused than in non-abused infants.

**Conclusion of review of the literature**

Compression of the chest, especially in infants, is a frequent component of inflicted trauma. Rib fractures are a form of injury associated with child abuse and can occur with compression of the chest. However, while they are associated with child abuse, rib fractures can also occur due to CPR, birth, and various metabolic diseases. Rib fractures can occur in a variety of locations including the anterior and lateral portion of the shaft and the rib head and neck. Fractures in the posterior segment of the rib (e.g., fractures at the neck) have been strongly associated with inflicted injury in the medical literature. Unfortunately, identification of rib fractures can be difficult, both radiographically and at the time of autopsy; yet, their identification could potentially assist investigators in evaluation of a death. As ribs fractures are difficult to identify radiologically and via gross inspection at autopsy, and as examination of high-yield areas of the skeleton to assess for injury (e.g., proximal and distal tibia to check for metaphyseal lesions) is advocated, removal and histologic examination of rib heads in infants at the time of autopsy may allow for identification of injuries otherwise undiagnosed. Evaluation of the association of rib head fractures with various other factors (e.g., possible abuse, CPR, birth, metabolic disease, socio-economic conditions) would improve the ability to use this examination in an effort to evaluate the circumstances of death of an infant.
CHAPTER 3: MATERIALS AND METHODS

General method of investigation

The method of investigation will be a retrospective analysis of the glass slides prepared for microscopic analysis of ribs taken from 90 children at autopsy, combined with a retrospective review of the material contained within the autopsy file. The material contained within an autopsy file includes, but is not limited to, nor inclusive of, the final autopsy report, autopsy notes, a coroner report, and medical records, including birth records. As part of the investigation, many coroners prepare a copy of the Sudden Unexpected Infant Death Investigation (SUIDI) form produced by the Centers for Disease Control and Prevention. This form contains numerous specifics about the circumstances of death, adult contacts with the deceased infant, past and present medical history, and some social factors, including age of the mother. The actual contents of each autopsy file and the accompanying coroner report are not standardized, and thus, given a retrospective review, not all of the same information was available for each case examined.

Basis for examination of rib heads at autopsy

Investigation of any infant death involves microscopic examination of multiple organs. For a proposed minimal acceptable investigation for a diagnosis of SIDS, Bergman et al. (1970) recommended histologic examination of the brain, heart, liver, lungs, kidneys, and any organs indicated by abnormalities identified in the medical or investigative history or upon gross examination at the time of autopsy. In agreement, Weber et al. (2012) studied 546 cases of sudden unexpected death in infancy, and 89 of 166 ultimately explained cases had the condition
causing death found upon histologic examination, 43% through examination of the lung, 8% the heart, 2% the liver, and <1% in the kidneys. The authors opined that microscopic examination of other organs had a low yield for establishing the cause of death. However, other authors have identified significant microscopic lesions in other organs, which helped identify the cause of death. For example, Sundararajan et al. (2005) and Eisenhut (2011) identified significant pathology microscopically in the diaphragm in sudden unexpected infant deaths. Sundararajan et al. (2005) opined that a full histologic examination was necessary in the investigation of sudden unexpected infant deaths, including organs other than those listed by Bergman et al. (2005) or Weber et al. (2012).

As has been described in the Review of the Literature, the rib head and neck is an area of critical importance when investigating infant deaths. The rib is so important in the diagnosis of child abuse that Cameron and Rae (1975) said, “In every case of suspected battered baby syndrome where there are no obvious rib fractures the whole length of the rib should be carefully examined for evidence of old injuries.” The AAP [American Academy of Pediatrics] Committee on Child Abuse and Neglect (2001:439) stated, “Thorough documentation of suspected skeletal injury may require specimen resection and high-detailed specimen radiography.” The rib head is an important region of the body when assessing children, and, as radiography and gross inspection are not ideal methods for the detection of injuries, histologic examination should be considered the gold standard method of examination.

McGraw et al. (2002) indicate that, when evaluating child abuse, the identification of as many injuries as possible is always best, and can help in interviews with caretakers. Conway et al. (1993) reviewed 83 children and found that bone scintigraphy detected 26 additional
abnormalities in 11 patients, when compared to radiography, with 15 of these abnormalities being rib fractures. Mandelstam et al. (2003) illustrated the complimentary nature of skeletal survey and bone scintigraphy, with each identifying fractures that the other missed in some cases in their study. Hansen et al. (2008:156) stated, “In some cases even one rib fracture may sway the diagnosis in regard to abuse: thus, there is a need for thorough, accurate documentation of rib fractures to aid in the accurate diagnosis of child abuse.”

As child abuse and inflicted trauma causing death (e.g., suffocation) can be subtle and difficult to detect, and as fractures of the posterior portion of the rib are so strongly associated with inflicted injury, and as resection of high yield segments of bone is advocated in the evaluation of child abuse or possible child abuse, microscopic examination of the rib heads in infant deaths is warranted. And, in addition to assessing for possible microscopic fractures, which may aid in the interpretation of the circumstances of the child’s death, microscopic examination of the ribs also allows for assessment of bone, bone marrow, and cartilage, and insures a representative section of skeletal muscle in each case.

**Purpose of investigation**

The purpose of this study is several fold: 1) to study microscopic rib head clefts and determine, if any, their association with birth method, CPR, cause and manner of death, and other features regarding the infant from whom the rib head sections were removed, including various socio-economic factors such as marital status of mother, age of mother, and presence of two biologic parents in the household; 2) investigation of the utility of removal of infant rib heads at autopsy for microscopic analysis, specifically, identification of features of importance in the investigation of the death that may not have been identified with only gross or radiologic
analysis of the ribs; 3) to study the association of other features of the rib head, both gross and microscopic (e.g., metaphyseal lesions, acute and remote fractures) with birth method, CPR, cause and manner of death, socio-economic factors, and other features. The goal is to provide for a better understanding of the cause of infant rib fractures, and to determine if any of their features or associated findings can be used to identify abuse. The ability to examine the skeleton and identify abuse is important to forensic anthropologists and forensic pathologists, for whom forensic anthropologists frequently consult. Although the finding of features associated with abuse may not by itself allow for a definitive determination of a cause of death as being due to inflicted trauma, the information can be made available to investigating law enforcement officers who can use it to question caretakers involved in a death, as, unfortunately, although not always readily forthcoming, a confession may be the only determining factor that a death was inflicted. Thus, a better understanding of the mechanism of rib fracture formation in infants and other children would assist both forensic anthropologists and the forensic pathologists that forensic anthropologists frequently consult for regarding skeletal trauma. An increase in the understanding of the meaning of rib head fractures would also assist physical anthropologists and archaeologists when they encounter skeletal remains.

Identification of subtle abuse, if possible, would also benefit siblings of the deceased infant. In one study, Weston (1974) found that of 36 children killed by an adult, 23 had evidence of previous injury. So, identification of an abusive injury in a living child followed by intervention may help to prevent future abuse and possible death of that child. Less satisfactory but still worthy is that the diagnosis of abuse in one deceased child may save the life of a sibling. Smith and Hanson (1974) found that of 10 battered children with a deceased sibling, four had
died under suspicious circumstances. So, while the life of a deceased child cannot be saved, identification of abuse in that child may help protect the lives of their siblings by removing them from the abusive environment.

Overview of research design

The glass slides with tissue sections of the rib head region and a variable amount of adjacent neck and shaft from ribs removed during autopsies conducted on 90 neonates, infants and young children (all less than 2 years of age) were reviewed and specific data, including number of rib heads available for analysis, the presence of microscopic clefts on the anterior surface of the rib head, and various measurements of those clefts, was collected (Appendix A). If a cleft or other feature of interest was identified, the side on which it was found (i.e., left or right) was indicated. Also, specific information about each neonate, infant, and young child (including age in months at the time of death, cause of death, manner of death, birth method (e.g., vaginal, Cesarean), whether or not CPR was conducted as part of the terminal course, estimated gestational age at time of birth, socio-economic factors (e.g., married versus unmarried mother, age of mother, type of male involved with care of child (e.g., biologic father or boyfriend), history of drug use by parents), and the presence of acute or remote gross fractures of the rib identified at autopsy) was extracted from the autopsy file (Appendix A). All information was compiled in a Numbers [Apple, Cupertino, CA] spreadsheet. Prior to data analysis, a condensed version of the above information was produced. This condensed version did not include all of the original information collected (Table 1). Once the data was collected it was analyzed using the statistical programs R [R Development Core Team, 2008] and SPSS [IBM, Armonk, NY].
<table>
<thead>
<tr>
<th>Table 1. Information contained in final data sheet used for analysis.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (in months) at death, obtained from autopsy report</td>
</tr>
<tr>
<td>Delivery method (vaginal or Cesarean), obtained from autopsy report, or medical records in autopsy file</td>
</tr>
<tr>
<td>Estimated gestational age at birth, obtained from autopsy report, or medical records in autopsy file</td>
</tr>
<tr>
<td>Sex and ancestry of child, obtained from autopsy report, coroner report, or medical records</td>
</tr>
<tr>
<td>Socio-economic factors, obtained from the autopsy report, coroner report, or medical records</td>
</tr>
<tr>
<td>• Marital status and age of mother</td>
</tr>
<tr>
<td>• Relationship of male figure in child’s death, and age of such male</td>
</tr>
<tr>
<td>• Residence type, and cleanliness of residence</td>
</tr>
<tr>
<td>• History of drug use by parents</td>
</tr>
<tr>
<td>Was CPR performed (yes/no), obtained from autopsy report or coroner report</td>
</tr>
<tr>
<td>Was CPR performed at least by EMTs (yes/no), obtained from autopsy report or coroner report</td>
</tr>
<tr>
<td>Cause of death, obtained from autopsy report</td>
</tr>
<tr>
<td>Manner of death, obtained from autopsy report</td>
</tr>
<tr>
<td>If both cause and manner were undetermined, was the death suspicious for inflicted trauma, obtained from autopsy report</td>
</tr>
<tr>
<td>Number of ribs sampled</td>
</tr>
<tr>
<td>Number of rib heads available for analysis</td>
</tr>
<tr>
<td>Number of rib heads available for analysis in 6 ribs randomized sample</td>
</tr>
<tr>
<td>Number of rib heads available for analysis in 10 ribs randomized sample</td>
</tr>
<tr>
<td>Extent of healing of cleft</td>
</tr>
<tr>
<td>• Extends to periosteum</td>
</tr>
<tr>
<td>• Number of osteoclasts present</td>
</tr>
<tr>
<td>• Presence of rim</td>
</tr>
<tr>
<td>• Healing with fibrosis, cartilage, or woven bone</td>
</tr>
<tr>
<td>Measurements of cleft</td>
</tr>
<tr>
<td>• Length of cleft from tip at growth plate to periosteum</td>
</tr>
<tr>
<td>• Distance from tip of cleft at growth plate to anterior edge of the growth plate</td>
</tr>
<tr>
<td>Whether or not cleft would be included in 6 and 10 ribs randomized samples</td>
</tr>
<tr>
<td>Presence of:</td>
</tr>
<tr>
<td>• Metaphyseal lesion with no or scant hemorrhage</td>
</tr>
<tr>
<td>• Metaphyseal lesions with eosinophilic cleft material</td>
</tr>
<tr>
<td>• Acute clefts (with no eosinophilic material)</td>
</tr>
<tr>
<td>• Salter-Harris type fractures</td>
</tr>
</tbody>
</table>
Sample description

The sample (Table 2) consists of rib sections from 90 children, 48 males and 42 females. In the sample were 74 whites, 14 Native Americans, and two Hispanics. For 59 children, the mother was married, and for 29 children, the mother was unmarried. For two children, the marital status of the mother was not available. For 12 children, the significant male figure was a boyfriend, for 68 children, the significant male figure was their biological father, for six children there was no apparent significant male figure in the child’s life, and for four children, this information was not available.

Of the children, twenty died as the result of SIDS (although in one death, there were suspicious circumstances in the history indicating the infant may have been an unwanted birth), thirty infants died while bed sharing with an adult, more than one adult, or with adults and sibling, and eighteen died as the result of an undetermined cause and manner. Of eighteen who died as the result of an undetermined cause and manner of death, ten died under suspicious circumstances, with the suspicious circumstances either based upon investigation, autopsy findings (excluding the presence or absence of the clefts currently being investigated), or both, and eight died not under suspicious circumstances, but because the possible causes included both natural and accidental manners, the manner of death was certified as undetermined (e.g., the cause of death differential might have been SIDS versus bed-sharing). Three infants died as the result of inflicted trauma. Six infants died as the result of an undetermined natural cause (with the cause of death ruled as undetermined natural cause and manner as natural). Two infants died under possible asphyxial circumstances, but which was not a bed-sharing environment. One
infant died due to head trauma of uncertain origin, but likely accidental. Ten infants died as the result of a confirmed natural death (e.g., pneumonia).

Regarding birth method and performance of CPR at the terminal event, 58 children were delivered vaginally, 25 children were delivered via Cesarean section, and the birth method was unknown in seven. Described in the medical records were two infants with shoulder dystocia, one with vacuum extraction required for delivery, and one requiring re-positioning during the birth. CPR was performed on 83 children, and not performed on six children, and a history of whether or not CPR was performed was unavailable for one child.

<table>
<thead>
<tr>
<th>Table 2: Category counts within the sample</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total number of children in entire sample</td>
</tr>
<tr>
<td>Sex of children: Males/Females</td>
</tr>
<tr>
<td>Ancestry of children: Caucasian/Native American/Hispanic</td>
</tr>
<tr>
<td>Marital status of mother: Married/unmarried/Unavailable</td>
</tr>
<tr>
<td>Status of father figure: Biological father/boyfriend/None/Unknown</td>
</tr>
<tr>
<td>Circumstances of death</td>
</tr>
<tr>
<td>• SIDS</td>
</tr>
<tr>
<td>• Bed-sharing</td>
</tr>
<tr>
<td>• Undetermined cause and manner, but suspicious circumstances</td>
</tr>
<tr>
<td>• Undetermined cause and manner, but non-suspicious circumstances</td>
</tr>
<tr>
<td>• Inflicted trauma</td>
</tr>
<tr>
<td>• Undetermined natural causes</td>
</tr>
<tr>
<td>• Possible asphyxial circumstances</td>
</tr>
<tr>
<td>• Head trauma of uncertain etiology</td>
</tr>
<tr>
<td>• Known natural</td>
</tr>
<tr>
<td>Birth method: Vaginal/Cesarean/Unknown</td>
</tr>
<tr>
<td>CPR performed: Yes/No/Unknown</td>
</tr>
</tbody>
</table>
Total numbers of ribs, rib heads and clefts analyzed

The 90 children in the sample had a mean age at death of 4.268 months (with a standard deviation of 4.337), and the age range represented was 0.03 (1 day) to 20 months (Fig. 9). The mean estimated gestational age at which the children autopsied were born was 37.975 weeks (with a standard deviation of 2.610 weeks), and the range represented was 28 to 41 weeks (Fig. 10).

![Figure 9](image1.png)  ![Figure 10](image2.png)

Figure 9. Histogram of age in months at death for each child (n=90) in the entire sample.
Figure 10. Histogram of estimated gestational age at birth (in weeks) for each child (n=90) in the entire sample.

The total number of ribs examined grossly was 2160 (24 per child). The total number of ribs originally submitted for microscopic analysis (All sample) was 972, and, of these, 851 had an intact rib head region available for examination on the glass slide. The 6R sample had 482 rib heads. The 10R sample had 781 rib heads. The 10R+ sample had 706 ribs heads. The total number of rib heads available for microscopic examination for each child varied between 3 and
20 in the All sample (in the 10R sample, the number available varied between 3 and 11) (Figs. 11 and 12).

**Figure 11.** Histogram of number of rib heads per child for analysis in entire sample.

**Figure 12.** Histogram of number of rib heads per child for analysis in the 10R sample.

**Institutional Review Board**

Approval or review of this study by the University of Montana Institutional Review Board (IRB) was not required. According to material provided by the University of Montana (University of Montana IRB, N.d.:1), which “provides guidance to UM investigators who may be uncertain if their study meets the definitions of human subjects research as stated in the federal regulations (45CFR46.102),” the material reviewed (i.e., the glass slides with sections of rib) does not constitute human subjects, specifically, the UM pamphlet (University of Montana IRB, N.d.:4) states that “cadavers, autopsy specimens or specimens/information from subjects now deceased is not human subjects.” Also, no genetic studies were conducted on the material.
used in this project and the results obtained do not impact the health or reveal the health of living patients. In review of the primary source material cited in the UM pamphlet for investigators, review of 45CFR46.101-505 (Protection of human subjects, 45CFR46.101-505 [1995]), under definitions, indicates that human subjects are a living individual about whom an investigator (whether professional or student) conducting research obtains (1) data through intervention or interaction with the individual, or (2) identifiable private information. Therefore, the analysis of the sections of the ribs did not involve human subjects research, and does not therefore require IRB review and either approval or waiver.

The information contained within the autopsy files was either obtained directly at the time of autopsy from the interview of investigators or during accumulation of outside medical records and other records, including material from birth certificates, as part of the death investigation. The amount of investigative information recommended recovered in all investigations of sudden unexplained infant deaths is extensive (Centers for Disease Control and Prevention, 2010), and includes the data extracted from the autopsy files for this research. Although not all the recommended information was necessarily obtained prior to completion of the autopsy report, the Centers for Disease Control and Prevention, based upon their death investigation form, would argue that all the information should have been obtained. Adding information to the autopsy file after completion of the report is not incorrect. The cause and manner of death certified can be amended if additional pertinent information becomes available.

According to Montana Code Annotated and the Code of Federal Regulations, confidential medical information may be released to medical examiners to assist in determination of cause and manner of death, and other duties. In addition, the Health Insurance Portability and
Accountability Act (HIPAA) Privacy rule indicates that covered entities may release protected health information under other certain circumstances. One of these circumstances is in the case of a deceased individual where the information is being sought for the purpose of research, and it is necessary for research. This disclosure by a covered entity does not require authorization from a personal representative or next of kin, or waiver of the authorization by an IRB, but the covered entity may request documentation of the death (Gostin, 2002; US Department of Health & Human Services, 2003; Centers for Disease Control and Prevention, 2003; National Institutes of Health, 2004a; National Institutes of Health, 2004b; US Department of Health & Human Services, 2006; National Institutes of Health, 2007). Thus, all information used in this study should have been available to the medical examiner conducting the death investigation. No confidential health information was obtained from a covered entity solely for the purpose of this research project, and, as is required, any confidential health information extracted from the autopsy files will be held confidential.

Specific materials and methods

Description of obtainment and processing of rib specimens

Although the rib head and neck sections were obtained as part of the autopsy itself, and this study entails review of the glass slides and information in the autopsy file, the methods by which the rib sections were obtained will be described here.

At the time of autopsy, after removal of the internal organs of the trunk, a block of left ribs and a block of right ribs (#5-#9 bilateral) was removed in the following manner: the lateral
aspect of the associated vertebral body was shaved off using the Stryker saw in an anterior to posterior cut, or, alternatively, the costovertebral cartilaginous junction between the rib head and vertebral body was incised with a scalpel. The rib shaft just distal to the rib neck and transverse process attachment was transected in an anterior to posterior vertical plane with the Stryker saw. Following these cuts, the rib block was dissected away from the body, severing attached soft tissue and incising the cartilaginous junction between the rib neck and the transverse process with a scalpel or scissors. If circumstances dictated (e.g., investigation of an infant death suspicious for inflicted trauma from investigative findings or autopsy findings), larger blocks of ribs (e.g., left and right ribs #3-10) were removed. With the exception of the two sentinel cases described above, in no case was the entire rib cage and vertebral column removed; and, therefore, as the vertebral column and rib cage was essentially intact, repair of the body by the funeral homes was not impaired. Even when the entire thoracic cage, including ribs and vertebral column, was removed, repair and subsequent open casket funeral with viewing was not precluded. Compared to the routine removal of the calotte and sternum with attached sternal ends of the ribs (the breast plate) at autopsy, the removal of the rib head blocks did not adversely affect the body with regards to future embalming or reconstruction efforts (Personal communication with Tyson Moore, funeral director, on 7-15-2013 via telephone).

After removal of the rib blocks from the body, each was fixed in formalin for a period of at least one week. Following fixation with formalin, each rib block was immersed in a 5% solution of nitric acid for the purpose of decalcification. The decalcification process was done for 24-48 hours, until the rib sections could be cut easily with a scalpel. For several children, additional rib sections were submitted up to four years past the initial autopsy to increase the
number of sections available for microscopic examination. In these cases, the decalcification process had continued to a variable extent resulting in pale hematoxylin and eosin staining, but with minimal or no distortion of architecture.

After decalcification, each rib section was cut with a scalpel in a horizontal plane from anterior to posterior through the midportion of the rib as viewed from anterior, most importantly to include a bisection of the rib head; however, due to the small nature of the ribs and their curved structure, equal bisection of the neck and any adjacent shaft in the section was not always successful. The cut sections of rib were placed in a tissue cassette, cut surface down, with one to three segments placed in each cassette. No effort to separate the ribs as to their location (e.g., rib #5 versus rib #7) was made. Following sectioning, the rib sections in their cassettes were sent in a plastic container filled with formalin to a local hospital (Clinical Laboratory Improvement Amendments (CLIA)-certified) to be processed according to their standard procedures for the purpose of production of glass slides with a section of tissue for microscopic review.

Histologic review of rib tissue sections

The glass slides with the rib sections were reviewed with an Olympus BX51 [Olympus, Center Valley, PA] microscope using 2x, 4x, 10x, 20x, and 40x objectives. In reviewing the rib sections, the following data were collected: number of left and right ribs, number of left and right rib heads available for analysis, number of left and right transverse process regions available for analysis, the number of microscopic acute and remote rib head, neck, and shaft fractures, metaphyseal lesions (with pink cleft material or with no or scant hemorrhage), the number of acute clefts with no eosinophilic material in left and right ribs, the number of clefts with
eosinophilic material in the left and right ribs, the number of healing clefts (with rim only, or with woven bone, cartilage, and/or fibrosis) in left and right ribs, the number of clefts extending to the periosteum, various measurements of the clefts (including length, area, distance from inner tip to anterior edge of growth plate, and distance to proximal and distal end of base of the fracture), and cartilage extensions (all of the above information is reflected in the data collection form found in Appendix B). After the data was collected, before statistical analyses, the ribs from the left and right sides were combined. So, for example, if an infant had four left side ribs and five right side ribs, each with a rib head available for histologic analysis, the final data recorded would have been nine ribs and nine rib heads. Also, some of the data was not included in statistical analyses because it was either relatively redundant (e.g., several measurements of various aspects of cleft size) or too subjective when recorded (e.g., presence of different forms of cartilage extensions).

Photographic documentation of findings

All abnormalities were photographed with an Olympus camera [U-TVO.5XC-3]. For most abnormalities, a photomicrograph using both the 4x and the 10x objectives was taken to allow for an overall and a close-up. For some abnormalities, the 2x and 20x or 40x objectives were used. The photographs were altered, only for contrast or removal of blemishes caused by particles of dirt, wax, or other substance superimposed on the image, using Photoshop CS5 [Adobe, San Jose, CA]. The length of the cleft and distance of the inner tip of the cleft from the anterior edge of the growth plate measurements were obtained using ImageJ [Wayne Rasband, National Institutes of Health, Bethesda, MA]. A calibration slide (scales stage micrometer) was
obtained from AmScope [Irvine, CA], and using ImageJ the length of 1 millimeter was measured on the calibration slide; this measurement was then used on the photomicrographs for measurement. Under 4x objective, 1 millimeter equals 581 pixels and under a 10x objective, 1 millimeter equals 1454 pixels, occasionally, the 2x objective was required, and the appropriate conversion was 1 millimeter equals 291 pixels. ImageJ was used to measure the feature on the photomicrograph, and the resultant measurement in pixels was converted to millimeters, by dividing by 291, 581, or 1454 as appropriate.

Collection of data

The data were collected onto record sheets (Appendix B). After all data had been collected, the information was compared to the photomicrographs to insure accuracy. During the examination of the rib sections microscopically, the other information regarding the deceased child (specifically cause and manner of death, birth history, and whether or not CPR was performed) was unavailable to me; however, the autopsy number was visible on the glass slide being examined. Also, the relative number of rib head sections available for analysis in each case was known.

Complications with data collection

In the collection of the data, two difficulties require explanation at this point so that the methods chosen for analysis can be better understood. First, the number of ribs originally submitted per child and the number of rib heads available for microscopic analysis per child were not constant. While in most cases ten ribs were submitted for histologic examination, in
some cases, more or less were submitted. When more ribs were submitted, it was most often because some aspect of the death was suspicious, either autopsy or investigation, and thus, closer evaluation of more rib heads was deemed appropriate. And, even though a rib was submitted, the rib head portion of the rib may not have been available for microscopic review, due to the section of the tissue on the slide not including the region of interest. For statistical analysis, this variation in number of rib heads per child to examine complicates matters. Simple weighting of the data by the number of rib heads available for analysis per child might not work, as each rib is not an independent event. If one rib has a fracture, the ribs adjacent to it may be more likely to be fractured than ribs more distant. For example, if left rib #6 is fractured, left ribs #5 and 7 are more likely to be fractured than left ribs #3 and 9 because the force applied to break the rib is most localized at left rib #6 and would likely decrease in intensity the further it is from this site.

To contend with this variability in the number of rib heads available for analysis, randomized datasets of ribs for each child were developed. A sample with each infant having around 6 rib heads was made and a sample with each infant having around 9-10 rib heads was made. The randomized datasets assumed that for each child in the study only 6 or 10 ribs had been chosen initially for microscopic review. Although this process involved the loss of data (e.g., a child with 20 rib heads available for histologic examination, but with only findings from 6 or 10 rib heads being used, would involve a loss of material), it did allow for the creation of relatively uniform datasets, with each child represented having a relatively equal number of rib heads available for histologic analysis. The process for producing these samples was as follows (refer to Appendix C for example):
1. The number of ribs submitted for analysis was determined (e.g., nine on the left and ten on the right).

2. If the number of rib heads available for analysis was the same as the number of ribs submitted (e.g., in the above case, nine on the left and ten on the right), go to step 4.

3. Using the statistical computer program, R [R Development Core Team, 2008], function, sample, the number of ribs on the left and right was sampled to determine which ribs had yielded the intact rib heads. Then, the process was repeated to determine which of the ribs on each side (three per side in the 6 rib sample, and five per side on the 10 rib sample) were selected for the randomized sample.

4. The ribs with a cleft or other feature of interest were numbered, using the R function, sample, and compared to the ribs sampled.

Thus, four datasets of rib heads available for analysis for each child were created from the original data, with each dataset including all children within the entire sample (unless such inclusion was impossible based upon a child having too few rib heads available for analysis, e.g., a child with 6 rib heads available for analysis could not be in the 10 rib head set). In short, the four datasets based upon number of rib heads available for analysis for each child was simply four different ways to order the entire sample, and the four datasets are 1) an All ribs sample, including all ribs removed from each child for analysis, 2) a 6 ribs randomized sample, assuming only 6 ribs had been taken from each child for analysis, 3) a 10 ribs randomized sample, assuming only 10 ribs had been taken from each child for analysis. Although a certain number of ribs were removed from each child, the actual number of rib heads available for analysis varied, because of the loss of some rib heads between collection and microscopic examination due to
sectioning artifact or histologic processing (e.g., although six ribs were submitted for a child for analysis, only three or four rib heads might have been available for microscopic analysis). Because many rib heads were lost in such a manner, to standardize the number of rib heads evaluated per child, a fourth dataset was created, 4) a 10 ribs randomized sample with 8-11 ribs per child, assuming only 10 ribs had been taken from each child for analysis and only including children where 8-11 rib heads were available for histologic examination. Only one child had 11 rib heads available for analysis without randomization and was included in the dataset. Children with 8-11 ribs were used, instead of just children with 10 ribs, in recognition of the inherent imperfections in obtaining exactly 10 rib heads for histologic analysis. For labeling purposes, the four datasets will be referred to as All (for All ribs sample), 6R (for 6 ribs randomized sample), 10R (for 10 ribs randomized sample), and 10R+ (for 10 ribs randomized sample, with 8-11 rib heads per child for analysis).

Groupings of children by cause and manner of death

The second difficulty was that while much of the data in this study is objective and either measured or otherwise determined (e.g., age at death, birth type, number of rib heads available, number of clefts, size of clefts), some of the data is also subjective (e.g., cause of death, manner of death, determination of suspicious nature of death). The cause and manner of death, while determined by the medical examiner through careful analysis of the autopsy findings, circumstances of the death (e.g., scene investigation and known medical history), and toxicology findings, is subjective. Therefore, to accommodate this subjectivity, for statistical analysis, the 90 children in the sample were divided into several groupings based upon their cause and/or
manner and circumstances of death, with each grouping containing all 90 children in the sample. Once again, as with the four datasets created based upon the number of rib heads available for analysis, the four groupings of circumstances of death was simply four different ways to order the children in the sample based upon this feature.

In general, the 90 children could be divided into five different general categories based on their cause and manner of death: 1) infants who died as the result of SIDS, 2) infants who died as the result of bed sharing, 3) infants who died as the result of non-accidental injury (i.e., inflicted trauma), or died under suspicious circumstances that indicated the possibility of inflicted trauma as a cause of death, 4) infants who died as the result of a natural cause other than SIDS, and 5) infants who died as the result of other circumstances, not suspicious for inflicted trauma, including a variety of causes of death that did not fit into one of the other four groups (e.g., an infant who died under circumstances between which bed sharing and SIDS as the cause of death could not be excluded.)

However, one infant who was certified as dying from SIDS had a suspicious historical circumstance that implied a lack of parental interest in the child, or in other words, that the child was unwanted, which may be concerning for the possibility of later abuse; but, there were no other investigative or autopsy findings suggesting abuse, and thus, the cause of death was certified as SIDS. Also, while some infants died as the result of a known natural cause (e.g., bronchopneumonia), some died as the result of a likely, but yet unknown, natural cause, but not SIDS, and were certified as undetermined natural causes. So, considering these discrepancies, four different combinations of groups, each composed of the entire sample, (Groups 1-4) were made for statistical analysis, each group in turn was composed of five subgroups (Subgroups A-
E) reflecting the placement of one child into SIDS versus suspicious, or the need to separate undetermined natural deaths from known natural deaths. The difference between Group 1 and Group 2, and Group 3 and Group 4, is that in Groups 1 and 3 the potentially suspicious SIDS death is included in the Suspicious subgroup (Subgroup C), and in Groups 2 and 4, the potentially suspicious SIDS death is included in the SIDS subgroup (Subgroup A). The difference between Groups 1 and 2 and Groups 3 and 4 is that in Groups 1 and 2, undetermined and known natural deaths are included together in Subgroup D, while in Groups 3 and 4, known natural deaths are in Subgroup D, and undetermined natural deaths are included in the miscellaneous category (Subgroup E) (Table 3). Essentially, Groups 1-4 represent four different ways of dividing the children studied into subgroups based upon the circumstances of their death. The fact that four separate groupings of the circumstances of death can be designated is an indication of the somewhat subjective nature of cause and manner of death determination.
Table 3. Groupings of cause/manner of death determinations for statistical analysis purposes.

<table>
<thead>
<tr>
<th>Main Group</th>
<th>Subgroup</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Group 1</td>
<td>A</td>
<td>SIDS</td>
</tr>
<tr>
<td></td>
<td>B</td>
<td>Bed share</td>
</tr>
<tr>
<td></td>
<td>C</td>
<td>NAI/Suspicious (including one SIDS)</td>
</tr>
<tr>
<td></td>
<td>D</td>
<td>Natural (not SIDS; includes undetermined natural)</td>
</tr>
<tr>
<td></td>
<td>E</td>
<td>Others</td>
</tr>
<tr>
<td>Group 2</td>
<td>A</td>
<td>SIDS (including one suspicious)</td>
</tr>
<tr>
<td></td>
<td>B</td>
<td>Bed share</td>
</tr>
<tr>
<td></td>
<td>C</td>
<td>NAI/Suspicious</td>
</tr>
<tr>
<td></td>
<td>D</td>
<td>Natural (not SIDS; includes undetermined natural)</td>
</tr>
<tr>
<td></td>
<td>E</td>
<td>Others</td>
</tr>
<tr>
<td>Group 3</td>
<td>A</td>
<td>SIDS</td>
</tr>
<tr>
<td></td>
<td>B</td>
<td>Bed share</td>
</tr>
<tr>
<td></td>
<td>C</td>
<td>NAI/Suspicious (including one SIDS)</td>
</tr>
<tr>
<td></td>
<td>D</td>
<td>Known natural</td>
</tr>
<tr>
<td></td>
<td>E</td>
<td>Others (includes undetermined natural)</td>
</tr>
<tr>
<td>Group 4</td>
<td>A</td>
<td>SIDS (including one suspicious)</td>
</tr>
<tr>
<td></td>
<td>B</td>
<td>Bed share</td>
</tr>
<tr>
<td></td>
<td>C</td>
<td>NAI/Suspicious</td>
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<td>D</td>
<td>Known natural</td>
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<td></td>
<td>E</td>
<td>Others (includes undetermined natural)</td>
</tr>
</tbody>
</table>

Statistical analysis methods

Statistical analyses were performed using the Apple Macintosh computer program Numbers [Apple, Cupertino, CA], and the computer statistical programs SPSS [IBM, Armonk, NY] and R [R Core Development Team, 2008]. Both R and Numbers were used for generating counts (e.g., number of infants who died from SIDS). SPSS was used for weighted statistical analyses. The remainder of the statistical analyses were performed using R (see Appendix D for R script used; note that in the R script, “E1” identifies Group 1 and “E3”, “E4”, and “E5” identify Groups 2, 3, and 4 respectively, and, when used as identifiers, the numbers “1”, “2”, “3”,

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“4”, and “5” represent Subgroups A, B, C, D, and E respectively). Two separate statistical analyses of the data were performed and designated, “preliminary” and “final”. The preliminary data analysis used various groupings of the 16 possible combinations of datasets (four datasets based upon number of rib heads available for analysis per child times four datasets based upon the possible groupings of circumstances of death, totaling 16). For example, the All rib sample, between Groups 1 through 4, or the 10R and 10R+ sample between Groups 1 through 4) for each statistical method performed, and the final analysis used only the 10R+ rib sample and compared results between Groups 3 and 4. As described, of the 16 datasets, each represented a unique grouping of the entire data sample based upon 1 of 4 methods of establishing number of rib heads available for analysis (All, 6R, 10R, or 10R+), and 1 of 4 methods for dividing the children based upon circumstances of death (Group 1-4).

**General interpretation of statistical analyses**

For the statistical analyses, non-parametric methods were favored over parametric methods for two reasons. First, the dependent variables studied, i.e., number of clefts per child, number of clefts per child with a length of greater than 1.00 mm, and number of clefts per child with a length of greater than 0.501 mm, did not have a normal distribution, and instead, apparently followed a Poisson distribution. Second, given the repercussions of a mistaken diagnosis of child abuse, a conservative method of analysis of the data is preferred.

In the Results and Discussion sections, both in the text and the tables, the exact p-value determined by the various statistical methods used will be indicated. For the purpose of determination of significance in regards to conclusions based upon the results of this research, a
p-value of less than 0.01 will be considered significant. The exact p-values are provided so that a reader may make their own determinations as to significance, understanding that lower or higher thresholds for significance may be used by others.

**Statistical methods for preliminary data analysis**

**General calculations**

The total number of rib heads available for analysis, total number of clefts, total number of clefts with a length of greater than 1.00 mm, and total number of clefts with a length of greater than 0.501 mm in each sample (i.e., All, 6R, 10R, and 10R+), and, when necessary, in each of the subcategories of Groups 1-4, and for the 1-4 month age group, 4.5-12 months age group, less than 2 weeks age group, and greater than 9 months age group, and infants with no clefts, were calculated using the function, `sum`, in Numbers. In the preliminary data analysis, sample means and standard deviations for age in months at death and estimated gestational age, and for number of clefts per child in each Group 1-4 and all four samples (All, 6R, 10R, and 10R+), were calculated using the functions, `mean` and `sd`, in R.

**Age-related comparison calculations**

Age of child at death is one variable that may affect whether or not clefts of the anterior surface of the rib head are present. To test whether or not the age of the child at death was a significant variable in determining the number of clefts per child or the number of clefts with a length of greater than 1.00 mm per child, comparison of the number of clefts per child and the
number of clefts with a length of greater than 1.00 mm per child between two age categories (1-4 months, and 4.5-12 months) was done. In addition, the proportion of ribs with a rib head cleft in children less than 2 weeks of age was compared to the proportion of ribs with a rib head cleft in children greater than 9 months of age. For the first testing of the effect of the age of the child at death upon the number of rib head clefts and clefts of a certain length, the two age categories were children age 1-4 months and children age 4.5-12 months. Note that no children in the sample had an age between 4 months and 4.5 months. These age categories were based upon the fact that most SIDS deaths occur in the 1-4 month age group (Bergman, 1970; DiMaio and DiMaio, 2001). When comparing the distribution of number of clefts and distribution of number of clefts with a length of greater than 1.00 mm per child between the different age categories (e.g., 1-4 months and 4.5-12 months), the Wilcoxon Rank Sum test was performed with R, using the appropriate function, wilcox.test. The Wilcoxon Rank Sum test was chosen for this analysis as there is a single independent variable that has two categories (i.e., two groups based upon age of death of the child) and the dependent variable has a non-normal distribution (Leeper, 2000).

To compare the proportion of number of rib heads with a cleft between infants with an age of less than two weeks and those with an age of greater than 9 months, a test of equal proportions was performed with R, using the appropriate function, prop.test.

Analysis of individual ribs

For the purpose of generating descriptive statistics regarding the clefts, the total number of ribs with a cleft, number of healing clefts in various stages, and number of ribs with a cleft of a specific length (e.g., greater than 1.00 mm) were calculated using the function, sum, in
Numbers. The mean age at death of ribs with a cleft, mean distance from the growth plate, and mean length of the cleft (and associated standard deviations) were calculated in R using the functions, mean and sd. To test whether or not the length of the cleft was related to whether or not the cleft was healed, the number of clefts that were healed and non-healed was compared between clefts that were greater than or less than 0.501 mm in length. A division of 0.501 was chosen as the mean length of the rib head clefts was near this measurement. As proportions were being tested, with the independent variable having two categories (healed or non-healed) and the dependent variable being categorical (either above or below 0.501 mm), a Chi-square and a Fisher exact test were both used for this analysis (Leeper, 2000).

Analysis of effects of estimated gestational age

Estimated gestational age (EGA) of a child at birth is one variable that may affect whether or not clefts of the anterior surface of the rib head are present. To test whether or not the EGA at birth was a significant variable in determining the number of clefts per child, the number of clefts with a length of greater than 1.00 mm per child, or the number of clefts with a length of greater than 0.501 mm, comparison of the number of clefts per child and the number of clefts with a length of greater than 1.00 mm or greater than 0.501 mm per child between two different categories of EGA were performed. As less than 37 weeks EGA is considered a less than term gestation, the number of the above clefts per child was compared between children with an EGA of 37 weeks or greater and those with an EGA of less than 37 weeks. Also, the number of the above clefts per child was compared between children with an EGA of greater than 33 weeks and with an EGA of less than 33 weeks. Counts of children with an EGA of less than 37 weeks, and
less than 33 weeks, and the number of children with clefts of various size were performed in Numbers using the function, *sum*. As EGA is a continuous independent variable and the dependent variable (i.e., number of clefts per child, number of clefts greater than 1.00 mm in length per child, and number of clefts greater than 0.501 mm in length per child) is non-normal in distribution, non-parametric correlation, specifically the Spearman method, was used in these analyses (Leeper, 2000).

Correlations comparing number of clefts per child, number of clefts greater than 1.00 mm in length, and number of clefts greater than 0.501 mm in length per child to the EGA of the child, using various circumstances of death (e.g., SIDS, bed-share) and the 10R and 10R+ samples were performed using the appropriate function, *cor*, and Spearman method, in R. The use of different combinations of circumstances of death as the sample population for this testing (e.g., only using infants who died as the result of SIDS, bed sharing, and known natural death, and not suspicious deaths) was done to attempt to remove the possible confounding factor of abuse from the statistical analysis. In addition to the above testing, to test whether or not EGA may affect the number of clefts per child, comparison of the incidence of children with at least one microscopic cleft, or greater than two microscopic clefts, and EGA of less than 37 weeks or less than 33 weeks were performed using a Chi-square and Fisher’s exact test. As above, because the independent variable has two categories (i.e., 37 weeks or greater, or less than 37 weeks, and 33 weeks or greater, or less than 33 weeks), and the dependent variable was categorical (based upon grouping into one or two groups based upon the number of microscopic clefts), the Chi-square and Fisher’s exact test were most appropriate for the analysis (Leeper, 2000).
The birth method (vaginal versus Cesarean section) and whether or not CPR was performed at the time of death are two variables that may affect whether or not clefts of the anterior surface of the rib head are present. To test whether or not the birth method (vaginal versus Cesarean section) or whether or not CPR was performed at the time of death were significant variables in determining the number of clefts per child or the number of clefts with a length of greater than 1.00 mm per child, comparisons of the number of clefts per child and the number of clefts with a length of greater than 1.00 mm per child between the two birth methods and between children with and without CPR performed were done. Comparison of the distribution of number of clefts per child, distribution of number of clefts with a length of greater than 1.00 mm or greater than 0.501 mm per child between vaginal and Cesarean section delivery and between the presence or absence of resuscitative attempts at the terminal event, among various combinations of circumstances of death using the 10R+ sample are performed in R with the Wilcoxon Rank Sum test and student’s t-test. The Wilcoxon Rank Sum test was used because the independent variable is categorical and has two independent populations (i.e., two forms of birth method, or presence or absence of CPR at death), and the dependent variable has a non-normal distribution (Leeper, 2000). For comparison, a student’s t-test, the parametric equivalent of the Wilcoxon Rank Sum test, was performed. In addition, using the Chi-square test and Fisher’s exact test, comparison of the distribution of infants with and without clefts and infants with and without a cleft of greater than 1.00 mm in length between the two birth methods, and the number of infants with and without acute clefts, and with and without CMLs with no
hemorrhage or CMLs with cleft material, and the presence or absence of CPR was performed. The Chi-square and Fisher’s exact test were performed in all these combinations because proportions were being tested, and in each case the independent variable had two categories and the dependent variable was categorical (Leeper, 2000).

Analysis of presence of clefts and the various subgroups (i.e., SIDS, bed-share, suspicious deaths, natural deaths, and other deaths)

The circumstance of death is one variable that may affect whether or not clefts of the anterior surface of the rib head are present. To test whether or not the circumstance of death of a child was a significant variable in determining the number of clefts per child or the number of clefts with a length of greater than 1.00 mm or greater than 0.501 mm per child, comparison of the number of clefts per child and the number of clefts with a length of greater than 1.00 mm or 0.501 mm per child between the various subgroup divisions of circumstances of death were performed. Also, the proportion of children with and without greater than two microscopic clefts, with and without a cleft, and with and without a cleft greater than 1.00 mm in length was compared among the five subgroups representing the circumstances of death. The second analysis described above was performed using Chi-square and Fisher’s exact test and the 10R and 10R+ samples, and Group 4. As the measure tested was proportions, and the independent variable was categorical, and the dependent variable was categorical, the Chi-square and Fisher’s exact test were appropriate (Leeper, 2000). The first analysis described above was performed using the Kruskal-Wallis test. The mean number of clefts per child, mean number of clefts per child greater than 1.00 mm in length, and mean number of clefts greater than 0.501 mm in length per child was compared between either Subgroups A-D or Subgroups A-E in Groups 1-4, and
using all samples, with the Kruskal-Wallis test in R, using the appropriate function, \textit{kruskal.test}. The Kruskal-Wallis test was appropriate as the measure tested was distributions, and not means, the independent variable was categorical, and had three or more populations (five, exactly, the five subgroups of circumstances of death), and the dependent variable (i.e., the three measures of the clefts, each one in three separate tests) has a non-normal distribution (Leeper, 2000). Follow-up pairwise comparison of the various subgroups was performed with the Wilcoxon Rank Sum test, with the function, \textit{wilcox.test}, in R. No Bonferroni correction was applied. Comparison of the effects of weighted and unweighted samples (e.g., based upon the number of rib heads available for analysis per child) was performed using analysis of variance (ANOVA), Kruskal-Wallis, and Mann-Whitney tests in SPSS. As each child did not have a consistent number of rib heads available for histologic analysis, one method for addressing this problem is by weighting the statistical analyses based upon the number of rib heads available for analysis. However, weighting was felt to be inappropriate as each rib sampled is not an independent event. The comparison of weighted and unweighted samples was performed to test this proposal.

\textit{Analysis of association of gross rib fractures}

To test the association of microscopic features with gross features, as gross features may have played a role in the determination of the circumstances of death, the number of gross rib fractures of various types per child (e.g., acute anterior, remote head/neck) were compared to various features, including number of clefts per child, number of clefts with a length of greater than 1.00 mm, and number of clefts with a length of greater than 0.501 mm. In addition, groupings based upon various features (e.g., number of children with no clefts, versus number of
children with one or more clefts), were compared to children with and without certain types of gross fractures. All analyses were performed with R using the function, \textit{cor}, spearman method, and Chi-square and Fisher’s exact test. When the analysis compared a continuous independent variable (e.g., number of gross rib fractures) to a non-normally distributed independent variable (e.g., number of clefts per child), a non-parametric correlation, specifically the Spearman method, was appropriate, and when the measure tested was proportions, and the independent variable had two categories, and the independent variable was categorical, the Chi square test and Fisher’s exact test were appropriate (Leeper, 2000).

**Statistical methods for final data analysis**

In the final data analysis, counts of the number of children in each group (Group 3 10R+ and Group 4 10R+), number of children delivered vaginally or via Cesarean section, and number of children who received cardiopulmonary resuscitation, were performed by reading data into R, and then using the function, \textit{length}. The R functions, \textit{mean} and \textit{median}, and the Sign-test and a one-sample Wilcoxon Rank Sum test were used to determine the sample median and a 95% confidence interval for the median, for the age at death of the children, the estimated gestational age, the number of clefts per child and number of clefts with a length of greater than 1.00 mm and with a length of >.501 mm for the entire sample (i.e., all children included), as well as individually for each category of cause of death (e.g., SIDS, bed-share), as well as for the non-suspicious deaths lumped together versus the suspicious category of deaths. The Sign test is in the R package, \textit{BSDA} (Arnholt, 2010), and is used to perform inferences on the median. As the rib head clefts are not a normal histologic finding, and as a child should have no rib head clefts,
for the purpose of the Sign test, the null hypothesis is that the median is zero. Both the Kruskal-Wallis and ANOVA were performed to evaluate the distribution of and mean number of clefts per child, distribution of and mean number of clefts greater than 1.00 mm in length per child, and distribution of and mean number of clefts greater than .501 mm in length per child between the five categories of cause of death. The appropriateness for using the Kruskal-Wallis test is the same as described for preliminary data analysis. To test for differences in the mean number of clefts and mean number of clefts of greater than 1.00 mm and 0.501 mm in length per child, an ANOVA was performed. Although the dependent variables tested were not normally distributed, because of the relatively large sample, in accordance with the central limit theorem, the ANOVA was performed to assess its utility in evaluation of the rib head clefts. An ANOVA test would be appropriate in this circumstance as the one independent variable (i.e., circumstance of death) is divided into five populations; however, to test the assumptions of ANOVA (i.e., normality and equal variances), both a Bartlett test and a Levene test were performed (Leeper, 2000). The Levene test is in the R package, *car* (Fox and Weisberg, 2011). Following the ANOVA, a Tukey HSD (Honestly Significant Difference) was performed to assess differences in means between all pairings of the five categories of death to determine where differences occurred. After the Kruskal-Wallis test, a pair-wise Wilcoxon Rank Sum test was performed with R, utilizing a Bonferroni correction. The pairwise Wilcoxon Rank Sum test was performed to assess differences in distributions between all pairings of the five categories of death to determine where differences occurred. The contribution of various factors to the number of clefts per child and the number of clefts with a length greater than 1.00 mm per child was assessed with a Poisson regression, with model fit assessed with the AIC statistic (Akaike’s
Information Criterion), and chi-square assessing residual deviance and degrees of freedom of residual deviance. A Poisson regression model was used because the dependent variables have a Poisson distribution and there is a mixture of categorical and continuous dependent variables (Leeper, 2000). A logistic regression to assess the contribution of various factors to suspicious versus non-suspicious deaths was performed, with model fit assessed. A logistic regression model was used because there were more than one independent variable, which was a mixture of categorical and continuous and the dependent variable was categorical (i.e., suspicious or non-suspicious) (Leeper, 2000).

Reproducibility of results

The ideal method to determine reproducibility of results would be to randomly sample 10 of the 90 cases and have a pathologist, or forensic anthropologist familiar with microscopic examination of bone, independent of the author, examine the histologic sections; however, as the material reviewed for this study is not commonly examined by pathologists or forensic anthropologists, unfamiliarity with the morphologic features specific to this study would potentially bias a reviewer. Also, as the author reviewed each slide and each photomicrograph taken of the histologic features several times during the course of the study, his knowledge of the material would bias his own independent review of the material. However, the author obtained each measurement of the length of the rib cleft only once during the course of the study, and, as the length of each cleft was an important feature, re-measurement of the lengths of the cleft will serve as a method to determine reproducibility of results.
Using the R function, *sample*, 30 rib head clefts were randomly selected. The length of these 30 rib head clefts was re-measured using ImageJ. Note that at the time the measurement is being made, only the size in pixels is available to the person making the measurement; the pixel measurement requires a conversion to millimeters with a calculator. Thus, although the measurer may have a rough estimate of the size, the actual measurement in millimeters is not visible while the measurement is being made.

Of the 30 re-measured clefts, in seven cases, the second measurement was less than the original measurement. The mean cleft length from the first set of measurements was 0.700 mm and the mean cleft length from the second set of measurements was 0.735 mm. A paired t-test indicates there is not a significant difference between the two means, with a p-value of 0.04792. The percentage obtained from dividing the difference between the original measurement and the second measurement by the original measurement varied between 0.3% and 30.5%; however, only 11 were 10% or greater, and all but two of these 11 occurred on a cleft with an original measurement of 0.999 mm in length or less. The mean percentage is 9.33%, with a standard deviation of 8.04%. Therefore, an assumption that measurements of length may vary by 10-15% of the original measurement when repeated would be appropriate. Contributions to this discrepancy include healing changes obscuring clear-cut boundaries and artifact induced when the microscopic section was produced (e.g., partial loss of tissue, not impairing identification of a histologic feature, but impairing its measurement).
CHAPTER 4: RESULTS

The results will be divided into two main sections: the first section will contain images of the various morphologic features (e.g., clefts, CMLs) identified in the rib heads and the second section will contain a written and tabular representation of the data collected and the results of statistical analyses of that data.

Morphology of rib heads

Morphology of cartilage extensions

Although cartilage extensions have been described as associated with trauma and child abuse (Kleinman et al., 1991), a reliable objective evaluation of cartilage extensions projecting into the metaphysis from the anterior edge of the growth plate was not deemed possible for two reasons plus, as infants who died at the time of birth were identified who had cartilage extensions at the anterior edge of the growth plate (Fig. 13), the morphologic change may represent a normal anatomic variant in at least some cases. The two reasons that objective evaluation of the cartilage extensions identified in the sample was not possible were 1) although some cartilage extensions appeared either thin (Fig. 14) or thick (Fig. 15), variation was prominent, and therefore, assigning a morphologic classification was subjective and 2) as the exact point at which the cartilage extension began at the growth plate was not clear; any measurement of the extension was highly subjective.
However, some cartilage extensions did appear to contain residual material such as that seen lining the clefts (Fig. 16), or even had a cleft containing eosinophilic material within the
cartilage (Fig. 17), and thus, may represent a later stage of healing of the clefts at the anterior edge of the growth plate.

Figure 16. Cartilage extension with cleft material remnant at arrow. Hematoxylin and eosin, 40x.

Figure 17. Cartilage extension with cleft material remnant at arrow. Hematoxylin and eosin, 100x.

The cartilage in the cartilage extensions at the anterior surface of the growth plate had hyaline cartilage that appeared similar to that found in the zone of reserve cartilage in the epiphysis, with evidence of neither proliferation nor hypertrophy or any other change normally found at the growth plate. Also, as was described by Kleinman et al. (1991), cartilage formation was associated with the healing of clefts (Fig. 18). Therefore, although quantification of the nature of the cartilage extensions at the anterior edge of the growth plate was not considered possible, the change, in some circumstances, does occur during the healing process.
Figure 18. Rib cleft with cartilage outgrowth. The short arrow indicates the remnants of the cleft at the anterior edge of the growth plate, and the long arrow indicates the cartilage filling the cleft. Hematoxylin and eosin, 20x.

The main type of cleft at the anterior region of the rib head that was the focus of this study is illustrated in Figure 19, and whenever the term, “cleft” is used in this paper, this is the morphologic feature cited. As will be discussed, some acute clefts without eosinophilic material were identified, and these will be referred to as “acute clefts” in this paper when the morphologic feature is cited. The cleft is a triangular-shaped defect in the primary spongiosa with the tip at the growth plate and the base at the periosteum.

The cleft is filled with an amorphous, granular, eosinophilic, acellular material of uncertain origin. Dolinak and Matshes (2005) only provide a descriptive name for this material; and review of the literature did not produce a specific terminology for, or description of, the specific content of the material. However, although the content of the clefts has not been determined by previous authors, given its histologic appearance and context of appearance, the material most likely represents some combination of necrotic tissue and fibrin and other proteinaceous material associated with inflammation and repair.
The clefts could be small (Figs. 20 and 22) or large (Fig. 21), with some located immediately adjacent to the anterior edge of the growth plate, and having a very short length (less than .501 mm), while others were much farther from the anterior edge of the growth plate, and had a much longer length (1.00 mm or greater).

Occasionally, more than one cleft was identified in a rib head (Fig. 23). Although the two clefts in the rib head in Figure 23 are similar in apparent age (with both extending to the periosteum and being associated with a similar osteoclastic reaction and cartilage outgrowth), in some cases, a nearly healed cleft could be associated with a more recent cleft.
Figure 20. Small cleft at arrow. Hematoxylin and eosin, 100x.

Figure 21. Large cleft at arrow. Hematoxylin and eosin, 40x.

Figure 22. Small cleft at arrow. Hematoxylin and eosin, 100x.
In addition to clefts filled with the eosinophilic material, acute clefts with only a break in the spongiosa were identified (Figs. 24-26). Figure 24 was taken from a neonate who died on the day of birth after vaginal delivery. Also in support of clefts caused by delivery, Figure 26 shows a cleft that was found in an infant who died one month after a vaginal delivery, with the clefts having a pronounced component of osteoclasts and fibrosis, and cartilage extension.

Figure 23. Two clefts at anterior edge of growth plate. The anterior edge of this growth plate has two clefts, both extending to the periosteum, and with cartilage extending between them, Hematoxylin and eosin, 40x.

Morphology of cleft healing

From the acute cleft stage, the clefts appeared to follow a general pattern for healing: infilling of the cleft with the amorphous eosinophilic material, followed by a thin layer forming between the cleft material and the periosteum, then infilling of the cleft with a varied combination of fibrosis, woven bone, and cartilage. Some clefts were identified that had a minimal amount of acellular eosinophilic material within them (Figs. 27-28), and could serve as...
a bridge between the acute cleft (see Figs. 24 and 25) and the clefts filled with the eosinophilic material (see Fig. 19).

Figures 24-26. Acute clefts and healing cleft after vaginal delivery. Fig. 24 (top left) and 25 (top right) illustrate acute clefts (arrow) with a break in the primary spongiosa and to or into the cartilage, but with little or no amorphous eosinophilic material filling the cleft. Both were identified in different children. Fig. 24 was found in a neonate who died on the day of birth after vaginal delivery. Fig. 26 (right) is a healing cleft that was found in a 1-month-old who had been born vaginally. Hematoxylin and eosin, 100x, 40x, 40x.

A thin rim, composed in part of osteoclasts, formed between the cleft material and the periosteum (Figs. 29-32). The rim in Figure 31, compared to the thin rim in other clefts, is actually fairly thick, but still early in development.
Figure 27. Cleft at anterior edge of growth plate. Fig. 27 illustrates a cleft containing a minimal amount of amorphous, eosinophilic material. Hematoxylin and eosin, 100x.

Figure 28. Cleft at anterior edge of growth plate. Fig. 28 illustrates a cleft containing a minimal amount of amorphous, eosinophilic material. Hematoxylin and eosin, 100x.

In data analysis regarding healing of clefts, those that extended to the periosteum were combined with those with a thin rim, as definitive identification of the thin rim was difficult. And, in some, although the cleft extended to the periosteum, there was apparently an increased number of osteoclasts and possible rim formation in a patchy distribution along the base of the cleft (Fig. 32).
Figure 29. Cleft at anterior edge of growth plate with thin rim. The cleft is filled with amorphous eosinophilic material, but there is a thin rim of cells (arrow) between the cleft and the periosteum. Hematoxylin and eosin, 100x.

Figure 30. Cleft at anterior edge of growth plate with thin rim. Like Fig. 30, this cleft has a thin rim between the cleft and the periosteum (arrow), and multiple osteoclasts are present. Hematoxylin and eosin, 100x.

Figure 31. Cleft at anterior edge of growth plate with thicker rim. The rim (arrow) is thicker than that in Fig. 29 or Fig. 30. Hematoxylin and eosin, 100x.

Figure 32. Cleft at anterior edge of growth plate with patchy rim. The arrows indicate a patchy rim between the cleft and the periosteum, but focally, the cleft extends to the periosteum. Hematoxylin and eosin, 100x.

Also, some clefts with a prominent amount of fibrosis, indicative of healing, still had extension to the periosteum (Figs. 33-34).
In concordance with Figs 33 and 34, fibrosis and infilling of the cleft apparently begins at the distal edge, and near the periosteum (Figs. 35a-b, 36).

Other healing changes (Figs 37-43) included woven bone formation and fibrosis, often near the periosteum, and cartilage extensions from the growth plate. Many healing clefts contained a residual cleft with the eosinophilic material. The healing changes themselves were variable, with healing clefts exhibiting a combination of the above-described elements. For example, some ribs had a cleft with in-filling with fibrosis distal and a cartilage outgrowth, but with a cleft that extended to the periosteum, except a partial thin rim.
Figures 35a-b, 36. Clefts with distal in-filling with fibrosis. Fig. 35a and b (top left and top right) are the same cleft, at low and high power, with the arrow indicating distal in-filling of the cleft with fibrosis. Fig. 36 (to side) is another example of a cleft with distal in-filling of the cleft with fibrosis. Hematoxylin and eosin, 40x, 100x, 40x.
Figures 37-39. Clefts with residual cleft and variable healing changes including cartilage outgrowth and woven bone formation. Fig. 37 (upper left) illustrates a healing cleft with prominent cartilage outgrowth from the growth plate and prominent woven bone formation (arrows). There is a residual cleft with amorphous eosinophilic material at the apex of the cartilage outgrowth. Fig 38 (upper right) illustrates a healing cleft with woven bone and some cartilage outgrowth (not as prominent as Fig. 37). A residual cleft is present. Fig. 39 (to side) illustrates similar changes as Fig. 38. Hematoxylin and eosin, 40x, 40x, 40x.
Figure 40-43. Clefts with healing changes. Fig. 40 (upper left) illustrates a healing cleft with prominent fibrosis, but little cartilage outgrowth or woven bone. Fig. 41 (upper right) illustrates a healing cleft with prominent fibrosis and cartilage outgrowth. Fig. 42 (lower left) illustrates a healing cleft with prominent cartilage outgrowth, but little fibrosis or woven bone. Fig. 43 (lower right) illustrates a healing cleft with prominent fibrosis and woven bone, but little cartilage (the cartilage present is associated with spicules in the spongiosa). Hematoxylin and eosin, 40x, 40x, 100x, 40x.
Other morphologic features

Some of the infant ribs evaluated were exposed to a prolonged period of decalcification prior to sectioning. This prolonged period of decalcification caused the tissue to have a generalized eosinophilia (with a loss of basophilia); however, the architecture was not disrupted, allowing for identification of clefts and other features (Figs. 44-45).

Figure 44. Cleft after prolonged decalcification. Even after extensive decalcification, and despite a generalized eosinophilia, a cleft is still identifiable (arrow). Hematoxylin and eosin, 40x.

Figure 45. Cleft after prolonged decalcification. Even after extensive decalcification, and despite a generalized eosinophilia, a cleft is still identifiable (arrow). Hematoxylin and eosin, 40x.

In addition to clefts in various states of healing, other features were identified and recorded, including metaphyseal lesions, with scant or no hemorrhage (Figs. 46-49) or with cleft material (Figs. 50a,b). Of interest, classic metaphyseal lesions (CMLs) with cleft material (see Figs. 50a,b) were found in an infant whose death was certified as SIDS.
Figure 46-49. Metaphyseal lesions with scant or no hemorrhage. Illustrated are acute metaphyseal lesions, with the fracture line extending along the growth plate through the primary spongiosa (arrow). Fig. 46 (top left) and Fig. 48 (bottom left) have a spur anterior. 20x, 20x, 20x, 40x.
Figures 50a,b. Metaphyseal lesions with cleft material. Both Fig. 50a (left, low power) and Fig. 50b (right, high power) are from an infant who was certified as dying from SIDS. The arrow in Figure 50a (left) indicates the metaphyseal lesion, extending through the spongiosa and along the growth plate. Visible in the metaphyseal lesion is an amorphous, eosinophilic material similar to that found in the rib head clefts discussed earlier. Hematoxylin and eosin, 40x, 100x.

Rarely, a healing CML was identified (Fig 51). And, finally, some rib head clefts, instead of being oriented oblique between the periosteum and growth plate, were oriented parallel to the growth plate (Fig. 52). Do these represent an early form of a CML? In addition, Salter-Harris fractures were identified and in one case, a Salter-Harris fracture was associated with a healing cleft (Figs. 53-54)
Results of data analysis of microscopic clefts--preliminary

A total of 223 clefts were identified microscopically in the All sample. In the 6R sample, 131 clefts were identified. The 10R sample had 205 clefts and the 10R+ sample had 192 clefts. The number of clefts per child varied between 0 and 12 in the All sample. When the 10R sample was used, the number of clefts per child varied between 0 and 10 (Figs. 55-56).

Of the 223 clefts in the All sample, 46 had a length of greater than 1.00 mm. In the 6R sample, there were 27 clefts with a length of greater than 1.00 mm. The 10R sample had 43 clefts with a length of greater than 1.00 mm and the 10R+ sample had 41 clefts with a length of greater than 1.00 mm. In both the All sample and the 10R sample, the number of clefts with a length of greater than 1.00 mm per child was 0 to 5 (Figs. 57-58). Of the 205 clefts in the 10R
sample, 115 had a length of greater than .501 mm, with from 0 to 8 such clefts per child, and in the 10R+ sample, 106 had a length of greater than .501 mm (Fig. 59).

Figure 53. Salter-Harris fracture. The arrow indicates a fracture extending parallel to the growth plate, but within the cartilage of the growth plate. Hematoxylin and eosin, 40x.

Figure 54. Salter-Harris fracture and healing cleft. The short arrow indicates a Salter-Harris-type fracture in the cartilage of the growth plate, while the long arrow indicates a healing (with fibrosis) cleft at the anterior edge of the rib head. Hematoxylin and eosin, 40x.

Extremes of the numbers of clefts per child associated with the circumstances of death

One infant in the All sample, who had 12 clefts (10 in the 10R and the 10R+ samples), died as the result of an undetermined cause and manner of death that was regarded as suspicious and had four clefts greater than 1.00 mm in length. Two infants, who had 10 clefts in the All sample (10 and 8 clefts in the 10R sample), died as the result of an undetermined cause and manner of death that was regarded as suspicious in one and as the result of an undetermined natural cause in the other. The child who died as the result of an undetermined cause and manner of death that was regarded as suspicious had five clefts that were greater than 1.00 mm in length,
while the child who died as the result of an undetermined natural cause had no clefts greater than 1.00 mm in length.

![Figure 55. Histogram of number of clefts per child for the entire sample.](image)

![Figure 56. Histogram of number of clefts per child for the 10R sample.](image)

![Figure 57. Histogram of number of clefts greater than 1.00 mm in length per child in the entire sample.](image)

![Figure 58. Histogram of number of clefts greater than 1.00 mm in length per child in the 10R sample.](image)
Infants with no clefts or one cleft

Twenty-four children had no clefts (with 3 to 13 rib heads available for analysis in the All sample). Four of these children died as the result of SIDS, one died as the result of an undetermined cause and manner of death that was regarded as suspicious and had 13 rib heads available for analysis, two died as the result of non-accidental injury, one of which had three rib heads available for analysis, and the other of which had 13 rib heads available for analysis. The others died as the result of undetermined natural causes, known natural causes, bed sharing, or an undetermined cause and manner that was not suspicious for inflicted trauma. The children were aged 0.03 to 9 months, with a mean age of 2.5292 months. Seventeen children had one cleft (with 5 to 12 rib heads available for analysis in the All sample). One died as the result of an undetermined cause and manner of death that was regarded as suspicious and had eight rib heads available for analysis. The other children died as the result of SIDS, bed-sharing or an unsafe sleep environment, a known natural disease, or an undetermined natural disease. Thus, in non-suspicious deaths, some children had multiple clefts of the rib heads, and, in suspicious deaths, some children had no or only one cleft of a rib head.
Comparison of 1-4 month age group and 4.5-12 month age group

There were 43 infants between 1 and 4 months of age (Table 4). In this age group, there were 421, 232, 378, and 361 ribs heads available for analysis in the All, 6R, 10R, and 10R+ samples respectively and 118, 69, 105, and 102 clefts identified in the All, 6R, 10R, and 10R+ samples respectively, with 41, 21, 38, and 38 being healing clefts. Of these infants aged 1-4 months, in the 10R sample, 16 have at least one cleft greater than 1.00 mm in length, 8 have one cleft, 5 have two clefts, 1 has three clefts, 1 has four clefts, and 1 has five clefts greater than 1.00 mm in length. There were 25 infants between 4.5 months and 12 months of age (see Table 4). In this age group, there were 231, 134, 212, and 178 rib heads available for analysis in the All, 6R, 10R, and 10R+ samples respectively and 53, 31, 50, and 48 clefts in the same samples, of which 10, 5, 10, and 9 were healing. Of these infants aged 4.5-12 months in the 10R sample, 5 have at least one cleft greater than 1.00 mm in length, 4 have one cleft, and 1 has two clefts greater than 1.00 mm in length.

<p>| Table 4. Comparison of number of clefts and age groups of children (1-12 months) |
|--------------------------------|---------------------------|---------------------------|</p>
<table>
<thead>
<tr>
<th>Age group</th>
<th>Number of clefts per number of rib heads in All sample</th>
<th>Number of clefts with length of greater than 1.00 mm in All sample</th>
</tr>
</thead>
<tbody>
<tr>
<td>1-4 months (n=43)</td>
<td>118 of 421</td>
<td>22 of 421</td>
</tr>
<tr>
<td>4.5 to 12 months (n=25)</td>
<td>53 of 231</td>
<td>6 of 231</td>
</tr>
</tbody>
</table>
Wilcoxon Rank Sum testing comparing the distribution of the number of clefts per child between infants aged 1-4 months and infants aged 4.5-12 months indicated a p-value of 0.4189 when the 10R sample was used and a p-value of 0.7858 when the 10R+ sample was used. Wilcoxon Rank Sum testing comparing the distribution of the number of clefts greater than 1.00 mm in length per child between infants aged 1-4 months and infants aged 4.5-12 months indicated a p-value of 0.1173 for both the 10R and the 10R+ sample. The function, `prop.test`, in R comparing the proportion of healing clefts in infants aged 1-4 months and the proportion of healing clefts in infants aged 4.5-12 months indicated a p-value of 0.06401. These age groups (i.e., 1-4 months and 4.5-12 months) were compared because a large portion of autopsies are conducted on children in the 1-4 months age group, as this is the most frequent age at which SIDS occurs. None of the p-values indicated are significant.

Comparison between the less than 2 weeks old age group and the greater than 9 months old age group

When it was noticed that children of less than 2 weeks of age had few clefts, while older child had numerous clefts, comparison of these two age categories was deemed appropriate. Eleven of the 90 children were 2 weeks or less of age. In these children, of 105 total rib heads evaluated, only three had clefts (Table 5). These clefts were all found in one infant who was 2 weeks old and died while bed sharing. In contrast, eight of the 90 children were greater than 9 months of age, and, of 77 total ribs heads evaluated in this group, 42 clefts were identified (with between 1-10 clefts identified per child in the All sample). The function, `prop.test`, in R comparing the proportion of number of rib heads with clefts in infants less than 2 weeks of age
and those greater than 9 months of age (used total number of clefts and total number of ribs) indicated a p-value of $5.647^{-15}$. This p-value is highly significant.

<table>
<thead>
<tr>
<th>Age group</th>
<th>Number of clefts per number of rib heads in All sample</th>
</tr>
</thead>
<tbody>
<tr>
<td>Less than 2 weeks (n=11)</td>
<td>3 of 105</td>
</tr>
<tr>
<td>Greater than 9 months of age (n=8)</td>
<td>42 of 77</td>
</tr>
</tbody>
</table>

Table 5. Comparison of number of clefts and age groups of children (less than 2 weeks and greater than 9 months)

Data analysis of individual ribs with clefts

If each rib with a cleft is considered as an individual, the number of ribs with a cleft is 223. The mean age at death of ribs with a cleft is 5.792 months (with a standard deviation of 5.509 months and a range of 0.5-20 months). For all 223 clefts, the mean distance of the tip of the cleft at the growth plate to the anterior edge of the growth plate was 0.589 mm, with a range of 0.001 to 3.265 mm, and a standard deviation of 0.550 mm. For all 223 clefts, the mean length of the cleft from the growth plate to the periosteum was 0.676 mm, with a range of 0.001 to 2.467 mm, and a standard deviation of 0.479 mm.

Of the 223 clefts, 68 were healed or healing (the range of ages associated with these clefts was 0.75 to 20.00 months), 49 had a rim between the eosinophilic material within the cleft and the periosteum (the range of ages associated with these clefts was 0.50 to 20.00 months), and 107 clefts extended between the periosteum and the growth plate, with no evidence of healing, including formation of woven bone, cartilage, or fibrosis, or with evidence of a rim (the range of ages associated with these clefts was 0.5 to 20.00 months). The length of the cleft and whether or not it was significantly healed were compared. For the purpose of these calculations, those
clefts showing some combination of woven bone formation, fibrosis, and cartilage production that occupied a noticeable portion of the cleft were classified as healed, and those clefts where the eosinophilic cleft material extended to the periosteum, or where a thin rim was between the cleft material and the periosteum, were classified as non-healed. Table 6 compares the size of the cleft and whether or not it is healed or non-healed. There are two comparisons of healed versus non-healed based upon different cut-off points (0.501 and 1.00 cm) for length of cleft. The table represents the total number of clefts found in the entire sample of all 90 children, and does not indicate a number per child.

Table 6. Comparison of number of healed and non-healed clefts based upon size of cleft

<table>
<thead>
<tr>
<th>Cleft size</th>
<th>Healed</th>
<th>Non-healed</th>
</tr>
</thead>
<tbody>
<tr>
<td>Less than 0.501 mm in length</td>
<td>8</td>
<td>93</td>
</tr>
<tr>
<td>Greater than 0.500 mm in length</td>
<td>60</td>
<td>62</td>
</tr>
<tr>
<td>Less than 1.00 mm in length</td>
<td>34</td>
<td>144</td>
</tr>
<tr>
<td>Greater than 0.999 mm in length</td>
<td>34</td>
<td>11</td>
</tr>
</tbody>
</table>

Of the 11 clefts that were 1.00 mm or greater in length and not healing, two were found in one child who died from SIDS and was born vaginally, one was found in a child who died from a natural cause (other than SIDS) and was born by Cesarean section, five were found in 4 children who died while bed-sharing with all four born vaginally, and two were found in infants who died from an undetermined cause and manner of death that was suspicious.

Comparison of the distribution of the number of rib clefts that were healed and non-healed divided between those clefts greater than 0.500 mm in length and those less than 0.500
mm in length, revealed a p-value of $7.231^{-11}$ by Chi-square analysis, and a p-value of $4.739^{-12}$ by Fisher’s exact test. Comparison of the distribution of the number of rib clefts that were healed and non-healed and divided between those clefts greater than 1.00 mm in length and less than 1.00 mm in length, revealed a p-value of $7.605^{-13}$ by Chi-square analysis, and a p-value of $1.98^{-12}$ by Fisher exact test. All four p-values listed above are highly significant.

**Effects of randomization of datasets**

Before statistical analysis of differences in the number of clefts per child and the number of clefts greater than 1.00 mm per child between the various groups are performed, two points must be made to foster an appreciation for the loss of data in developing the randomized sets for data analysis. First, a chart indicating the total number of rib heads available for analysis and the total number of rib head clefts identified for each of several groups is provided as well as these same numbers and others, when the 10R and 10R+ samples are considered against the All sample (Tables 7 and 8a,b). Second, Wilcoxon Rank Sum test comparing the distribution of number of clefts per child in Group 4 All sample against the Group 4 6R sample, the Group 4 10R sample, and the Group 4 10R+ sample each individually was performed, indicating no statistical significance.
Table 7: Number of children in each circumstance of death category, and total number of rib heads available for analysis and total number of rib head clefts identified in the All sample

<table>
<thead>
<tr>
<th></th>
<th>Number of deaths in total sample</th>
<th>Overall number of rib heads in this category</th>
<th>Total number of clefts</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bed-sharing</td>
<td>30</td>
<td>285</td>
<td>74</td>
</tr>
<tr>
<td>SIDS, not including suspicious</td>
<td>19</td>
<td>175</td>
<td>41</td>
</tr>
<tr>
<td>SIDS, including suspicious</td>
<td>20</td>
<td>185</td>
<td>45</td>
</tr>
<tr>
<td>Undetermined and suspicious plus NAI (without suspicious SIDS)</td>
<td>13</td>
<td>148</td>
<td>77</td>
</tr>
<tr>
<td>Undetermined and suspicious plus NAI (with suspicious SIDS)</td>
<td>14</td>
<td>158</td>
<td>81</td>
</tr>
<tr>
<td>Known natural</td>
<td>10</td>
<td>81</td>
<td>16</td>
</tr>
<tr>
<td>Others (unknown natural, possible asphyxia)</td>
<td>17</td>
<td>152</td>
<td>31</td>
</tr>
<tr>
<td>Shoulder dystocia</td>
<td>4</td>
<td>30</td>
<td>5</td>
</tr>
</tbody>
</table>

Table 8a: Number of children in each circumstance of death category, and total number of rib heads available for analysis and total number of rib head clefts of each size identified in the 10R sample

<table>
<thead>
<tr>
<th></th>
<th>N</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bed-sharing</td>
<td>30</td>
<td>265</td>
<td>69</td>
<td>54</td>
<td>15</td>
<td>29</td>
<td>40</td>
</tr>
<tr>
<td>SIDS, not including suspicious</td>
<td>19</td>
<td>172</td>
<td>40</td>
<td>33</td>
<td>7</td>
<td>17</td>
<td>23</td>
</tr>
<tr>
<td>SIDS, including suspicious</td>
<td>20</td>
<td>182</td>
<td>44</td>
<td>35</td>
<td>9</td>
<td>19</td>
<td>25</td>
</tr>
<tr>
<td>Undetermined and suspicious plus NAI (without suspicious SIDS)</td>
<td>13</td>
<td>108</td>
<td>50</td>
<td>33</td>
<td>17</td>
<td>15</td>
<td>35</td>
</tr>
<tr>
<td>Undetermined and suspicious plus NAI (with suspicious SIDS)</td>
<td>14</td>
<td>118</td>
<td>54</td>
<td>35</td>
<td>19</td>
<td>17</td>
<td>37</td>
</tr>
<tr>
<td>Known natural</td>
<td>10</td>
<td>79</td>
<td>16</td>
<td>16</td>
<td>0</td>
<td>11</td>
<td>5</td>
</tr>
<tr>
<td>Others (unknown natural, possible asphyxia)</td>
<td>17</td>
<td>147</td>
<td>27</td>
<td>25</td>
<td>2</td>
<td>17</td>
<td>10</td>
</tr>
<tr>
<td>Shoulder dystocia</td>
<td>4</td>
<td>30</td>
<td>5</td>
<td>5</td>
<td>0</td>
<td>2</td>
<td>3</td>
</tr>
</tbody>
</table>

1: Number of rib heads
2: Total number of clefts
3: Number of clefts with length of <1.00 mm
4: Number of clefts with length of >1.00 mm
5: Number of clefts with length of < or = 0.501 mm
6: Number of clefts with length of >0.501 mm
Table 8b: Number of children in each circumstance of death category, and total number of rib heads available for analysis and total number of rib head clefts of each size identified in the 10R+ sample

<table>
<thead>
<tr>
<th></th>
<th>N</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bed-sharing</td>
<td>27</td>
<td>247</td>
<td>61</td>
<td>47</td>
<td>14</td>
<td>27</td>
<td>34</td>
</tr>
<tr>
<td>SIDS, not including suspicious</td>
<td>18</td>
<td>166</td>
<td>40</td>
<td>33</td>
<td>7</td>
<td>17</td>
<td>23</td>
</tr>
<tr>
<td>SIDS, including suspicious</td>
<td>19</td>
<td>176</td>
<td>44</td>
<td>35</td>
<td>9</td>
<td>19</td>
<td>25</td>
</tr>
<tr>
<td>Undetermined and suspicious plus NAI (without suspicious SIDS)</td>
<td>11</td>
<td>98</td>
<td>50</td>
<td>33</td>
<td>17</td>
<td>15</td>
<td>35</td>
</tr>
<tr>
<td>Undetermined and suspicious plus NAI (with suspicious SIDS)</td>
<td>12</td>
<td>108</td>
<td>54</td>
<td>35</td>
<td>19</td>
<td>17</td>
<td>37</td>
</tr>
<tr>
<td>Known natural</td>
<td>7</td>
<td>63</td>
<td>12</td>
<td>12</td>
<td>0</td>
<td>9</td>
<td>3</td>
</tr>
<tr>
<td>Others (unknown natural, possible asphyxia)</td>
<td>14</td>
<td>122</td>
<td>26</td>
<td>25</td>
<td>1</td>
<td>17</td>
<td>9</td>
</tr>
</tbody>
</table>

1: Number of rib heads
2: Total number of clefts
3: Number of clefts with length of <1.00 mm
4: Number of clefts with length of >1.00 mm
5: Number of clefts with length of < or = 0.501 mm
6: Number of clefts with length of >0.501 mm

Mean number of clefts per Group and Subgroup

The mean number of clefts per child (and standard deviation) per Subgroup, considering each Group and across all four datasets (All, 6R, 10R, and 10R+), using unweighted samples, was determined (Table 9).
<table>
<thead>
<tr>
<th>Table 9. Mean number of clefts (and standard deviation) per child</th>
</tr>
</thead>
<tbody>
<tr>
<td>All sample</td>
</tr>
<tr>
<td>-----------</td>
</tr>
<tr>
<td><strong>Group 1</strong></td>
</tr>
<tr>
<td>A</td>
</tr>
<tr>
<td>B</td>
</tr>
<tr>
<td>C</td>
</tr>
<tr>
<td>D</td>
</tr>
<tr>
<td>E</td>
</tr>
<tr>
<td><strong>Group 2</strong></td>
</tr>
<tr>
<td>A</td>
</tr>
<tr>
<td>B</td>
</tr>
<tr>
<td>C</td>
</tr>
<tr>
<td>D</td>
</tr>
<tr>
<td>E</td>
</tr>
<tr>
<td><strong>Group 3</strong></td>
</tr>
<tr>
<td>A</td>
</tr>
<tr>
<td>B</td>
</tr>
<tr>
<td>C</td>
</tr>
<tr>
<td>D</td>
</tr>
<tr>
<td>E</td>
</tr>
<tr>
<td><strong>Group 4</strong></td>
</tr>
<tr>
<td>A</td>
</tr>
<tr>
<td>B</td>
</tr>
<tr>
<td>C</td>
</tr>
<tr>
<td>D</td>
</tr>
<tr>
<td>E</td>
</tr>
</tbody>
</table>

Results of analysis of effects of estimated gestational age

Seventeen children had an estimated gestational age (EGA) of less than 37 weeks and three had an EGA of less than 33 weeks. These seventeen children have 148 rib heads available for analysis in the 10R sample, and, within these 148 rib heads, 52 clefts are identified. One
child had 10 clefts, one each in the 10 rib heads available for analysis. Of the 52 clefts, 23 are healing clefts. Of the 17 children with an EGA of less than 37 weeks, five have at least one cleft greater than 1.00 mm in length (one has 5 clefts, one has 3 clefts, two have 2 clefts, and one has 1 cleft). The children with the five and three clefts with a length greater than 1.00 mm died as the result of an undetermined cause and manner of death that was suspicious. The infants with two clefts greater than 1.00 mm in length died as the result of SIDS, and the child with one cleft that was greater than 1.00 mm in length died while bed-sharing. Of three infants born at less than 33 weeks EGA, none have at least one cleft greater than 1.00 mm in length, and of 23 rib heads available for analysis in the 10R sample, there are only two clefts, one of which is healing. A Spearman correlation, comparing the EGA of various Groups and Subgroups to number of clefts per child, number of clefts greater than 1.00 mm in length per child, and number of clefts greater than 0.501 mm in length per child (Table 10), indicated a not quite significant p-value of 0.01271 when correlating Group 1, Subgroups A and D to the number of clefts when using the 10R sample, and a significant p-value of 0.006648 for the 10R+ sample when using the same Group and Subgroups, and a non-significant p-value of 0.04067 when correlating the Group 4, Subgroups A and D to the number of clefts when using the 10R+ sample. Thus, when using certain groupings of circumstances of death (Group 1) and the most natural death sample (SIDS and other naturals), there is a correlation between EGA and the number of clefts per child, and, as expected if a short EGA is a risk factor for fracture, the correlation between the two is negative (-0.5007) Chi-square analysis and the Fisher exact test comparing the various distributions of cleft features with combinations of estimated gestational age indicated non-significant p-values of 0.09692 (Chi-square) and 0.05543 (Fisher exact test) when comparing the
distribution of children with an EGA of greater or less than 37 weeks and the presence or absence of at least one microscopic cleft (Table 11).

<table>
<thead>
<tr>
<th>Group 1: A-E 10R (n=90)</th>
<th>Number of clefts per child</th>
<th>Number of clefts &gt;1 mm in length per child</th>
<th>Number of clefts &gt;0.501 mm in length per child</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>-0.0312</td>
<td>0.7834</td>
<td>0.0115</td>
</tr>
<tr>
<td></td>
<td>0.7834</td>
<td></td>
<td>0.9193</td>
</tr>
</tbody>
</table>

| Group 1: A-E 10R+ (n=77) |                          |                                           | 0.0180                                        |
|                          | -0.1513                   | 0.2181                                    | 0.9193                                        |

| Group 1: A and D 10R (n=35) |                          |                                           | -0.0453                                       |
|                            | -0.4291                   | 0.01271                                   | 0.9482                                        |

| Group 1: A and D 10R+ (n=30) |                          | 0.0118                                    | -0.1852                                       |
|                            | -0.0838                   | 0.4968                                    | 0.7138                                        |

| Group 1: A, B, D, E 10R (n=76) |                          |                                           | -0.1529                                       |
|                              | -0.5007                   | 0.006648                                  | 0.4371                                        |

| Group 1: A, B, D, E 10R+ (n=65) |                          |                                           | -0.0498                                       |
|                               | -0.0101                   | 0.9594                                    | 0.6915                                        |

| Group 3: A, D 10R (n=29) |                          |                                           | -0.1102                                       |
|                          | -0.3470                   | 0.07044                                   | 0.8185                                        |

| Group 3: A, D 10R+ (n=25) |                          |                                           | -0.0454                                       |
|                           | -0.4207                   | 0.04067                                   | 0.9866                                        |

| Group 3: A, B, D, E 10R (n=76) |                          |                                           | -0.0498                                       |
|                              | -0.1155                   | 0.3557                                    | 0.6915                                        |

| Group 3: A, B, D, E 10R+ (n=65) |                          |                                           | -0.1102                                       |
|                               | -0.2471                   | 0.06631                                   | 0.4188                                        |

| Group 4: A, D 10R (n=30) |                          |                                           | -0.1362                                       |
|                          | -0.3814                   | 0.04121                                   | 0.6915                                        |

| Group 4: A, D 10R+ (n=26) |                          |                                           | -0.0332                                       |
|                           | -0.4564                   | 0.02185                                   | 0.8749                                        |

| Group 4: A, B, D, E 10R (n=77) |                          |                                           | -0.0644                                       |
|                              | -0.1362                   | 0.2716                                    | 0.6044                                        |

| Group 4: A, B, D, E 10R+ (n=66) |                          |                                           | -0.1259                                       |
|                               | -0.2696                   | 0.04257                                   | 0.3506                                        |

*p-values are reported on the bottom of the respective cell; the correlation (rho) is reported on top significant p-values (<0.01) are bold-faced
Table 11. Comparison of children with EGA of <37 weeks or <33 weeks, with features of rib clefts

<table>
<thead>
<tr>
<th></th>
<th>Chi square Two-tailed p-value**</th>
<th>Fisher’s exact test Two-tailed p-value**</th>
</tr>
</thead>
<tbody>
<tr>
<td>10R+ sample (n=68)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>EGA &lt;37 weeks versus at least one microscopic cleft present</td>
<td>0.09692*</td>
<td>0.05543</td>
</tr>
<tr>
<td>10 R+ sample (n=68) EGA &lt;37 weeks versus &gt;2 microscopic clefts present</td>
<td>0.2174</td>
<td>0.2166</td>
</tr>
<tr>
<td>10R+ sample (n=68) EGA &lt;37 weeks versus at least one microscopic cleft &gt;1.00mm present</td>
<td>1.0000*</td>
<td>1.0000</td>
</tr>
<tr>
<td>10R+ sample (n=68) EGA &lt;33 weeks versus at least one microscopic cleft present</td>
<td>1.0000*</td>
<td>1.0000</td>
</tr>
<tr>
<td>10R+ sample (n=68) EGA &lt;33 weeks versus &gt;2 microscopic clefts present</td>
<td>0.2174</td>
<td>0.2166</td>
</tr>
<tr>
<td>10R+ sample (n=68) EGA &lt;33 weeks versus at least one microscopic cleft &gt;1.00mm present</td>
<td>1.0000*</td>
<td>1.0000</td>
</tr>
</tbody>
</table>

* Some cells have an expected count of <5/ **None of the p-values are significant (i.e., <0.01

The number of rib head clefts per child are plotted against the estimated gestational age at birth for each child for Groups 1 and 4, Subgroups A and D, SIDS and Natural deaths respectively (Figs. 60 and 61).

![Figure 60. Plot of number of rib head clefts per child versus estimated gestational age at birth of child for Group 1, Subgroups A and D (SIDS and other natural deaths).](image)

![Figure 61. Plot of number of rib head clefts per child versus estimated gestational age at birth of child for Group 4, Subgroups A and D (SIDS and other natural deaths).](image)
Results of analysis of effects of birth method and cardiopulmonary resuscitation

Comparison of the number of clefts, number of clefts greater than 1.00 mm in length, and number of clefts greater than 0.501 mm in length per child between vaginal and Cesarean delivery and between CPR and no CPR being performed as part of the terminal event for various groupings of children using the 10R+ sample via Wilcoxon Rank Sum test and Welch’s two sample t-test indicated a non-significant p-value of 0.03749 when comparing the mean number of clefts greater than 1.00 mm in length between children born vaginally versus those born by Cesarean section for the children in Group 3, Subgroups A, B, and D. Otherwise, no p-values were even less than 0.5. The corresponding Wilcoxon Rank Sum test assessing the distribution had a non-significant p-value of 0.1386 (Table 12).

In addition to Wilcoxon Rank Sum test and Welch’s two sample t-test analysis of the relation between delivery methods and CPR and various features of the microscopic clefts, Chi-square and Fisher’s exact tests were performed to compare the distribution of the number of infants with clefts versus without clefts, and the number of infants with and without a cleft of greater than 1.00 mm in length against those infants who were born vaginally or via Cesarean section. Also, comparisons of the distribution of children with acute clefts, CMLs with scant or no hemorrhage, and CMLs with cleft material between those receiving CPR and those not receiving CPR were performed. No p-values of <0.1 were associated with the distribution of children with various cleft features between vaginal and Cesarean section delivery, or between those who received CPR and those who did not (Table 13) These provide no statistical evidence that either birth method or CPR performed have any effect on the incidence of rib head clefts per child, or on the incidence of rib head clefts with a length of greater than 1.00 mm per child.
Table 12. Comparison of cleft features (e.g., length of greater than 1.00 mm) for various groupings of children with birth method and CPR

*the cleft features are per child; e.g., number of clefts per child

<table>
<thead>
<tr>
<th>Group 1: A-E</th>
<th>Vaginal vs. Cesarean</th>
<th>CPR vs. No CPR</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Wilcoxon Rank Sum</td>
<td>t-test</td>
</tr>
<tr>
<td></td>
<td>p-value</td>
<td>p-value</td>
</tr>
<tr>
<td>Vaginal</td>
<td>10R+</td>
<td></td>
</tr>
<tr>
<td># of clefts</td>
<td>.1918</td>
<td>3348</td>
</tr>
<tr>
<td># of clefts &gt;1mm</td>
<td>.4356</td>
<td>.1515</td>
</tr>
<tr>
<td># of clefts &gt;.501 mm</td>
<td>.4405</td>
<td>7535</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Group 1: A, B, D</th>
<th>Vaginal vs. Cesarean</th>
<th>CPR vs. No CPR</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Wilcoxon Rank Sum</td>
<td>t-test</td>
</tr>
<tr>
<td></td>
<td>p-value</td>
<td>p-value</td>
</tr>
<tr>
<td>Vaginal</td>
<td>10R+</td>
<td></td>
</tr>
<tr>
<td># of clefts</td>
<td>.3717</td>
<td>3849</td>
</tr>
<tr>
<td># of clefts &gt;1mm</td>
<td>.2225</td>
<td>.08178</td>
</tr>
<tr>
<td># of clefts &gt;.501 mm</td>
<td>.2500</td>
<td>3027</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Group 2: A, B, D</th>
<th>Vaginal vs. Cesarean</th>
<th>CPR vs. No CPR</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Wilcoxon Rank Sum</td>
<td>t-test</td>
</tr>
<tr>
<td></td>
<td>p-value</td>
<td>p-value</td>
</tr>
<tr>
<td>Vaginal</td>
<td>10R+</td>
<td></td>
</tr>
<tr>
<td># of clefts</td>
<td>.2702</td>
<td>3089</td>
</tr>
<tr>
<td># of clefts &gt;1mm</td>
<td>.4369</td>
<td>3727</td>
</tr>
<tr>
<td># of clefts &gt;.501 mm</td>
<td>.1987</td>
<td>2555</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Group 3: A, B, D</th>
<th>Vaginal vs. Cesarean</th>
<th>CPR vs. No CPR</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Wilcoxon Rank Sum</td>
<td>t-test</td>
</tr>
<tr>
<td></td>
<td>p-value</td>
<td>p-value</td>
</tr>
<tr>
<td>Vaginal</td>
<td>10R+</td>
<td></td>
</tr>
<tr>
<td># of clefts</td>
<td>.7385</td>
<td>9482</td>
</tr>
<tr>
<td># of clefts &gt;1mm</td>
<td>.1386</td>
<td>.03749</td>
</tr>
<tr>
<td># of clefts &gt;.501 mm</td>
<td>.5820</td>
<td>7304</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Group 4: A, B, D</th>
<th>Vaginal vs. Cesarean</th>
<th>CPR vs. No CPR</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Wilcoxon Rank Sum</td>
<td>t-test</td>
</tr>
<tr>
<td></td>
<td>p-value</td>
<td>p-value</td>
</tr>
<tr>
<td>Vaginal</td>
<td>10R+</td>
<td></td>
</tr>
<tr>
<td># of clefts</td>
<td>.5606</td>
<td>7661</td>
</tr>
<tr>
<td># of clefts &gt;1mm</td>
<td>.3244</td>
<td>3032</td>
</tr>
<tr>
<td># of clefts &gt;.501 mm</td>
<td>.4728</td>
<td>6276</td>
</tr>
</tbody>
</table>

no p-values are significant (i.e., <0.01)
Table 13. Comparison of distribution of various features of clefts between children with different birth methods, and for children with presence or absence of CPR at death

<table>
<thead>
<tr>
<th>Comparison</th>
<th>Chi-square Two-sided p-value</th>
<th>Fisher’s exact Two-sided p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Vaginal vs Cesarean; 10R sample (n=83) Number of infants with clefts vs Number of infants without</td>
<td>0.9234</td>
<td>0.7853</td>
</tr>
<tr>
<td>Vaginal vs Cesarean; 10R sample (n=83) Number of infants with clefts &gt;1.00 mm vs Number of infants without</td>
<td>0.2897</td>
<td>0.2967</td>
</tr>
<tr>
<td>CPR vs no-CPR; 10R sample (n=89) Number of infants with acute clefts vs Number of infants without</td>
<td>1.000*</td>
<td>1.000</td>
</tr>
<tr>
<td>CPR vs no-CPR; 10R sample (n=89) Number of infants with CML with no or scant hemorrhage vs Number of infants without</td>
<td>0.8810*</td>
<td>1.000</td>
</tr>
<tr>
<td>CPR vs no-CPR; 10R sample (n=89) Number of infants with CML with cleft material vs Number of infants without</td>
<td>0.9648*</td>
<td>1.0000</td>
</tr>
</tbody>
</table>

* some cells have an expected count of <5; no p-values are significant (i.e., <0.01)

Results of analysis of the distribution of clefts between the subgroups

Using Chi-square and Fisher’s exact test and Group 4 and both the 10R and the 10R+ samples separately, the distribution of infants with and without a cleft, with and without greater than two clefts, and with and without a cleft of greater than 1.00 mm in length was compared between the five Subgroups (A-E). When using the 10R and 10R+ samples, both the Chi-square analysis and Fisher’s exact test (four tests total) indicated not quite significant p-values of greater than 0.01 but less than 0.05 (all 0.01791 or less) for each test when comparing the distribution of the number of children with and without a cleft of greater than 1.00 mm in length across the five subgroups; however, no other comparison indicated a p-value of less than 0.05, which were all insignificant. The p-values for distribution of children with greater than two clefts between the five subgroups when using the 10R or 10R+ sample were both insignificant (Table 14).
Table 14. Comparison of various features of clefts between subgroups A-E in Group 4, using the 10R and 10R+ sample.

<table>
<thead>
<tr>
<th>Subgroup</th>
<th>Chi-square Two-tailed p-values</th>
<th>Fisher’s exact Two-tailed p-values</th>
</tr>
</thead>
<tbody>
<tr>
<td>Group 4, 10R sample A-E subgroups, presence vs absence of cleft</td>
<td>0.2984*</td>
<td>0.3327</td>
</tr>
<tr>
<td>Group 4, 10R sample A-E subgroups, presence vs absence of &gt;2 microscopic clefts</td>
<td>0.06608*</td>
<td>0.06707</td>
</tr>
<tr>
<td>Group 4, 10R sample A-E subgroups, presence vs absence of clefts &gt;1.00 mm in length</td>
<td>0.01791*</td>
<td>0.01335</td>
</tr>
<tr>
<td>Group 4, 10R+ sample A-E subgroups, presence vs absence of cleft</td>
<td>0.2398*</td>
<td>0.2552</td>
</tr>
<tr>
<td>Group 4, 10R+ sample A-E subgroups, presence vs absence of &gt;2 microscopic clefts</td>
<td>0.0668*</td>
<td>0.06487</td>
</tr>
<tr>
<td>Group 4, 10R+ sample A-E subgroups, presence vs absence of clefts &gt;1.00 mm in length</td>
<td>0.01268*</td>
<td>0.01085</td>
</tr>
</tbody>
</table>

*some cells have an expected count of <5; no p-values are significant (i.e., <0.01)

To compare the distribution of the number of clefts per child, number of clefts greater than 1.00 mm in length per child, and number of clefts greater than 0.501 mm in length per child across the five different subgroups representing SIDS, bed sharing, NAI and suspicious deaths, natural deaths (other than SIDS), and the miscellaneous category of other deaths for infants not fitting into one of the other four categories, a Kruskal-Wallis test using various combinations of subgroups (A-E and A-D), and all datasets (All, 6R, 10R, and 10R+) was performed. For all Groups, except Group 4, for some groupings, testing indicated a p-value of less than 0.05, but none less than 0.01 when comparing the distribution of number of clefts per child across the selected subgroups, and for comparisons using a cleft length of greater than 1.00 mm and greater than 0.501 mm, all Groups, including Group 4, had some p-values of <0.05 and even <0.01 when comparing the distribution across the selected subgroups (Table 15). The selection of the samples for analysis (e.g., 6R vs 10R+) and the grouping of the children based upon circumstances of death (e.g., Groups 1, 2, 3, or 4); therefore, played a role in the outcome (i.e.,
significant or insignificant p-value). See Table 15 for an indication of which datasets (i.e., rib numbers available per child, A1, 6R, 10R, and 10R+) and which Groupings (i.e., Group 1, 2, 3, or 4) affected the statistical results.

<table>
<thead>
<tr>
<th>Groups</th>
<th>Varying combinations of ribs</th>
<th>A-E All sample</th>
<th>A-D All sample</th>
<th>A-E 6R sample</th>
<th>A-D 6R sample</th>
<th>A-E 10R sample</th>
<th>A-D 10R sample</th>
<th>A-E 10R+ sample</th>
<th>A-D 10R sample</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td></td>
<td>0.0625</td>
<td>0.1646</td>
<td>0.1404</td>
<td>0.5862</td>
<td>0.04044</td>
<td>0.1541</td>
<td>0.02546</td>
<td>0.04156</td>
</tr>
<tr>
<td>2</td>
<td></td>
<td>0.08416</td>
<td>0.2287</td>
<td>0.1507</td>
<td>0.6255</td>
<td>0.05606</td>
<td>0.2240</td>
<td>0.03871</td>
<td>0.06754</td>
</tr>
<tr>
<td>3</td>
<td></td>
<td>0.0859</td>
<td>0.1351</td>
<td>0.1490</td>
<td>0.7519</td>
<td>0.0554</td>
<td>0.1686</td>
<td>0.03336</td>
<td>0.03559</td>
</tr>
<tr>
<td>4</td>
<td></td>
<td>0.115</td>
<td>0.1939</td>
<td>0.1599</td>
<td>0.7998</td>
<td>0.07643</td>
<td>0.2488</td>
<td>0.05051</td>
<td>0.05877</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Groups</th>
<th>Varying combinations of ribs</th>
<th>A-E All sample</th>
<th>A-D All sample</th>
<th>A-E 6R sample</th>
<th>A-D 6R sample</th>
<th>A-E 10R sample</th>
<th>A-D 10R sample</th>
<th>A-E 10R+ sample</th>
<th>A-D 10R sample</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td></td>
<td>0.007713</td>
<td>0.01476</td>
<td>0.04626</td>
<td>0.06216</td>
<td>0.005453</td>
<td>0.01502</td>
<td>0.002754</td>
<td>0.00864</td>
</tr>
<tr>
<td>2</td>
<td></td>
<td>0.01880</td>
<td>0.03648</td>
<td>0.0825</td>
<td>0.1106</td>
<td>0.01356</td>
<td>0.03698</td>
<td>0.007745</td>
<td>0.02465</td>
</tr>
<tr>
<td>3</td>
<td></td>
<td>0.006462</td>
<td>0.01502</td>
<td>0.03944</td>
<td>0.06846</td>
<td>0.004788</td>
<td>0.01502</td>
<td>0.002780</td>
<td>0.01189</td>
</tr>
<tr>
<td>4</td>
<td></td>
<td>0.01581</td>
<td>0.03697</td>
<td>0.07064</td>
<td>0.1193</td>
<td>0.01193</td>
<td>0.03698</td>
<td>0.007817</td>
<td>0.03308</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Groups</th>
<th>Varying combinations or ribs</th>
<th>A-E, 10R+ sample</th>
<th>A-D, 10R+ sample</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td></td>
<td>0.004336</td>
<td>0.01537</td>
</tr>
<tr>
<td>2</td>
<td></td>
<td>0.005372</td>
<td>0.01847</td>
</tr>
<tr>
<td>3</td>
<td></td>
<td>0.004762</td>
<td>0.009961</td>
</tr>
<tr>
<td>4</td>
<td></td>
<td>0.005897</td>
<td>0.01197</td>
</tr>
</tbody>
</table>

*significant p-values, i.e., <0.01, are indicated by bold face
Table 16. Wilcoxon Rank Sum Testing comparing individual groups (using 10R and 10R+ samples).

<table>
<thead>
<tr>
<th></th>
<th># of clefts per child</th>
<th># of clefts &gt;1mm per child</th>
<th># of clefts &gt;0.501 mm per child</th>
</tr>
</thead>
<tbody>
<tr>
<td>SIDS vs BedShare</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>1: 0.7575</td>
<td>2: 0.4064</td>
<td>1: 0.5767</td>
</tr>
<tr>
<td></td>
<td>2: 0.9120</td>
<td>2: 0.6330</td>
<td>2: 0.6714</td>
</tr>
<tr>
<td></td>
<td>3: 0.7545</td>
<td>3: 0.4064</td>
<td>3: 0.5767</td>
</tr>
<tr>
<td></td>
<td>4: 0.9120</td>
<td>4: 0.6330</td>
<td>4: 0.6714</td>
</tr>
<tr>
<td>SIDS vs BedShare (10R+ sample)</td>
<td>4: 0.8472</td>
<td>4: 0.6865</td>
<td>4: 0.9166</td>
</tr>
<tr>
<td>SIDS vs Susp</td>
<td>1: 0.08275</td>
<td>1: 0.04634</td>
<td>1: 0.07165</td>
</tr>
<tr>
<td></td>
<td>2: 0.1454</td>
<td>2: 0.1254</td>
<td>2: 0.09146</td>
</tr>
<tr>
<td></td>
<td>3: 0.08275</td>
<td>3: 0.04634</td>
<td>3: 0.07165</td>
</tr>
<tr>
<td></td>
<td>4: 0.1454</td>
<td>4: 0.1254</td>
<td>4: 0.09146</td>
</tr>
<tr>
<td>SIDS vs Susp (10R+ sample)</td>
<td>4: 0.02934</td>
<td>4: 0.06194</td>
<td>4: 0.02508</td>
</tr>
<tr>
<td>SIDS vs Nat</td>
<td>1: 0.3965</td>
<td>1: 0.1174</td>
<td>1: 0.2384</td>
</tr>
<tr>
<td></td>
<td>2: 0.3130</td>
<td>2: 0.07183</td>
<td>2: 0.1878</td>
</tr>
<tr>
<td></td>
<td>3: 0.4387</td>
<td>3: 0.0873</td>
<td>3: 0.1977</td>
</tr>
<tr>
<td></td>
<td>4: 0.3573</td>
<td>4: 0.0628</td>
<td>4: 0.1567</td>
</tr>
<tr>
<td>SIDS vs Nat (10R+ sample)</td>
<td>4: 0.4088</td>
<td>4: 0.1077</td>
<td>4: 0.1069</td>
</tr>
<tr>
<td>SIDS vs Others</td>
<td>1: 0.05431</td>
<td>1: 0.2534</td>
<td>1: 0.8063</td>
</tr>
<tr>
<td></td>
<td>2: 0.04216</td>
<td>2: 0.1749</td>
<td>2: 0.05616</td>
</tr>
<tr>
<td></td>
<td>3: 0.0901</td>
<td>3: 0.2503</td>
<td>3: 0.1264</td>
</tr>
<tr>
<td></td>
<td>4: 0.06729</td>
<td>4: 0.1552</td>
<td>4: 0.09024</td>
</tr>
<tr>
<td>SIDS vs Others (10R+ sample)</td>
<td>4: 0.1776</td>
<td>4: 0.1049</td>
<td>4: 0.1318</td>
</tr>
<tr>
<td>BedShare vs Susp</td>
<td>1: 0.1105</td>
<td>1: 0.1058</td>
<td>1: 0.08390</td>
</tr>
<tr>
<td></td>
<td>2: 0.1557</td>
<td>2: 0.1894</td>
<td>2: 0.09608</td>
</tr>
<tr>
<td></td>
<td>3: 0.1105</td>
<td>3: 0.1058</td>
<td>3: 0.08390</td>
</tr>
<tr>
<td></td>
<td>4: 0.1557</td>
<td>4: 0.1894</td>
<td>4: 0.09608</td>
</tr>
<tr>
<td>BedShare vs Susp (10R+ sample)</td>
<td>4: 0.02062</td>
<td>4: 0.0802</td>
<td>4: 0.01259</td>
</tr>
<tr>
<td>BedShare vs Nat</td>
<td>1: 0.2498</td>
<td>1: 0.01636</td>
<td>1: 0.06687</td>
</tr>
<tr>
<td></td>
<td>2: 0.2498</td>
<td>2: 0.01636</td>
<td>2: 0.06687</td>
</tr>
<tr>
<td></td>
<td>3: 0.3019</td>
<td>3: 0.02072</td>
<td>3: 0.05970</td>
</tr>
<tr>
<td></td>
<td>4: 0.3019</td>
<td>4: 0.02072</td>
<td>4: 0.05970</td>
</tr>
<tr>
<td>BedShare vs Nat (10R+ sample)</td>
<td>4: 0.4485</td>
<td>4: 0.04923</td>
<td>4: 0.06972</td>
</tr>
<tr>
<td>BedShare vs Other</td>
<td>1: 0.03054</td>
<td>1: 0.0610</td>
<td>1: 0.01404</td>
</tr>
<tr>
<td></td>
<td>2: 0.03054</td>
<td>2: 0.0610</td>
<td>2: 0.01404</td>
</tr>
<tr>
<td></td>
<td>3: 0.04526</td>
<td>3: 0.03939</td>
<td>3: 0.02272</td>
</tr>
<tr>
<td></td>
<td>4: 0.04526</td>
<td>4: 0.03939</td>
<td>4: 0.02272</td>
</tr>
<tr>
<td>BedShare vs Other (10R+ sample)</td>
<td>4: 0.2223</td>
<td>4: 0.03397</td>
<td>4: 0.07403</td>
</tr>
<tr>
<td>Nat vs Susp</td>
<td>1: 0.06283</td>
<td>1: 0.002440</td>
<td>1: 0.01422</td>
</tr>
<tr>
<td></td>
<td>2: 0.08974</td>
<td>2: 0.004449</td>
<td>2: 0.01911</td>
</tr>
<tr>
<td></td>
<td>3: 0.07004</td>
<td>3: 0.005778</td>
<td>3: 0.01593</td>
</tr>
<tr>
<td></td>
<td>4: 0.09533</td>
<td>4: 0.00855</td>
<td>4: 0.02114</td>
</tr>
<tr>
<td>Nat vs Susp (10R+ sample)</td>
<td>4: 0.04352</td>
<td>4: 0.01318</td>
<td>4: 0.01160</td>
</tr>
<tr>
<td>Nat vs Other</td>
<td>1: 0.2559</td>
<td>1: 0.8279</td>
<td>1: 0.6452</td>
</tr>
<tr>
<td></td>
<td>2: 0.2559</td>
<td>2: 0.8279</td>
<td>2: 0.6452</td>
</tr>
<tr>
<td></td>
<td>3: 0.4619</td>
<td>3: 0.2933</td>
<td>3: 1.000</td>
</tr>
<tr>
<td></td>
<td>4: 0.4619</td>
<td>4: 0.2933</td>
<td>4: 1.000</td>
</tr>
<tr>
<td>Nat vs Other (10R+ sample)</td>
<td>4: 0.9671</td>
<td>4: 0.5294</td>
<td>4: 0.5318</td>
</tr>
<tr>
<td>Susp vs Other</td>
<td>1: 0.01685</td>
<td>1: 0.01271</td>
<td>1: 0.004722</td>
</tr>
<tr>
<td></td>
<td>2: 0.02342</td>
<td>2: 0.02035</td>
<td>2: 0.007580</td>
</tr>
<tr>
<td></td>
<td>3: 0.02246</td>
<td>3: 0.00503</td>
<td>3: 0.005688</td>
</tr>
<tr>
<td></td>
<td>4: 0.03302</td>
<td>4: 0.009607</td>
<td>4: 0.008836</td>
</tr>
<tr>
<td>Susp vs Other (10R+ sample)</td>
<td>4: 0.02054</td>
<td>4: 0.004056</td>
<td>4: 0.005401</td>
</tr>
</tbody>
</table>
To analyze the difference in number of clefts per child, number of clefts that are greater than 1.00 mm in length per child, and the number of clefts that are greater than 0.501 mm in length per child, a Wilcoxon Rank Sum test was performed to compare the distribution between two subgroups. All combinations of the five general categories were made, using the 10R sample, and all five Subgroups from each of the four Groups each independently (Table 16). Note that no Bonferroni correction was applied. As there were 10 pairwise comparisons, if a Bonferroni correction had been applied, the resulting p-values would be 10 times greater than that listed in Table 16.

Testing of non-weighted versus weighted samples

As described in Materials and Methods, one difficulty encountered was the uneven distribution of the number of rib heads available for analysis between the different children in the sample population (with the extreme ends of the spectrum being three rib heads available for analysis in one child versus 20 rib heads available for analysis in another child). As the rib fractures are not felt to be independent events, weighting the statistical analyses seemed inappropriate; however, confirming this assumption was appropriate. As described above, non-parametric testing was favored over parametric testing as the distribution of number of clefts per child and number of clefts greater than 1.00 mm in length per child appeared to follow a Poisson distribution and not a normal distribution, and to be conservative in conclusions; so the Kruskal-Wallis test was performed to assess whether or not the different subgroups had an identical distribution for the number of clefts, number of clefts greater than 1.00 mm in length, and number of clefts greater than 0.501 mm in length per child. However, assessment of the utility
of ANOVA testing was appropriate so as to test the conclusion that the data was not normally distributed, as would be determined by the associated statistical methods to test the ANOVA assumptions of a normal distribution and homogeneity of variance.

Using the All sample and Group 1 and comparing the distribution of number of clefts per child between the five subgroups indicated a non-significant p-value of 0.022 for a non-weighted sample (with a Levene statistic p-value of 0.087), but a highly significant p-value of 0.000 when weighting the sample by the number of rib heads available for analysis per child (with a Levene statistic p-value of 0.000, which is highly significant, and indicates at least one of the ANOVA assumptions is unfulfilled).

Repeating the above analyses using the 10R sample produced comparable results (p-values of 0.026 and 0.199 respectively for non-weighted sample and p-values of 0.000 and 0.000 respectively for a weighted sample). Using the 10R+ sample and Group 4 and comparing the distribution of number of clefts per child between the five subgroups indicated a p-value of 0.027 for a non-weighted sample (with a Levene statistic p-value of 0.354), but a p-value of 0.000 when weighting the sample by the number of rib heads available for analysis per child (with a Levene statistic p-value of 0.000). Table 17 illustrates ANOVA results when comparing mean number of clefts per child across the five Subgroups (A-E), using Group 4, and through all four datasets, both weighting and not weighting of the analysis based upon the number of rib heads available for analysis per child.

The results are the same as above, with weighting routinely indicating a p-value of <0.000 for both the ANOVA analysis and the corresponding Levene statistic. Performance of the Kruskal-Wallis test using non-weighted and weighted samples and various combinations of
Groups, subgroups, and datasets as well as performance of the Mann-Whitney using the All sampled weighting by the number of rib heads per child available for analysis, indicated p-values of <0.0009 for almost every comparison when using weighed samples (Tables 18 and 19).

Table 17. ANOVA analysis of distribution of number of clefts per child between the various subgroups using all datasets and both weighted and unweighted samples

<table>
<thead>
<tr>
<th>Number of clefts per child</th>
<th>p-value</th>
<th>Levene statistic</th>
</tr>
</thead>
<tbody>
<tr>
<td>All ribs sample, Group 4, non-weighted</td>
<td>0.041</td>
<td>0.047</td>
</tr>
<tr>
<td>All ribs sample, Group 4, weighted by number of rib heads</td>
<td><strong>0.000</strong></td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>6R sample, Group 4, non-weighted</td>
<td>0.348</td>
<td>0.707</td>
</tr>
<tr>
<td>6R sample, Group 4, weighted by number of rib heads</td>
<td><strong>0.000</strong></td>
<td>0.001</td>
</tr>
<tr>
<td>10R sample, Group 4, non-weighted</td>
<td>0.059</td>
<td>0.199</td>
</tr>
<tr>
<td>10R sample, Group 4, weighted by number of rib heads</td>
<td><strong>0.000</strong></td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>10R+ sample, Group 4, non-weighted</td>
<td>0.027</td>
<td>0.354</td>
</tr>
<tr>
<td>10R+ sample, Group 4, weighted by number of rib heads</td>
<td><strong>0.000</strong></td>
<td><strong>0.000</strong></td>
</tr>
</tbody>
</table>

*significant p-values (<0.01) are indicated by bold-face

Table 18. Results of Kruskal-Wallis testing using both weighted and unweighted samples

<table>
<thead>
<tr>
<th>Number of clefts per child</th>
<th>p-value Non-weighted</th>
<th>p-value Weighted by number of rib heads</th>
</tr>
</thead>
<tbody>
<tr>
<td>Group 1, A-E (n=90), All sample</td>
<td>0.063</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 1, A-D (n=79), All sample</td>
<td>0.165</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 2, A-E (n=90), All sample</td>
<td>0.084</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 2, A-E (n=79), All sample</td>
<td>0.229</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 3, A-E (n=90), All sample</td>
<td>0.086</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 3, A-D (n=79), All sample</td>
<td>0.135</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 4, A-E (n=90), All sample</td>
<td>0.115</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 4, A-D (n=79), All sample</td>
<td>0.194</td>
<td><strong>0.000</strong></td>
</tr>
</tbody>
</table>

Group 1, A-E (n=90), 10R sample

<table>
<thead>
<tr>
<th>Number of clefts per child</th>
<th>p-value Non-weighted</th>
<th>p-value Weighted by number of rib heads</th>
</tr>
</thead>
<tbody>
<tr>
<td>Group 1, A-E (n=90), 10R sample</td>
<td>0.040</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 1, A-D (n=79), 10R sample</td>
<td>0.154</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 2, A-E (n=90), 10R sample</td>
<td>0.056</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 2, A-D (n=79), 10R sample</td>
<td>0.224</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 3, A-E (n=90), 10R sample</td>
<td>0.055</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 3, A-D (n=79), 10R sample</td>
<td>0.169</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 4, A-E (n=90), 10R sample</td>
<td>0.076</td>
<td><strong>0.000</strong></td>
</tr>
<tr>
<td>Group 4, A-D (n=79), 10R sample</td>
<td>0.249</td>
<td><strong>0.000</strong></td>
</tr>
</tbody>
</table>

*significant p-values (<0.01) are indicated by bold-face
<table>
<thead>
<tr>
<th>Table 19: Results of weighted Mann-Whitney test</th>
</tr>
</thead>
<tbody>
<tr>
<td>p-values are indicated significant p-values are bold faced</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>SIDS vs Bed sharing</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>SIDS vs Suspicious</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>SIDS vs Natural</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>SIDS vs Other</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Bed sharing vs Suspicious</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Bed sharing vs Natural</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Bed sharing vs Other</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Suspicious vs Natural</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Suspicious vs Other</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Natural vs Other</td>
</tr>
<tr>
<td></td>
</tr>
</tbody>
</table>

Results of data analysis of microscopic clefts–final

As the preliminary analyses were performed using various combinations of datasets (All, 6R, 10R, and 10R+) and groups (Groups 1-4), and subgroups (A-E), the above reported results are complex; however, they do serve to illustrate the variation in results when various numbers of ribs per child are analyzed (e.g., 6 versus 10) and how the subjective assessment of cause and manner of death can affect the data analyses. To provide for a more straight-forward analysis of the data, the final data analysis will be performed using the 10R+ sample, where each child had between 8-11 rib heads available for analysis, and Groups 3 and 4, which both had a subgroup of known natural deaths (e.g., bronchopneumonia), and included the undetermined natural deaths in the other category, and varied by inclusion of the suspicious SIDS death in the suspicious subgroup (Group 3) or in the SIDS subgroup (Group 4), statistical methods as described above.
were conducted. As described above, the purpose was two fold: 1) for simplification, as using four different samples (All, 6R, 10R, and 10R+) and four different groupings of circumstances of death (Groups 1-4), resulted in up to 16 different pairings for statistical analysis for each test conducted in the preliminary analysis, and complex results and 2) to compare the inclusion of a suspicious death in either the SIDS category or the suspicious category and the changes it would create in analysis. Also, these final results will include analysis of socio-economic factors such as the effect of married versus unmarried mothers and the presence of biological fathers versus boyfriends.

*General characteristics*

In both Group 3 and Group 4, there were 77 children, 42 males and 35 females (Table 20). Division into ancestry included 64 whites, 12 Native Americans, and one Hispanic. Forty-eight of the children had married mothers, 27 had unmarried mothers, and for two children, the marital status of the mother was unavailable. Regarding the dominant male figure in the child’s life, 58 had a biological father, 11 had a boyfriend, five had no male figure, and for three children, this situation was unknown. For 18 children, their living quarters were apparently clean, and for nine children, their living quarters were apparently messy; however, for the remainder of the children, this designation was not available. Of these children, 54 were delivered vaginally, 18 via Cesarean section, and, for five, the birth method was unavailable. In addition, 72 received CPR, four did not, and for one, the presence or absence of CPR attempts was unavailable. The age at death ranged from 0.03 to 18 months, with a mean of 4.103 months and a median of 3.000 months (95% CI of 2.187-3.000). The estimated gestational age ranged
from 28 to 41 weeks, with a mean of 38.221 weeks, and a median of 39 weeks (95% CI of 38.000-39.000)

In both Group 3 and Group 4, each child had between 0 and 10 clefts (Table 20). The mean number of clefts per child was 2.494, with a median of 2.000 (95% CI of 1.000-3.000). Each child had between 0 and 5 clefts that had a length of greater than 1.00 mm. The mean number of clefts with a length of greater than 1.00 mm per child was 0.532, with a median of 0 (95% CI of 0.000-0.000). Each child had between 0 and 8 clefts that had a length of greater than 0.501 mm. The mean number of clefts with a length of greater than 0.501 mm per child was 1.377, with a median of 1 (95% CI of 0.000-2.000).

Comparison of cleft characteristics between five circumstances of death

Although the mean number of clefts per child, mean number of clefts with a length of greater than 1.00 mm in length per child, and mean number of clefts with a length of greater than 0.501 mm in length per child varied between Groups 3 and 4 for the various categories of circumstances of death (Tables 21-23), the median did not, although the 95% CI for the calculated median using the sign test did vary when comparing the SIDS subgroup and Suspicious subgroup.
Table 20. General characteristics of final data analysis sample

<table>
<thead>
<tr>
<th></th>
<th>Group 3</th>
<th>Group 4</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of children in sample</td>
<td>77</td>
<td>77</td>
</tr>
<tr>
<td>Number delivered vaginally</td>
<td>54</td>
<td>54</td>
</tr>
<tr>
<td>Number delivered via Cesarean section</td>
<td>18</td>
<td>18</td>
</tr>
<tr>
<td>Delivery method unknown</td>
<td>5</td>
<td>5</td>
</tr>
<tr>
<td>Number who received CPR</td>
<td>72</td>
<td>72</td>
</tr>
<tr>
<td>Number who did not receive CPR</td>
<td>4</td>
<td>4</td>
</tr>
<tr>
<td>Status of CPR use unknown</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Age at death</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Range</td>
<td>0.3-18 months</td>
<td>0.3-18 months</td>
</tr>
<tr>
<td>Mean</td>
<td>4.103 months</td>
<td>4.103 months</td>
</tr>
<tr>
<td>Median</td>
<td>3 months</td>
<td>3 months</td>
</tr>
<tr>
<td>Median with sign test</td>
<td>3.000 months</td>
<td>3.000 months</td>
</tr>
<tr>
<td>Estimated gestational age at birth</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Range</td>
<td>28-41 weeks</td>
<td>28-41 weeks</td>
</tr>
<tr>
<td>Mean</td>
<td>38.221 weeks</td>
<td>38.221 weeks</td>
</tr>
<tr>
<td>Median</td>
<td>39 weeks</td>
<td>39 weeks</td>
</tr>
<tr>
<td>Median with sign test</td>
<td>39 weeks</td>
<td>39 weeks</td>
</tr>
<tr>
<td>Number of clefts per child</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Range</td>
<td>0-10</td>
<td>0-10</td>
</tr>
<tr>
<td>Mean</td>
<td>2.494</td>
<td>2.494</td>
</tr>
<tr>
<td>Median</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Median with sign test</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Number of clefts greater than 1.00 mm in length per child</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Range</td>
<td>0-5</td>
<td>0-5</td>
</tr>
<tr>
<td>Mean</td>
<td>0.532</td>
<td>0.532</td>
</tr>
<tr>
<td>Median</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Median with sign test</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Number of clefts greater than 0.501 mm in length per child</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Range</td>
<td>0-8</td>
<td>0-8</td>
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<tr>
<td>Mean</td>
<td>1.377</td>
<td>1.377</td>
</tr>
<tr>
<td>Median</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Median with sign test</td>
<td>1</td>
<td>1</td>
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</tbody>
</table>

186
Table 21. Comparison of number of clefts per child between five circumstances of death

<table>
<thead>
<tr>
<th></th>
<th>Group 3</th>
<th>Group 4</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>SIDS deaths</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Number of children</td>
<td>18</td>
<td>19</td>
</tr>
<tr>
<td><strong>Number of clefts per child</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Mean</td>
<td>2.222</td>
<td>2.316</td>
</tr>
<tr>
<td>• Median</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>• Median with Sign test</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>95% CI = 1 to 3</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Bed-Sharing deaths</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Number of children</td>
<td>27</td>
<td>27</td>
</tr>
<tr>
<td><strong>Number of clefts per child</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Mean</td>
<td>2.259</td>
<td>2.259</td>
</tr>
<tr>
<td>• Median</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>• Median with Sign test</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>95% CI = 1 to 3</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Suspicious and NAI deaths</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Number of children</td>
<td>12</td>
<td>11</td>
</tr>
<tr>
<td><strong>Number of clefts per child</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Mean</td>
<td>4.5</td>
<td>4.545</td>
</tr>
<tr>
<td>• Median</td>
<td>4</td>
<td>4</td>
</tr>
<tr>
<td>• Median with Sign test</td>
<td>4</td>
<td>4</td>
</tr>
<tr>
<td>95% CI = 3.000 to 6.894</td>
<td></td>
<td></td>
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<tr>
<td><strong>Known natural deaths</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Number of children</td>
<td>7</td>
<td>7</td>
</tr>
<tr>
<td><strong>Number of clefts per child</strong></td>
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<tr>
<td>• Mean</td>
<td>1.714</td>
<td>1.714</td>
</tr>
<tr>
<td>• Median</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>• Median with Sign test</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>95% CI = 0.000 to 4.371</td>
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<td></td>
</tr>
<tr>
<td><strong>Other</strong></td>
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<tr>
<td>• Number of children</td>
<td>13</td>
<td>13</td>
</tr>
<tr>
<td><strong>Number of clefts per child</strong></td>
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<td></td>
</tr>
<tr>
<td>• Mean</td>
<td>1.923</td>
<td>1.923</td>
</tr>
<tr>
<td>• Median</td>
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<tr>
<td>• Median with Sign test</td>
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<td>1</td>
</tr>
<tr>
<td>95% CI = 0.000 to 2.606</td>
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</table>
Table 22. Comparison of number of clefts with length greater than 1.00 mm per child between five circumstances of death

<table>
<thead>
<tr>
<th>Group</th>
<th>Group 3</th>
<th>Group 4</th>
</tr>
</thead>
<tbody>
<tr>
<td>SIDS deaths</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of children</td>
<td>18</td>
<td>19</td>
</tr>
<tr>
<td>Number of clefts per child greater than 1.00 mm in length</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean</td>
<td>0.389</td>
<td>0.474</td>
</tr>
<tr>
<td>Median</td>
<td>0</td>
<td>0</td>
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<tr>
<td>Median with Sign test</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>95% CI = 0.000 to 0.708</td>
<td>0</td>
<td>95% CI = 0 to 1</td>
</tr>
<tr>
<td>Bed-Sharing deaths</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of children</td>
<td>27</td>
<td>27</td>
</tr>
<tr>
<td>Number of clefts per child greater than 1.00 mm in length</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean</td>
<td>0.519</td>
<td>0.519</td>
</tr>
<tr>
<td>Median</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Median with Sign test</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>95% CI = 0 to 1</td>
<td>0</td>
<td>95% CI = 0 to 1</td>
</tr>
<tr>
<td>Suspicious and NAI deaths</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of children</td>
<td>12</td>
<td>11</td>
</tr>
<tr>
<td>Number of clefts per child greater than 1.00 mm in length</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean</td>
<td>1.583</td>
<td>1.545</td>
</tr>
<tr>
<td>Median</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Median with Sign test</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>95% CI = 0.000 to 2.894</td>
<td>1</td>
<td>95% CI = 0.000 to 3.287</td>
</tr>
<tr>
<td>Known natural deaths</td>
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<tr>
<td>Number of children</td>
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<tr>
<td>Number of clefts per child greater than 1.00 mm in length</td>
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<td></td>
</tr>
<tr>
<td>Mean</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Median</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Median with Sign test</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>95% CI = 0 to 0</td>
<td>0</td>
<td>95% CI = 0 to 0</td>
</tr>
<tr>
<td>Other</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of children</td>
<td>13</td>
<td>13</td>
</tr>
<tr>
<td>Number of clefts per child greater than 1.00 mm in length</td>
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</tr>
<tr>
<td>Mean</td>
<td>0.077</td>
<td>0.077</td>
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<tr>
<td>Median</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Median with Sign test</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>95% CI = 0 to 0</td>
<td>0</td>
<td>95% CI = 0 to 0</td>
</tr>
</tbody>
</table>
Table 23. Comparison of number of clefts with length greater than 0.501 mm in length per child between five circumstances of death

<table>
<thead>
<tr>
<th></th>
<th>Group 3</th>
<th>Group 4</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>SIDS deaths</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of children</td>
<td>18</td>
<td>19</td>
</tr>
<tr>
<td>Number of clefts per child greater than 0.501 mm in length</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Mean</td>
<td>1.278</td>
<td>1.316</td>
</tr>
<tr>
<td>• Median</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>• Median with Sign test</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>95% CI = 0 to 2</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Bed-Sharing deaths</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of children</td>
<td>27</td>
<td>27</td>
</tr>
<tr>
<td>Number of clefts per child greater than 0.501 mm in length</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Mean</td>
<td>1.259</td>
<td>1.259</td>
</tr>
<tr>
<td>• Median</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>• Median with Sign test</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>95% CI = 0 to 2</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Suspicious and NAI deaths</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of children</td>
<td>12</td>
<td>11</td>
</tr>
<tr>
<td>Number of clefts per child greater than 0.501 mm in length</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Mean</td>
<td>3.083</td>
<td>3.182</td>
</tr>
<tr>
<td>• Median</td>
<td>2.5</td>
<td>3</td>
</tr>
<tr>
<td>• Median with Sign test</td>
<td>2.5</td>
<td>3</td>
</tr>
<tr>
<td>95% CI = 2 to 4</td>
<td></td>
<td></td>
</tr>
<tr>
<td>95% CI = 1.425 to 4.575</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Known natural deaths</strong></td>
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<td></td>
</tr>
<tr>
<td>Number of children</td>
<td>7</td>
<td>7</td>
</tr>
<tr>
<td>Number of clefts per child greater than 0.501 mm in length</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Mean</td>
<td>0.429</td>
<td>0.429</td>
</tr>
<tr>
<td>• Median</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>• Median with Sign test</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>95% CI = 0.000 to 2.057</td>
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<td></td>
</tr>
<tr>
<td>95% CI = 0.000 to 2.057</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Other</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of children</td>
<td>13</td>
<td>13</td>
</tr>
<tr>
<td>Number of clefts per child greater than 0.501 mm in length</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Mean</td>
<td>0.692</td>
<td>0.692</td>
</tr>
<tr>
<td>• Median</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>• Median with Sign test</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>95% CI = 0 to 1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>95% CI = 0 to 1</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
To better visualize differences in mean number of clefts per child in each of the five circumstances of death categories and mean number of clefts per child with a length of greater than 1.00 mm, boxplots are provided (Figs. 62-63).

A Kruskal-Wallis test and pairwise Wilcoxon Rank Sum test (with Bonferroni correction) comparing distribution of number of clefts per child, number of clefts with a length of greater than 1.00 mm per child, and number of clefts with a length of greater than 0.501 mm per child between the subgroups of circumstances of death indicated only three significant differences (Table 24). In Group 3 and 4, the difference between the distribution of number of clefts greater
than 1.00 mm in length for suspicious deaths and other deaths had non-significant p-values of less than 0.05 (0.023 and 0.041 respectively) and, in Group 3, the difference between the distribution of number of clefts greater than 0.501 mm in length for suspicious deaths and other deaths had a non-significant p-value of 0.039 (of note, Group 4 had a p-value of 0.054 for the same pairing).

Table 24. Comparison of number of clefts per child and cleft size to circumstances of death by Kruskal-Wallis (first line for each feature tested) and pairwise Wilcoxon (each subsequent pairing)

| Kruskal-Wallis and pairwise Wilcoxon p-values are indicated; significant p-values are bold-faced |
|-----------------------------------------------|----------------|
| Group 3                                      | Group 4        |
| Number of clefts per child                   | 0.0336         | 0.0505         |
| • Suspicious and Bed-sharing                 | 0.14           | 0.21           |
| • Suspicious and known natural               | 0.36           | 0.44           |
| • Suspicious and other                       | 0.15           | 0.21           |
| • Suspicious and SIDS                        | 0.15           | 0.29           |
| Number of clefts greater than 1.00 mm in length per child | **0.00278** | **0.00782** |
| • Other and Bed-sharing                      | 0.340          | 0.340          |
| • Suspicious and known natural               | 0.095          | 0.132          |
| • Suspicious and other                       | 0.023          | 0.041          |
| • Suspicious and SIDS                        | 0.211          | 0.619          |
| Number of clefts greater than 0.501 mm in length per child | **0.00476** | **0.00590** |
| • Suspicious and Bed-sharing                 | 0.113          | 0.126          |
| • Suspicious and known natural               | 0.103          | 0.116          |
| • Suspicious and other                       | 0.039          | 0.054          |
| • Suspicious and SIDS                        | 0.220          | 0.251          |

The Bonferroni correction was applied to p-values determined by pairwise Wilcoxon Rank Sum test.
An analysis of variance, comparing the mean number of clefts per child, mean number of clefts that are greater than 1.00 mm in length per child, and mean number of clefts that are greater than 0.501 mm in length per child between the five circumstances of death (Table 25), indicates problems with normality (significant Shapiro-Wilk tests) and homogeneity of variance (significant Levene and Bartlett tests).

Comparison of cleft characteristics between suspicious and non-suspicious deaths

Although the above testing compared five subgroups, the most important distinction was between those deaths which were 1) outright due to non-accidental injury (NAI), or had autopsy or investigative features suspicious for NAI, and are grouped together under the category of “suspicious”, or 2) had no suspicious findings during the investigation or at autopsy, and are grouped together under the category of “non-suspicious”. In Group 3, there were 12 suspicious deaths and 65 non-suspicious deaths, and in Group 4, there were 11 suspicious deaths and 66 non-suspicious deaths (Table 26).
Table 25. Analysis of variance testing for differences in cleft characteristics between circumstances of death, using Group 3 and all categories of death

<table>
<thead>
<tr>
<th>Groups</th>
<th>Difference</th>
<th>95% CI</th>
<th>p-value**</th>
</tr>
</thead>
<tbody>
<tr>
<td>Susp-BedShare</td>
<td>2.241</td>
<td>0.108 to 4.374</td>
<td>0.0347</td>
</tr>
<tr>
<td>Susp-Natural</td>
<td>2.786</td>
<td>-0.138 to 5.710</td>
<td>0.0693</td>
</tr>
<tr>
<td>Susp-Other</td>
<td>2.577</td>
<td>0.116 to 5.038</td>
<td>0.0356</td>
</tr>
<tr>
<td>Susp-SIDS</td>
<td>2.278</td>
<td>-0.0136 to 4.569</td>
<td>0.0521</td>
</tr>
</tbody>
</table>

Results of Tukey HSD

Levene test* p-value of 0.8152  Bartlett test p-value of 0.1467  Shapiro-Wilk test* p-value of 0.0003613

Results of Tukey HSD

<table>
<thead>
<tr>
<th>Groups</th>
<th>Difference</th>
<th>95% CI</th>
<th>p-value**</th>
</tr>
</thead>
<tbody>
<tr>
<td>Susp-BedShare</td>
<td>1.0648</td>
<td>0.233 to 1.897</td>
<td><strong>0.00541</strong></td>
</tr>
<tr>
<td>Susp-Natural</td>
<td>1.583</td>
<td>0.443 to 2.724</td>
<td><strong>0.00204</strong></td>
</tr>
<tr>
<td>Susp-Other</td>
<td>1.506</td>
<td>0.547 to 2.466</td>
<td><strong>0.000357</strong></td>
</tr>
<tr>
<td>Susp-SIDS</td>
<td>1.194</td>
<td>0.301 to 2.0879</td>
<td><strong>0.00327</strong></td>
</tr>
</tbody>
</table>

Results of Tukey HSD

Levene test* p-value of 0.0009794  Bartlett test p-value of 0.00000  Shapiro-Wilk test* p-value of 0.00000

Results of Tukey HSD

<table>
<thead>
<tr>
<th>Groups</th>
<th>Difference</th>
<th>95% CI</th>
<th>p-value**</th>
</tr>
</thead>
<tbody>
<tr>
<td>Susp-BedShare</td>
<td>1.824</td>
<td>0.332 to 3.317</td>
<td><strong>0.00890</strong></td>
</tr>
<tr>
<td>Susp-Natural</td>
<td>2.655</td>
<td>0.609 to 4.701</td>
<td><strong>0.00464</strong></td>
</tr>
<tr>
<td>Susp-Other</td>
<td>2.391</td>
<td>0.669 to 4.113</td>
<td><strong>0.00204</strong></td>
</tr>
<tr>
<td>Susp-SIDS</td>
<td>1.806</td>
<td>0.202 to 3.409</td>
<td>0.0195</td>
</tr>
</tbody>
</table>

*Square root transformation of the data did not correct the Shapiro-Wilk or Levene tests.

**Significant p-values are bold faced
<table>
<thead>
<tr>
<th>Table 26. Cleft characteristics of suspicious versus non-suspicious deaths</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Number of children</strong></td>
</tr>
<tr>
<td>• Group 3</td>
</tr>
<tr>
<td>• Group 4</td>
</tr>
<tr>
<td><strong>Mean number of clefts per child</strong></td>
</tr>
<tr>
<td>• Group 3</td>
</tr>
<tr>
<td>• Group 4</td>
</tr>
<tr>
<td><strong>Median number of clefts per child</strong></td>
</tr>
<tr>
<td>• Group 3</td>
</tr>
<tr>
<td>• Group 4</td>
</tr>
<tr>
<td><strong>Median (with sign test) number of clefts per child</strong></td>
</tr>
<tr>
<td>• Group 3</td>
</tr>
<tr>
<td>95% CI = 3.000 to 6.894</td>
</tr>
<tr>
<td>• Group 4</td>
</tr>
<tr>
<td>95% CI = 2.425 to 7.287</td>
</tr>
<tr>
<td><strong>Mean number of clefts per child greater than 1.00 mm in length</strong></td>
</tr>
<tr>
<td>• Group 3</td>
</tr>
<tr>
<td>• Group 4</td>
</tr>
<tr>
<td><strong>Median (with sign test) number of clefts per child greater than 1.00 mm in length</strong></td>
</tr>
<tr>
<td>• Group 3</td>
</tr>
<tr>
<td>95% CI = 0.000 to 2.894</td>
</tr>
<tr>
<td>• Group 4</td>
</tr>
<tr>
<td>95% CI = 0.000 to 3.287</td>
</tr>
<tr>
<td><strong>Median (with sign test) number of clefts per child greater than 0.501 mm in length</strong></td>
</tr>
<tr>
<td>• Group 3</td>
</tr>
<tr>
<td>95% CI = 2 to 4</td>
</tr>
<tr>
<td>• Group 4</td>
</tr>
<tr>
<td>95% CI = 1.425 to 4.575</td>
</tr>
</tbody>
</table>

Histograms indicating the frequency of number of clefts per child for suspicious and non-suspicious deaths, and the frequency of the number of clefts greater than 1.00 mm in length per child for suspicious and non-suspicious deaths are provided. As the difference between Group 3 and Group 4 is one child, only Group 3 cases are used for the purpose of constructing the histograms (Figs. 64-69).
Figure 64. Histogram of number of clefts per child in Group 3 suspicious deaths.

Figure 65. Histogram of number of clefts per child in Group 3 non-suspicious deaths.

Figure 66. Histogram of number of clefts per child with a length of greater than 1.00 mm in Group 3 suspicious deaths.

Figure 67. Histogram of number of clefts per child with a length of greater than 1.00 mm in Group 3 non-suspicious deaths.
The mean number of clefts per child in the suspicious deaths was 4.5 in Group 3 and 4.545 in Group 4, and in the non-suspicious deaths was 2.123 and 2.152 in Group 3 and 4 respectively. The median number of clefts per child was 4 in the suspicious deaths and 2 in the non-suspicious deaths in both Group 3 and 4. With the Sign test, the median number of clefts per child was 4 in the suspicious group (with a 95% CI of 3.000 to 6.894) in Group 3 and 4 in the suspicious group (with a 95% CI of 2.425 to 7.287) in Group 4. However, in the non-suspicious group, the median was 2 in both Group 3 and 4, with 95% CI of 1.000 to 2.897, and 1 to 3 respectively. The mean number of clefts with a length of greater than 1.00 mm per child in the suspicious deaths was 1.583 in Group 3 and 1.545 in Group 4, and in the non-suspicious deaths was 0.338 and 0.364 in Group 3 and 4 respectively. The median number of clefts with a length of greater than 1.00 mm per child was 1 in the suspicious deaths and 0 in the non-suspicious deaths in both Group 3 and 4. With the sign test, the median number of clefts with a length of greater than 1.00 mm per child was 1 in the suspicious group (with a 95% CI of 0.000 to 2.894)
in Group 3 and 1 in the suspicious group (with a 95% CI of 0.000 to 3.287) in Group 4. However, in the non-suspicious group, the median was 0 in Group 3 and 0.5 in Group 4, with 95% CI of 0 to 1 in both. Using Wilcoxon Rank Sum testing to assess the difference in distribution of number of clefts per child, number of clefts greater than 1.00 mm in length per child, and number of clefts greater than 0.501 mm in length per child between suspicious and non-suspicious deaths revealed significant p-values in all three categories, in both Groups 3 and 4 (Table 27).

**Table 27. Comparison of number of clefts per child and cleft size to suspicious versus non-suspicious deaths**

<table>
<thead>
<tr>
<th></th>
<th>Group 3</th>
<th>Group 4</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Wilcoxon Rank Sum test</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of clefts per child</td>
<td>p-value: 0.00326</td>
<td>p-value: 0.00658</td>
</tr>
<tr>
<td></td>
<td>Pseudomedian = 2.000</td>
<td>Pseudomedian = 2.000</td>
</tr>
<tr>
<td></td>
<td>95% CI = 1.000 to 4.000</td>
<td>95% CI = 1.000 to 4.000</td>
</tr>
<tr>
<td>Number of clefts greater than 1.00 mm in length per child</td>
<td>p-value: 0.00145</td>
<td>p-value: 0.00620</td>
</tr>
<tr>
<td></td>
<td>Pseudomedian = 1.000</td>
<td>Pseudomedian = 1.000</td>
</tr>
<tr>
<td></td>
<td>95% CI = 0.000 to 2.000</td>
<td>95% CI = 0.000 to 1.000</td>
</tr>
<tr>
<td>Number of clefts greater than 0.501 mm in length per child</td>
<td>p-value: 0.00145</td>
<td>p-value: 0.00214</td>
</tr>
<tr>
<td></td>
<td>Pseudomedian = 2.000</td>
<td>Pseudomedian = 2.000</td>
</tr>
<tr>
<td></td>
<td>95% CI = 1.000 to 3.000</td>
<td>95% CI = 1.000 to 3.000</td>
</tr>
</tbody>
</table>

*Significant p-values (<0.01) are bold-faced

**Poisson and logistic regression**

As the distribution of number of clefts per child and number of clefts greater than 1.00 mm in length per child follow a Poisson distribution, a Poisson regression model was developed, with the dependent variable being the number of clefts per child, and the explanatory variables being whether or not the death was suspicious or non-suspicious, age at death of child, delivery method, presence or absence of CPR, and estimated gestational age at delivery, and returned a significant p-value for both the suspicious or non-suspicious death and age at death coefficients.
(0.00229 and 0.00419 respectively), but none of the other variables. The residual deviance of the model was 107.20 on 59 degrees of freedom, and the AIC (Akaike’s Information Criterion) was 270.13. A chi-square test of deviance and residual degrees of freedom indicated a p-value of 0.0001267, indicating a lack of fit. A second Poisson regression model was developed, with the dependent variable being the number of clefts per child greater than 1.00 mm in length, and the explanatory variables being suspicious or non-suspicious death, age at death of child, delivery method, presence or absence of CPR, and estimated gestational age at delivery, and returned a significant p-value only for the suspicious or non-suspicious death coefficient (0.000005), and none others. The residual deviance of the model was 67.774 with 59 degrees of freedom, and the AIC was 135.81. A chi-square test of deviance and residual degrees of freedom indicated a p-value of 0.2028, supporting goodness of fit. A logistic regression model was developed, with the dependent variable being whether or not the death is suspicious, and the explanatory variable being the number of clefts greater than 1.00 mm per child. A calculation of the odds ratio indicated a 3.051 increased risk of a suspicious death for every cleft that was 1.00 mm in length, which was present; however, analysis of the goodness of fit indicated problems.

Analysis of socio-economic factors

A Wilcoxon Rank Sum test was performed to analyze the difference in distribution of the number of clefts per child, number of clefts per child greater than 1.00 mm in length, and number of clefts per child greater than 0.501 mm in length, between males and females, whites and Native Americans, married and unmarried mothers, boyfriends and biological fathers, and
positive or negative history of drug use. No statistically significant differences were identified (i.e., all p-values were above 0.05). The results are aggregated in Table 28.

<table>
<thead>
<tr>
<th>Groups</th>
<th>Number of clefts</th>
<th>Number of clefts with length &gt; 1.00 mm</th>
<th>Number of clefts with length &gt; 0.501 mm</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male and Female</td>
<td>p-value: 0.7674</td>
<td>p-value: 0.8577</td>
<td>p-value: 1.000</td>
</tr>
<tr>
<td></td>
<td>95% CI: -1.000 to 1.000</td>
<td>95% CI: -0.000027 to 0.000050</td>
<td>95% CI: -0.000049 to 0.000055</td>
</tr>
<tr>
<td>White and Native American</td>
<td>p-value: 0.9136</td>
<td>p-value: 0.8705</td>
<td>p-value: 0.3141</td>
</tr>
<tr>
<td></td>
<td>95% CI: -1.000 to 1.000</td>
<td>95% CI: -0.0000215 to 0.0000478</td>
<td>95% CI: -0.0000543 to 1.000</td>
</tr>
<tr>
<td>Married and Unmarried</td>
<td>p-value: 0.5088</td>
<td>p-value: 0.6431</td>
<td>p-value: 0.8845</td>
</tr>
<tr>
<td></td>
<td>95% CI: -1.000 to 1.000</td>
<td>95% CI: -0.0000288 to 0.0000811</td>
<td>95% CI: -0.100 to 0.000020</td>
</tr>
<tr>
<td>Biological father and Boyfriend</td>
<td>p-value: 0.881</td>
<td>p-value: 0.8161</td>
<td>p-value: 0.8899</td>
</tr>
<tr>
<td></td>
<td>95% CI: -1.000 to 2.000</td>
<td>95% CI: -0.0000519 to 0.0000508</td>
<td>95% CI: -1.000 to 1.000</td>
</tr>
<tr>
<td>Drug use—Yes or No</td>
<td>p-value: 0.0685</td>
<td>p-value: 0.3701</td>
<td>p-value: 0.1560</td>
</tr>
<tr>
<td></td>
<td>95% CI: -0.0000524 to 2.000</td>
<td>95% CI: -0.0000589 to 0.0000187</td>
<td>95% CI: -0.0000282 to 2.000</td>
</tr>
</tbody>
</table>

No p-values were significant (i.e., <0.01)

Results of gross inspection of ribs

Acute fractures of anterior shaft

The number of infants with grossly observable acute fractures of the anterior or the anterior-lateral portion of the rib shaft was 10 of 90, or 11.1%. The age range of these infants was from 0.3 to 5.0 months. The mean was 2.330 months, with a standard deviation of 1.425. A Wilcoxon Rank Sum test and a Welch two-sample t-test comparing the age of children without
acute anterior rib fractures and those with acute anterior rib fractures indicated p-values of 0.2002 and 0.002593 respectively. A Wilcoxon Rank Sum test and a Welch two-sample t-test comparing the number of clefts per child in the 10R sample between those with and without acute fractures of the anterior portion of the shaft indicated p-values of 0.6103 and 0.5540 respectively. Regarding delivery method, eight of the infants were delivered via vaginally and two via Cesarean. The cause of death was SIDS in four, bed sharing in four, and undetermined and suspicious in two. All infants had undergone CPR. The estimated gestational age was from 35 to 41 weeks, with a mean of 38.5 and a standard deviation of 2.0138. A Wilcoxon Rank Sum test and a Welch two-sample t-test comparing the EGA of children without acute anterior rib fractures and those with acute anterior rib fractures indicated p-values of 0.5009 and 0.4143 respectively. Four of these 10 infants also had acute fractures of the rib head/neck, in two of these infants the cause of death was bed sharing and in the other two it was undetermined and suspicious. The number of acute fractures varied from 1-15 per infant, with three infants having 1 fracture, one having 2 fractures, three having 4 fractures, and one infant each having 5, 8, and 15 fractures. The four infants dying of SIDS had 1, 1, 5, and 8 fractures. The four infants dying while bed sharing had 2, 4, 4, and 4 fractures, and the two infants dying of an undetermined cause under suspicious circumstances had 1 and 15 fractures. None of the infants had gross acute fractures of the lateral portion of the shaft. One infant (undetermined cause and manner of death and suspicious circumstances) had one remote fracture of a rib head/neck, and one remote fracture of the anterior portion of a shaft. Two infants had CMLs with no or scant hemorrhage: three and two CMLs (both bedshare deaths), none had CMLs with cleft material, two had acute
fractures of the rib head (2 in one bed share death, and 1 in one suspicious and undetermined death)

_Acute fractures of lateral shaft_

One neonate, who died as the result of an undetermined cause and manner of death that was regarded as suspicious, had one acute lateral rib fracture. The age at death was 0.75 months, and CPR had been performed. The neonate also had one acute gross fracture of a rib head/neck, four gross remote fractures of the lateral portion of ribs, and CMLs with cleft material present (eight CMLs present in All sample and five CMLs present in 10R sample). No other child had acute lateral rib fractures.

_Acute fractures of rib head/neck_

Six children had acute fractures of a rib head/neck. The mean age in months at death of these children was 1.425 months, with a standard deviation of 0.9715. Five of the children were born vaginally and one via Cesarean section. As for a cause of death, two of these children died while bed sharing and four died an undetermined cause and manner of death that was regarded as suspicious. All had received CPR. The mean EGA was 38.1667 weeks, with a standard deviation of 2.2286. Five of the children had 1 fracture and one child had 15 fractures. In addition, four children had gross acute fractures of the anterior shaft (1, 4, 4, and 15 fractures). Chi-square and Fisher’s exact tests comparing the distribution of children with and without acute anterior fractures and with and without acute posterior fractures indicated p-values of 0.0001391
and 0.001009 respectively (the Pearson chi-square returned a warning as some cell counts were less than 5). One child had a gross acute fracture of the lateral shaft. Two children had gross remote fractures of the rib head/neck (5 and 1 fractures), one child had 1 gross remote fracture of the anterior shaft, and three children had gross remote fractures of the lateral shaft (1, 1 and 4 fractures). One child had CMLs with no or scant hemorrhage (3 CMLs). Two children had CMLs with cleft material present (1 CML with none in 10 rib sample, or 8 CMLs with 5 in 10 rib sample). Two children had acute clefs of the anterior rib head (2 in one and 1 in another)

_Gross remote fractures of the anterior shaft_

Three children had gross remote fractures of the anterior shaft, with two children having 1 fracture, and one child having 3 fractures. All three had been delivered vaginally. The mean age at death was 2.6667 months, with a standard deviation of 0.5774. The cause of death was different for each: bed sharing, SIDS, and undetermined cause and manner under suspicious circumstances. The mean EGA was 39.3333 weeks, with a standard deviation of 0.5774. One child had one gross acute fracture of a rib head and one gross acute fracture of an anterior shaft. Two children had a gross remote fracture of the rib head/neck. Chi-square and Fisher’s exact tests comparing the distribution of children with and without remote anterior fractures and with and without remote posterior fractures indicated p-values of 0.01093 and 0.02002 respectively (the Pearson chi-square returned an warning as some cell counts were less than 5).

_Gross remote fractures of the lateral shaft_
Four children had gross remote fractures of the lateral shaft. All four were delivered vaginally. The number of fractures was 1, 2, 2, and 4. All four died as the result of an undetermined cause and manner of death regarded as suspicious. The mean age at death was 6.1875 months, with a standard deviation of 7.9461. The mean EGA was 38.00 weeks, with a standard deviation of 2.4495. Two children had 1 gross acute fracture of a rib head/neck. One child had a gross acute fracture of a lateral segment of a rib. Two children had gross remote fractures of the rib head/neck (5 and 3 fractures). None had gross remote fractures of anterior shaft. One child had CMLs with no or scant hemorrhage. Three children had CMLs with cleft material (1, 1, 8 with 0, 1, 5 in 10R sample respectively).

**Gross remote fractures of rib head/neck**

Eight children had gross remote fractures of the rib head/neck. The number of remote fractures present was 1 (in four children) and 2, 3, 4, 5, in one child each. The mean age at death was 3.0625 months, with a standard deviation of 0.8634. Six children were delivered vaginally and two via Cesarean section. The mean EGA was 37.625 weeks, with a standard deviation of 1.4079. The cause of death was SIDS in one case, bed sharing in three cases, a known natural cause in one case, and undetermined cause and manner of death regarded as suspicious in three cases. Of the two bed sharing cases, both involved intoxicated adults. Two children had gross acute fractures of rib head/neck. One had a gross acute fracture of anterior shaft. Two had gross remote fractures of anterior rib. Two children had gross remote fractures of lateral shaft of rib. One had CMLs with scant or no hemorrhage. Two had CMLs with cleft material.
Additional microscopic findings

Three children had Salter-Harris fractures. In the 10R sample, there was one fracture in two children, and two fractures in one child. Of the children with Salter-Harris fractures, two were delivered vaginally and in one, the birth method was unknown. The ages at death were 12, 15, and 18 months. All had received CPR. One died as the result of an undetermined natural cause, one died as the result of non-accidental injury, and one died as the result of an undetermined cause and undetermined manner that was regarded as suspicious. The EGA were 39 and 41 weeks for two children and unknown in the third.

Nine children had a CML with scant or no hemorrhage. In the 10R sample, there were two infants with 1 CML, four infants with 2 CMLs, one infant with 3 CMLs, one infant with 4 CMLs, and one infant with 5 CMLs. Of the children with CMLs with no or scant hemorrhage, seven were delivered vaginally and two via Cesarean section. The age at death ranged from 0.25 to 3 months, with a mean of 1.25 months, and a standard deviation of 0.9670. All had received CPR. The cause of death in four was bed sharing, in one was an undetermined cause and undetermined manner that was not regarded as suspicious, in two was an undetermined cause and undetermined manner that was regarded as suspicious, in one was a known natural, and in one was possible accidental trauma. The mean EGA was 38.625 weeks, with a standard deviation of 1.408.

Seven children had a CML with cleft material. In the 10R sample, of these, one infant had 0 CMLs (found in other ribs that would not have been available), four with 1 CML, one with 2 CMLs, and two with 5 CMLs—the one child with no CML identified in the 10R sample was a 3-month-old who was delivered vaginally and died from an undetermined cause and
undetermined manner that was regarded as suspicious. Six of the children were delivered vaginally and one via Cesarean section. The age at death ranged from 0.75 to 15 months, with a mean of 6.107 and a standard deviation of 5.881. All had received CPR. Two died as the result of SIDS, three died as the result of an undetermined cause and manner that was regarded as suspicious, one died while bed sharing and one died from NAI.

Nineteen children had acute clefts of the anterior rib head. All acute clefts were small, and only found with the tip at or immediately adjacent to the anterior edge of the growth plate (i.e., none would have been greater than 1.00 mm in length). In 10R sample, 14 with one cleft, 3 with two clefts, 1 with four clefts, and 1 with five clefts. Twelve children were born vaginally, four via Cesarean section, and in three children, the birth method was unknown. The age at death ranged between 0.03 and 20 months, with a mean of 3.610, and a standard deviation of 5.518. Sixteen children had received CPR and 1 did not. Regarding cause of death, seven were from bed-sharing, three were from SIDS, two were from a known natural cause, one was from an undetermined natural cause, five were from an undetermined cause and manner that was not regarded as suspicious, and one was from accidental trauma.

Spearman correlations are performed comparing the number of clefts per child, number of clefts with a length of greater than 1.00 mm per child, number of clefts with a distance from the anterior edge of the growth plate of greater than 1.00 mm per child, and number of healing clefts per child to various gross and microscopic findings. The Spearman correlations were performed using the 10R+ sample with all children, and with subgroups A, B, D, and E of Group 4. In this sample, the correlation for remote fractures of lateral shaft is not available, and not listed in the table. In the 10R+ sample using all causes of death when comparing number of
gross remote fractures of the rib head/neck against all four categories of rib cleft features, as well as for gross remote fractures of the lateral shaft, gross acute fractures of the rib head/neck (not including when comparing against number of clefts per child), age at death (only when comparing against number of clefts per child), number of CMLs with cleft material, and number of acute microscopic clefts resulted in p-values of $<0.01$; however, in the 10R+ dataset using Group 4, Subgroups A, B, D, and E only, the only comparison resulting in a p-value of $<0.01$ was age at death against number of clefts per child (Table 29).

Chi-square and Fisher’s exact test were performed using the 10R+ dataset and comparing the distribution among all subgroups (A-E) of Group 4 who had various gross findings. Chi-square and Fisher’s exact test were also performed using the 10R+ dataset and comparing the distributions of children with various features of the clefts with other microscopic findings and gross findings. When comparing the distribution of children with and without gross acute fractures of the rib head/neck and with and without gross remote fractures of the lateral shaft, and the five subgroups of Group 4, significant p-values of $<0.01$ resulted. In addition, for the Fisher’s exact test, significant p-values of $<0.01$ were indicated when comparing the distribution of children with and without greater than 2 microscopic clefts and with and without a cleft of length greater than 1.00 mm as well as when comparing infants with and without a cleft greater than 1.00 mm in length and with and without a gross remote fracture of the lateral shaft (Table 30).

Chi-square and Fisher’s exact test were performed using all children comparing presence of gross rib fractures and birth method as well as the presence of shoulder dystocia or another difficult delivery. No significant p-values were obtained (Table 31).
Table 29. Spearman correlations comparing cleft features to various gross and microscopic findings, using 10R+ sample and Group 4; correlations (rho) are listed first, p-values second, significant p-values bold faced.

<table>
<thead>
<tr>
<th>Feature</th>
<th># of clefts per child</th>
<th># of clefts with length &gt;1mm/ child</th>
<th># of clefts with tip &gt;1mm from anterior edge/ child</th>
<th># of healing clefts per child</th>
</tr>
</thead>
<tbody>
<tr>
<td># of gross remote fractures of</td>
<td>0.2911</td>
<td>0.2728</td>
<td>0.3006</td>
<td>0.2095</td>
</tr>
<tr>
<td>rib head/neck</td>
<td>0.01020</td>
<td>0.01640</td>
<td>0.0079</td>
<td>0.06741</td>
</tr>
<tr>
<td># of gross remote fractures of</td>
<td>0.1040</td>
<td>0.0415</td>
<td>0.0360</td>
<td>-0.00392</td>
</tr>
<tr>
<td>anterior shaft</td>
<td>0.3678</td>
<td>0.7203</td>
<td>0.7556</td>
<td>0.9730</td>
</tr>
<tr>
<td># of gross remote fractures of</td>
<td>0.2751</td>
<td>0.4037</td>
<td>0.3656</td>
<td>0.3292</td>
</tr>
<tr>
<td>lateral shaft</td>
<td>0.0155</td>
<td>0.0002722</td>
<td>0.00107</td>
<td>0.003458</td>
</tr>
<tr>
<td>Estimated gestational age</td>
<td>-0.1513</td>
<td>-0.0838</td>
<td>-0.0388</td>
<td>-0.1459</td>
</tr>
<tr>
<td># of gross acute fractures of rib</td>
<td>0.1098</td>
<td>0.2601</td>
<td>0.2331</td>
<td>0.2212</td>
</tr>
<tr>
<td>head/neck</td>
<td>0.3417</td>
<td>0.02236</td>
<td>0.04134</td>
<td>0.05318</td>
</tr>
<tr>
<td># of gross acute fractures of</td>
<td>0.0170</td>
<td>0.0666</td>
<td>0.0150</td>
<td>0.1647</td>
</tr>
<tr>
<td>anterior shaft</td>
<td>0.8833</td>
<td>0.5650</td>
<td>0.8969</td>
<td>0.1522</td>
</tr>
<tr>
<td># of gross acute fractures of lateral</td>
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<td>0.1997</td>
<td>0.1310</td>
<td>0.0770</td>
</tr>
<tr>
<td>shaft</td>
<td>0.6348</td>
<td>0.08165</td>
<td>0.2561</td>
<td>0.5056</td>
</tr>
<tr>
<td>Age at death</td>
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<td>0.1149</td>
<td>0.1517</td>
</tr>
<tr>
<td></td>
<td>0.0004473</td>
<td>0.9932</td>
<td>0.3197</td>
<td>0.1879</td>
</tr>
<tr>
<td># of Salter-Harris fractures</td>
<td>0.1293</td>
<td>0.1061</td>
<td>0.1061</td>
<td>0.1265</td>
</tr>
<tr>
<td></td>
<td>0.2624</td>
<td>0.3584</td>
<td>0.3585</td>
<td>0.2729</td>
</tr>
<tr>
<td># of CMLs with scant or no</td>
<td>-0.2092</td>
<td>-0.1235</td>
<td>-0.1234</td>
<td>-0.1273</td>
</tr>
<tr>
<td>hemorrhage</td>
<td>0.06777</td>
<td>0.2845</td>
<td>0.2846</td>
<td>0.2700</td>
</tr>
<tr>
<td># of CMLs with cleft material</td>
<td>0.3007</td>
<td>0.2874</td>
<td>0.2952</td>
<td>0.3152</td>
</tr>
<tr>
<td></td>
<td>0.00787</td>
<td>0.01125</td>
<td>0.009146</td>
<td>0.005236</td>
</tr>
<tr>
<td># of acute microscopic clefts</td>
<td>-0.2956</td>
<td>-0.1895</td>
<td>-0.2005</td>
<td>-0.1421</td>
</tr>
<tr>
<td></td>
<td>0.009055</td>
<td>0.09878</td>
<td>0.08038</td>
<td>0.2177</td>
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</tbody>
</table>

Spearman correlations comparing cleft features to various gross and microscopic findings, using 10R+ sample and Group 4, subgroups A, B, D, and E (n=66)

<table>
<thead>
<tr>
<th>Feature</th>
<th># of clefts</th>
<th># of clefts with length &gt;1mm</th>
<th># of clefts with tip &gt;1mm from anterior edge</th>
<th># of healing clefts</th>
</tr>
</thead>
<tbody>
<tr>
<td># of gross remote fractures of rib</td>
<td>0.1894</td>
<td>0.0577</td>
<td>0.1055</td>
<td>0.0203</td>
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<tr>
<td>head/neck</td>
<td>0.1276</td>
<td>0.6451</td>
<td>0.3993</td>
<td>0.8715</td>
</tr>
<tr>
<td># of gross remote fractures of</td>
<td>0.0158</td>
<td>-0.1070</td>
<td>-0.1029</td>
<td>-0.1343</td>
</tr>
<tr>
<td>anterior shaft</td>
<td>0.8997</td>
<td>0.3923</td>
<td>0.4108</td>
<td>0.2822</td>
</tr>
<tr>
<td>Estimated gestational age</td>
<td>-0.2670</td>
<td>-0.0651</td>
<td>-0.0636</td>
<td>-0.2690</td>
</tr>
<tr>
<td></td>
<td>0.04257</td>
<td>0.6303</td>
<td>0.6384</td>
<td>0.04328</td>
</tr>
<tr>
<td># of gross acute fractures of rib</td>
<td>-0.1086</td>
<td>0.0714</td>
<td>-0.1030</td>
<td>0.0188</td>
</tr>
<tr>
<td>head/neck</td>
<td>0.3854</td>
<td>0.5691</td>
<td>0.4107</td>
<td>0.8808</td>
</tr>
<tr>
<td># of gross acute fractures of</td>
<td>-0.0264</td>
<td>0.0862</td>
<td>-0.0775</td>
<td>0.1712</td>
</tr>
<tr>
<td>anterior shaft</td>
<td>0.8333</td>
<td>0.4911</td>
<td>0.5362</td>
<td>0.1693</td>
</tr>
<tr>
<td>Age at death</td>
<td>0.3936</td>
<td>-0.0461</td>
<td>0.1133</td>
<td>0.0968</td>
</tr>
<tr>
<td></td>
<td>0.001076</td>
<td>0.7134</td>
<td>0.3651</td>
<td>0.4395</td>
</tr>
<tr>
<td># of Salter-Harris fractures</td>
<td>-0.0663</td>
<td>-0.0751</td>
<td>-0.0722</td>
<td>-0.0943</td>
</tr>
<tr>
<td></td>
<td>0.5971</td>
<td>0.5489</td>
<td>0.5644</td>
<td>0.4515</td>
</tr>
<tr>
<td># of CMLs with scant or no</td>
<td>-0.2773</td>
<td>-0.1913</td>
<td>-0.1840</td>
<td>-0.2401</td>
</tr>
<tr>
<td>hemorrhage</td>
<td>0.02417</td>
<td>0.1239</td>
<td>0.1392</td>
<td>0.0522</td>
</tr>
<tr>
<td># of CMLs with cleft material</td>
<td>0.2550</td>
<td>0.0714</td>
<td>0.1423</td>
<td>0.2311</td>
</tr>
<tr>
<td></td>
<td>0.03882</td>
<td>0.5691</td>
<td>0.2543</td>
<td>0.06193</td>
</tr>
<tr>
<td># of acute microscopic clefts</td>
<td>-0.2962</td>
<td>-0.1284</td>
<td>-0.1957</td>
<td>-0.0902</td>
</tr>
<tr>
<td></td>
<td>0.01574</td>
<td>0.3041</td>
<td>0.1154</td>
<td>0.4712</td>
</tr>
</tbody>
</table>
Table 30. Chi-square and Fisher’s exact test analysis of distribution of various gross and microscopic cleft features among subgroups of Group 4.

<table>
<thead>
<tr>
<th>Subgroups vs presence/absence of</th>
<th>Chi-square</th>
<th>Fisher’s exact</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Two-tailed</td>
<td>Two-tailed</td>
</tr>
<tr>
<td></td>
<td>p-value</td>
<td>p-value</td>
</tr>
<tr>
<td>Group 1, A-E, 10R+ sample (n=77)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Subgroups vs presence/absence of gross acute fractures of rib head/neck</td>
<td>0.003301*</td>
<td>0.01061</td>
</tr>
<tr>
<td>Subgroups vs presence/absence of gross acute fractures of anterior shaft</td>
<td>0.3520*</td>
<td>0.3703</td>
</tr>
<tr>
<td>Subgroups vs presence/absence of gross acute fractures of lateral shaft</td>
<td>0.1933*</td>
<td>0.2338</td>
</tr>
<tr>
<td>Subgroups vs presence/absence of gross remote fractures of rib head/neck</td>
<td>0.3049*</td>
<td>0.4100</td>
</tr>
<tr>
<td>Subgroups vs presence/absence of gross remote fractures of anterior shaft</td>
<td>0.7905*</td>
<td>0.8177</td>
</tr>
<tr>
<td>Subgroups vs presence/absence of gross remote fractures of lateral shaft</td>
<td>0.0004348</td>
<td>0.0002697</td>
</tr>
</tbody>
</table>

| Subgroups vs presence/absence of gross acute fractures of rib head/neck | 0.01206 | 0.007591         |
| Subgroups vs presence/absence of gross acute fractures of anterior shaft | 0.1588* | 0.08315          |
| Subgroups vs presence/absence of gross acute fractures of lateral shaft | 0.8545* | 0.7195          |
| Subgroups vs presence/absence of gross remote fractures of anterior shaft | 0.7063* | 0.3247          |
| Subgroups vs presence/absence of gross remote fractures of lateral shaft | 0.1291* | 0.1040          |
| Subgroups vs presence/absence of gross remote fractures of anterior shaft | 1.000* | 1.000*          |
| Subgroups vs presence/absence of gross remote fractures of lateral shaft | 0.01577* | 0.009348 |

*some cells have counts of <5
**significant p-values are bold faced
Table 31. Chi-square and Fisher’s exact test analysis of distribution of gross rib fractures among birth methods

<table>
<thead>
<tr>
<th></th>
<th>Chi-square</th>
<th>Fisher’s exact test</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Two-tailed</td>
<td>Two-tailed</td>
</tr>
<tr>
<td></td>
<td>p-value</td>
<td>p-value</td>
</tr>
<tr>
<td>All infants</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gross remote fractures rib head/neck versus birth method (n=83)</td>
<td>1.000*</td>
<td>1.000</td>
</tr>
<tr>
<td>All infants</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gross remote fractures rib head/neck versus shoulder dystocia/difficult delivery (n=83)</td>
<td>0.8424*</td>
<td>0.3386</td>
</tr>
<tr>
<td>All infants</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gross remote fractures of anterior shaft versus birth method (n=83)</td>
<td>0.6049*</td>
<td>0.5502</td>
</tr>
<tr>
<td>All infants</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gross remote fractures of anterior shaft versus shoulder dystocia/difficult delivery (n=83)</td>
<td>0.3291*</td>
<td>0.1393</td>
</tr>
<tr>
<td>All infants</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gross remote fractures of lateral shaft versus birth method (n=83)</td>
<td>0.4311*</td>
<td>0.3104</td>
</tr>
<tr>
<td>All infants</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gross remote fractures of lateral shaft versus shoulder dystocia/difficult delivery (n=83)</td>
<td>1.000*</td>
<td>1.0000</td>
</tr>
</tbody>
</table>

*some cells have counts of <5

Plots of some of the above significant correlations (notably number of clefts per child and number of clefts per child with a length of greater than 1.00 mm, and the number of gross remote fractures of the rib head/neck and lateral shaft, and the age of the child at death) are provided (Figs. 70-75).
Figure 70. Plot of number of clefts per child versus number of gross remote fractures of the rib head/neck per child.

Figure 71. Plot of number of clefts per child with a length of >1.00 mm versus number of gross remote fractures of the rib head/neck per child.

Figure 72. Plot of number of clefts per child versus number of gross remote fractures of the lateral rib shaft per child.

Figure 73. Plot of number of clefts per child with a length of >1.00 mm versus number of gross remote fractures of the lateral rib shaft per child.
Bed sharing involving intoxicated and non-intoxicated adults

As some children who died while bed sharing were with intoxicated adults and others were not, comparisons of the two samples were performed. Twenty-five children died while bed sharing with non-intoxicated adults, while in five deaths, the adult or adults were intoxicated. Using the All sample, Wilcoxon Rank Sum tests comparing the distribution of children who died while bed sharing with an intoxicated adult or with a non-intoxicated adult and the age at death (p-value of 0.3143), total number of clefts per child (p-value of 0.3102), and number of clefts with length of greater than 1.00 mm per child (p-value of 0.5866) did not yield significant results. Using the 10R dataset and comparing number of clefts (p-value of 0.3515) and number of clefts with length of greater than 1.0 mm (p-value of 0.5866) also did not yield significant
results. Using the 10R+ dataset, in which 22 children died while bed sharing with non-intoxicated adults and five died while bed-sharing with intoxicated adults, comparing the age at death (p-value of 0.2819), number of clefts per child (p-value of 0.2952), number of clefts with length of greater than 1.0 mm in length per child (p-value of 0.6175) also did not yield significant results.

Age at death: SIDS versus bed sharing

The mean estimated gestational age of infants who died as the result of SIDS (n=20) was 38.30 and the mean estimated gestational age of infants who died as the result of bed-sharing (n=30) was 38.17; however, the mean age at death of infants who died as the result of SIDS was 3.9375 months (with a standard deviation of 1.4687) and the mean age at death of infants who died as the result of bed sharing was 3.2017 months (with a standard deviation of 4.2786). If only the infants who died while bed-sharing and had an age in months of less than 10 were considered (n=28), the mean age at death was 2.2161 months (with a standard deviation of 1.9710). Using a Welch two-sample t-test comparing the mean age at death of infants who died from SIDS with all those who died while bed sharing indicated an insignificant p-value of 0.3906; however, a Wilcoxon Rank Sum test with continuity correction comparing the same two distributions, indicated a significant p-value of 0.001523, and, when the Welch two-sample t-test was performed a second time, using only infants who had died while bed sharing with an age at death of 10 months or less compared against the mean age of those infants who died as the result of SIDS, the p-value was significant at 0.001156.
CHAPTER 5: DISCUSSION

Rib fractures in infants, especially fractures in the posterior portion of the rib, which include fractures of the rib head and neck, are strongly associated with inflicted injury (Thomas, 1977; Kleinman et al., 1995; Dolinak and Matshes, 2005); however, Bulloch et al. (2000) and Cadzow and Armstrong (2000), using statistical methods, did not identify an association between abuse and posterior rib fractures in their studies, but, the authors did support the general association between rib fractures in infants and abuse. The mechanism by which these posterior fractures occur is opined to be either 1) anterior-to-posterior (AP) compression of the trunk, levering the rib against the transverse process and causing a fracture near the rib neck (Kleinman, 1987a; Kleinman and Schlesinger, 1997), which, anatomically, is plausible, or 2) lateral compression of the chest (DiMaio and DiMaio, 2001). AP compression could also result in fractures of the rib by pulling the rib head away from the vertebral body and causing a rib fracture at this location (Worn and Jones, 2007). Compression of the chest does not necessarily have to cause death; however, if fractures occur in these locations because of this mechanism, a less than lethal amount of force applied may produce less than striking anatomic changes (i.e., small fractures). Therefore, small fractures of the rib head and neck may be indicative of past episodes of chest compression that were non-fatal. Also, although in some cases inflicted fatal injury in children is identifiable grossly (e.g., inflicted head trauma manifesting as a subdural hemorrhage and retinal hemorrhages), other forms of death (e.g., suffocation) may produce no or only subtle changes. As compression of the chest is a common mechanism used to some extent during many forms of abuse in young children, especially infants, and as compression of the chest may cause fractures of the rib head and neck, careful examination of this region may help
differentiate a subtle homicide from SIDS, or, may help reveal a past pattern of forceful compression of an infant’s chest, and provide investigators with information they can use when interviewing caretakers. Forensic anthropologists, in their investigation of skeletal trauma, would benefit from such information, and forensic pathologists, who often consult forensic anthropologists, would also benefit. The amount of literature on child abuse is substantial; however, the amount of literature on child abuse written from a forensic anthropologist’s perspective is minimal, and this material could offer a boost.

The main purpose of this research was to determine if an association exists between the rib head clefts identified microscopically by the author and suspicious deaths. Suspicious deaths are those in which fatal inflicted trauma was suspected as a mechanism of death, but unproven in the opinion of the forensic pathologist conducting the autopsy. Although abuse can be a single event, abuse can also occur from multiple events. If microscopic rib head clefts are associated with suspicious deaths, they may provide for a marker that can be used to further the investigation of infant deaths. The hypothesis being that since chest compression can cause fractures of the ribs and since chest compression is a component of forms of child abuse, especially of infants, that if rib head clefts are associated with suspicious infant deaths, that they represent a marker of recent or past episodes of forceful chest compression, and, when identified, can be communicated to investigators who can use the information when interviewing the child’s caretakers about recent or past treatment of the child.

Fractures of the rib can be detected by one of several modalities, including radiography, scintigraphy, CT scans, gross inspection at autopsy, and microscopic examination. In the examination of the skeletal system, removal of soft tissue and direct inspection is the gold
standard if preservation of the bone is required (Catteneo et al., 2006; Kemp et al., 2013); however, for the examination of infant rib heads, because of the small size of the bones and the subtle morphologic changes (e.g., the clefts measured varied from 0.001 mm to greater than 3 mm in length), histologic examination is the gold standard (McGraw et al., 2002; Klotzbach et al., 2003). Love et al. (2014) opine that removal of soft tissue is the gold standard for examination of infant ribs; however, compared to removal of rib heads at autopsy, complete removal of soft tissue from the ribs is much more extensive, time-consuming, and invasive, and not necessarily a procedure to be performed on a routine basis for infant autopsies, whereas microscopic examination of the rib head and neck can be a routine autopsy procedure.

In cases of child abuse, or suspected child abuse, Kleinman et al. (1986) and Cooperman and Merten (2011) advocate removal of the proximal humerus, distal femur, and proximal and distal tibia to examine the bone with high-detailed radiography and with microscopic examination to assess for the presence of metaphyseal lesions and Dolinak and Matshes (2005) advocate removal of the left and right side of the rib cage to further assess for fractures if multiple recent or remote fractures are identified upon in-situ inspection. Although these steps are advocated only in the autopsy of a known or suspected victim of inflicted injury, removal of the rib heads is much less destructive and routine microscopic examination of this high yield location for detecting the presence of injuries may be useful. Also, when abuse is confirmed by autopsy and investigation, microscopic examination of the rib heads may help identify past episodes of abuse, and increase the number of injuries found. The identification of as many injuries as possible and of past patterns of abuse may help both in the determination of child abuse and in its prosecution (McGraw et al., 2002). Opinion as to how to perform an autopsy as
to best allow for identification of skeletal injuries is not limited to forensic pathologists, Love and Sanchez (2009) and Love et al. (2014) discuss examination for metaphyseal lesions at autopsy, and Dr. Love is a forensic anthropologist.

Histologic examination of the organs is a required component for the diagnosis of SIDS at autopsy as SIDS is a diagnosis of exclusion and occasionally the cause of death of an infant is identified within the tissue of organs (e.g., myocarditis or encephalitis) and could not be appreciated upon gross inspection (Bergman et al., 1970). Although a minimum number of organs to examine histologically is recommended (including the brain, heart, liver, lungs, and kidneys), examination of other organs (e.g., the diaphragm) has yielded information allowing for the determination of a cause of death (Bergman et al., 1970; Sundararajan et al., 2005; Eisenhut, 2011). Therefore, inclusion of histologic sections of the rib as a possible assessment for recent or past episodes of chest compression is acceptable. Although incorporation of microscopic examination of a block of rib heads removed from infants at autopsy is simple, an understanding of the microscopic findings in this region and their associations and implications is less simple. Based upon review of the literature, apparently only Kleinman et al. (1992) have studied the rib head microscopically. Kleinman et al. (1992) opined that isolated fractures of the rib head must be viewed with caution; thus, more information is required to properly interpret histologic changes in this region. The ribs examined from the 90 children in this current study have begun this process.

The statement by Schmidt (1979:105) that “a child’s rib cage may be pressed as far back as the spine without fracture occurring” highlights the general opinion that children’s ribs are rather resistant to fracture. In addition, multiple reports and reviews indicate the rarity of infant
rib fractures due to CPR (Feldman et al., 1984; Hobbs, 1989; Betz and Liebhardt, 1994; Spevak et al., 1994; Bush et al., 1996; Matshes and Lew, 2010b). Considering that CPR involves compression of the chest and routinely causes fractures when performed in adults (Matshes and Lew, 2010b), the absence or near absence of rib fractures reported in children after CPR serves to support the idea that infant’s ribs are highly flexible and only likely to break after the application of extreme force. One must remember however that imaging techniques for living patients (e.g., x-ray and CT) are not good at identifying recent rib fractures, which would contribute to the idea that rib fractures in children occurring as a result of CPR are rare.

In this current study, clefts at the anterior surface of the rib head were quite common, with 66 of 90 children having at least one cleft identified in their 3 to 20 rib heads available for analysis. The clefts have the same histologic appearance as fractures and are not described in the literature as a normal morphologic finding. None of the clefts were associated with an exuberant periosteal reaction, indeed almost all had no or essentially no periosteal reaction. Kleinman (1987a, 2008) has described how metaphyseal lesions can have no hemorrhage and heal without the formation of new subperiosteal bone formation. Metaphyseal lesions are strongly associated with abuse (Silverman, 1974; Cameron and Rae, 1975; Cooperman and Merten, 2001; Arkader et al., 2007) and occur in the metaphysis, the same general location as these clefts. Metaphyseal lesions do not have to extend completely across the length of the metaphysis at the growth plate. If both these clefts and metaphyseal lesions have no periosteal reaction and occur in the metaphysis and have histologic features of a fracture, what distinguishes them? At what degree of force does a cleft become a metaphyseal lesion? The metaphysis is immature bone and once a fracture has begun it seems possible that not substantially more force may be required to cause
its propagation through the immature bone. Therefore, is abuse much more common than thought, or are infant bones more prone to fracture than previously believed? Of note, the clefts containing eosinophilic material are a common finding, present in approximately 2/3s of the children examined, while metaphyseal lesions containing the same eosinophilic material were much less common in this study (found in only 7 of 90 children). While metaphyseal lesions have been described in many bones, their identification in ribs is not apparent in the literature; however, the results of this study indicate they exist, and, if not carefully examined for (i.e., with histology), can be easily missed and not identified in any given case.

Although the main purpose of this study was to document clefts of the anterior surface of the rib head in infants and other children and statistically analyze for an association between various features of the clefts (e.g., number per child and length) and suspicious deaths, statistical analysis of cleft features and other features of the child with the clefts (e.g., age at death, circumstances of death, birth method, and socio-economic factors, including boyfriend versus biological father as the dominant male in a child’s life) was also important. More information was collected than was analyzed. Specifically, in most infants, the ribs examined were known to be left or right, and this siding was maintained in the collection of data (e.g., if a cleft was identified, the side of the rib on which it was found was noted). However, for three reasons: 1) some infants did not have a left and right designation for ribs (i.e., when the ribs were originally submitted for analysis, the side from which they were taken was not indicated), 2) to decrease the complexity of statistical analysis, and 3) because only the number of clefts per child and not the side from which they occurred was felt to be of importance in such a relatively small sample (n=90), the left and right sides were combined before final statistical analysis was performed.
As Kleinman et al. (1991) had described cartilage extensions from the growth plate associated with trauma, when a cartilage extension at the anterior surface of the growth plate was identified, it was documented, along with its general form (thin, thick, or intermediate) and whether or not the adjacent bone bark extended its entire length, or not. However, these determinations were subjective. First, even determining whether or not a cartilage extension was present was difficult, as a slight projection of cartilage at the anterior edge of the growth plate may be an extension or merely an accentuation of the curvature of the growth plate. Second, although the extreme ends of the shape of a cartilage extension (thin and thick) were usually apparent, the variation in form was marked, and dividing the extensions into thin, thick, and intermediate was difficult. Finally, any measurement of the cartilage extension was subjective as determination of where in the growth plate the extension began was difficult. Although cartilage extensions are associated with past trauma, they were identified in infants who died at birth, which, implies that the anterior edge of the growth plate has normal variation, which includes extensions of the cartilage. Some cartilage extensions were identified though that had the remains of apparent material from a cleft associated with them. Therefore, these cartilage extensions may in fact represent a healed small cleft occurring at the anterior edge of the growth plate. As multiple small clefts were identified at this same site in the ribs in the sample, the hypothesis appears plausible, but objective testing is difficult, and based upon the conclusions of this study, may not contribute substantial information, as the clefts of most importance are those away from the anterior edge of the growth plate, having a length of greater than 0.501 mm.

Examination of the data on a case-by-case basis indicates that the number of clefts per child is not, for any given child, a reliable marker of the circumstances of death. At the extremes
of the sample, one child, who died from an undetermined natural cause of death, with neither investigation nor autopsy indicating anything suspicious about the death, had 10 ribs available for analysis, and each rib had a cleft, only one of which was healing. In contrast, two children, who died as the result of non-accidental injury (NAI), with both having a subdural hemorrhage and retinal hemorrhages, likely occurring during an episode of shaking with or without slamming and that may have also included chest compression, each had no clefts (with three rib heads available for analysis in one child, and thirteen available in the second child). In addition, in a third child, whose death was from an undetermined cause and manner, but whose death was considered suspicious for the possibility of fatal abuse, no clefts were found among 13 rib heads available for analysis. And, one child, who died as the result of an undetermined cause and manner regarded as suspicious for fatal abuse, had a single cleft identified among eight rib heads. Therefore, the presence of multiple clefts does not absolutely indicate fatal or even past abuse, and the absence or near absence of clefts does not preclude fatal abuse; however, this is expected as the histologic appearance of the clefts is not entirely consistent with them having occurred at the time of death. However, although they might not indicate a fatal abusive episode, they may indicate past subtle abuse due to chest compression and, it must be considered that a child who otherwise died under non-suspicious circumstances (e.g., bed-sharing with both parents and a sibling) may have sustained abusive chest compression in the past, in an event that occurred before their death.

While individual cases may indicate that the absolute number of clefts per child is not necessarily indicative of the circumstances of death (i.e., children can die from fatal abuse or under suspicious circumstances and have no clefts, and children with multiple clefts can die
under non-suspicious circumstances), the distribution of the number of clefts per child for each Subgroup (A-E) of Groups 1-4, when compared to other Subgroups (A-E) within the same Group (1-4), is different, and the number of clefts per child for Subgroup C (representing the children who died from NAI or who died under suspicious circumstances) is consistently higher than the other four Subgroups (A, B, D, and E), across all four Groups (1-4) and all four datasets (All, 6R, 10R, and 10R+). A Kruskal-Wallis test comparing the distribution of the number of clefts per child among four or five subgroups of the four groupings, using all available datasets (a total of 32 individual tests), did not indicate significance (there was a p-value of <0.05 for 6 tests, but no test had a p-value of <0.01). All tests with a p-value of <0.05 used the 10R or 10R+ sample and Groups 1-3. Groups 1 and 3 included the one death certified as SIDS, which had suspicious circumstances, placed into Subgroup C; however, no p-value of <0.05 was identified when using Group 4. These results indicate that under certain conditions of testing (i.e., the right groupings of circumstances of death and number of ribs per child available for analysis), the distribution of the number of clefts per child is potentially not similar between the various subgroups of circumstances of death (i.e., SIDS, bed sharing, NAI/suspicious, other natural, and miscellaneous deaths). In the 10R+ sample, using only Subgroups A-D (excluding Subgroup E, which was the miscellaneous category), for both Groups 1 and 3 (which included the suspicious SIDS death in Subgroup C), the Kruskal-Wallis test had an insignificant p-value, but which was <0.05, and, using the 10R+ sample and Subgroups A-D from Groups 2 and 4 (which included the suspicious SIDS death in the SIDS category, as it was originally certified), the Kruskal-Wallis test had an insignificant p-value of greater than 0.05. So, the categorization of children into different subgroups based upon their circumstances of death definitely affects the results of
statistical analysis. Although the distribution of the numbers of clefts per child among the Subgroups A-E was of borderline significance (depending upon the dataset and Group (1-4) used), a Chi-square and Fisher’s exact test, comparing the distribution of the number of children with and without greater than two microscopic clefts and with and without a cleft with a length of greater than 1.00 mm, returned significant, or close to significant p-values of 0.01206 and 0.007591 respectively, indicating that children with more clefts may be more likely to have a cleft with a length of greater than 1.00 mm.

While the distribution of the number of clefts per child among the different subgroups within a group may be of questionable significance (as the results depend upon the specific Group (1-4) and Subgroups (A-E) used), the distribution of the number of clefts with a length of greater than 1.00 mm per child among the subgroups of each group is significant. As with analysis of the number of clefts per child, a Kruskal-Wallis test comparing the distribution of number of clefts with a length of greater than 1.00 mm per child among four or five subgroups of the four groupings, using all available datasets was performed (a total of 32 individual tests), indicating a p-value of <0.05 for 26 tests, and 9 tests had a significant p-value of <0.01. In the 10R+ sample, using Subgroups A-E and Subgroups A-D, the p-value was 0.03308 or less for Groups 1-4. The inclusion of the E Subgroup (which included the miscellaneous causes of death) resulted in a lower p-value than testing of the distribution of number of clefts with a length of greater than 1.00 mm per child among only the first four of the Subgroups (A-D) in each grouping. These results indicate that the distribution of the number of clefts with a length of greater than 1.00 mm per child is not identical among the subgroups (circumstances of death). Similar results were obtained when comparing the distribution of the number of clefts with a
Combining all non-suspicious circumstances of death (i.e., SIDS, bed-sharing, natural, and other) into one group (both Group 3 and Group 4 were used, as one SIDS death was either suspicious or non-suspicious, depending upon the interpretation of the historical evidence), and comparing the distribution of the number of clefts per child and number of clefts with a length of greater than 1.00 mm per child, or with a length of greater than 0.501 mm between the two groups (i.e., suspicious and non-suspicious) also yielded significant p-values for each analysis. For Group 3, the p-values determined when comparing the number of clefts per child, number of clefts greater than 1.00 mm per child, and number of clefts greater than 0.501 mm per child between suspicious and non-suspicious deaths were 0.00326, 0.00145, and 0.00145 respectively, and for Group 4, were 0.00658, 0.00620, and 0.00214 respectively, indicating that the median number of clefts per child, and median number of clefts with a length of greater than 0.501 and 1.00 mm per child was different between suspicious and non-suspicious deaths.

For testing purposes, Group 4 most accurately reflected the death certification process. All deaths certified as SIDS (including the one death with potentially suspicious historical information) were included in the SIDS subgroup (Subgroup A). All bed sharing related deaths were included in the bed sharing subgroup (Subgroup B). All deaths certified as homicide and those certified as an undetermined cause and manner of death, but which were suspicious for fatal abuse, were included in Subgroup C. All known natural deaths were included in Subgroup D. And Subgroup E included all of the miscellaneous deaths, including those in which both an accidental or natural death of the child was possible (e.g., a death where both SIDS and bed sharing were in the final differential diagnosis for possible causes of death), and also, included
those deaths certified as undetermined natural, for which an argument could easily be made to place them within the miscellaneous category, as the cause and manner of death could have been certified as undetermined cause and manner, not regarded as suspicious, and not certified as undetermined natural. Of the datasets, the 10R+, although it has fewer samples represented, is the most consistent with regards to number of rib heads available for histologic analysis per child, as each child in the sample has from 8-11 rib heads available for analysis, while, in the entire sample, each child has between 3-20 rib heads available for analysis. In the All sample, the number of rib heads available for histologic analysis per child is 3 to 20, and, as shown, weighting of the statistical results by number of rib heads available for analysis per child is not acceptable, as when weighted samples were used, essentially all statistical testing returned a p-value of <0.0009. The 6R sample has too few rib heads available for analysis per child and the 10R sample includes infants with 3-7 rib heads available for analysis, and comparing the number of clefts in a child with three rib heads available for analysis to one with eleven is not appropriate.

When the 10R+ sample is used for a Kruskal-Wallis test analyzing the distribution of numbers of clefts per child among Subgroups A-E for Group 4, the p-value is 0.05051, and when analyzing the distribution of numbers of clefts per child among Subgroups A-D for Group 4, the p-value is 0.05877; however, when analyzing the distribution of numbers of clefts with a length of greater than 1.00 mm per child among Subgroups A-E for Group 4, the p-value is significant at 0.007817, and when analyzing the distribution of numbers of clefts per child among Subgroups A-D for Group 4, the p-value is insignificant at 0.03308. Therefore, the distribution of numbers of clefts greater than 1.00 mm in length per child is not identical between the five
Subgroups (A-E). Chi-square analysis and Fisher’s exact test supported this conclusion, showing an unequal distribution of numbers of children with and without a cleft with a length of greater than 1.00 mm between the five subgroups of circumstances of death in Group 4 (using the 10R and 10R+ sample).

To identify the subgroups responsible for this finding, Wilcoxon Rank Sum testing was performed comparing the distribution of the number of clefts per child, the number of clefts greater than 1.00 mm in length per child, and the number of clefts greater than 0.501 mm in length per child between all combinations of subgroups in all four groups, using the 10R sample for all, and the 10R+ sample for Group 4 (a total of 150 tests). Using the 10R+ sample and Group 4, of 30 tests, 12 had p-values of <0.05 (although, the Bonferroni correction was not applied). When comparing SIDS versus suspicious deaths and bed-share versus suspicious deaths, the number of clefts per child and the number of clefts greater than 0.501 mm in length had un-similar or borderline un-similar distributions (with p-values of 0.02934 or less); however, the distribution of the number of clefts greater than 1.00 mm per child was not significantly different (p-values of 0.06194 and 0.0802 respectively). The lack of a significant difference between the SIDS group and the suspicious group for number of clefts greater than 1.00 mm in length may reflect the inclusion of the one SIDS death with a suspicious historical fact in the SIDS group instead of in Subgroup C, with the other suspicious deaths. The distribution of the number of clefts with a length greater than 1.00 mm per child was not significantly different when comparing bed-sharing versus known natural deaths (p-value of 0.04923), bed-sharing versus the other category (p-value of 0.03397), known natural versus suspicious deaths (p-value of 0.01318), but was significantly different when comparing suspicious versus the other category.
(p-value of 0.004056). The fact that the most significant p-value is in the Wilcoxon Rank Sum test comparing the distribution of the number of clefts per child that are greater than 1.00 mm in length between Subgroup C (the NAI and suspicious deaths) and Subgroup E (the collection of miscellaneous circumstances of death, including undetermined naturals and those for whom the cause and manner of death were undetermined, yet the death was not thought to be suspicious) is unexpected; however, it may indirectly serve to highlight two points. First, the determination of cause and manner of death is, while made based upon careful observations of various factors, subjective. Second, the cause and manner of death does not necessarily reflect past treatment of a child. For example, children who die while bed sharing with adults, who are legitimately accidentally asphyxiated, can easily have sustained past episodes of abuse. The cause and manner of death certification would reflect the bed sharing, but not necessarily the past episodes of abuse, but the past episodes of abuse may have created rib fractures that are identified.

When the cause and manner of death of the child was originally certified, or when a determination as to whether or not a death that had an undetermined cause and manner was suspicious for fatal abuse was made, the presence of microscopic rib head clefts, whether by number or by size, did not impact the decision; however, the presence of acute or remote rib fractures identified grossly at the time of autopsy did affect decisions. Therefore, any association between the presence of gross and microscopic fractures in children could be interpreted in one of two ways: 1) as the presence of lateral rib fractures influenced the determination of cause and manner of death, often causing both to be certified as undetermined unless there were other important circumstances, if an association between the gross rib fractures and the microscopic rib head clefts is identified, then any association between microscopic rib head clefts and suspicious
deaths could be seen as biased; however, the presence of grossly identifiable lateral rib fractures was not a sole factor in identifying a death as suspicious, or 2) the association of microscopic rib head clefts with suspicious deaths and with the presence of grossly identifiable lateral rib fractures provides support to the association of lateral rib fractures with possible child abuse.

Using Group 1, Subgroups A-E, and the 10R+ sample, Chi-square and Fisher’s exact test analysis of the association between the various types of fractures identified at autopsy (i.e., acute or remote fractures of the anterior shaft, lateral shaft, or rib head/neck area) and the subgroups of circumstances of death revealed only p-values of <0.05 for acute fractures of the rib head/neck and remote fractures of the lateral shaft; therefore, these were the only two types of fractures found grossly that had a potential unequal distribution among the five subgroups of circumstances of death. If a particular fracture type were used to determine the cause and manner of death, it would be expected that the Chi-square or Fisher’s exact test would show an unequal distribution of this fracture type among the subgroups. This was only true for acute fractures of the rib head/neck and remote fractures of the lateral shaft. When Chi-square and Fisher’s exact tests were used to analyze the distribution of numbers of children with and without a cleft with a length of greater than 1.00 mm against the six types of gross rib fracture, the only significant or borderline significant p-values (0.01577 and 0.009348 for Chi-square and Fisher’s exact test respectively) were obtained by comparison of number of children with and without a cleft greater than 1.00 mm in length and the presence or absence of a remote fracture of the lateral shaft.

Only four children had remote fractures of the lateral shaft, and all had deaths certified as undetermined cause and manner and were regarded as suspicious. Of note, the distribution of
children within subgroups should have played no role in this test, as the calculations merely compared the number of children with and without a cleft with a length of greater than 1.00 mm and those with and without a gross remote fracture of the lateral shaft. Instead, the testing would indicate that children with a remote fracture of the lateral shaft of a rib are more likely to have a rib head cleft that has a length greater than 1.00 mm. As children with an unexplained remote fracture of the lateral shaft of the rib, in general, are considered as possibly having sustained abuse, the cause and manner of death are more likely to both be certified as undetermined, and the death to be considered suspicious. Because the distribution of remote fractures of the lateral shaft of the rib is unequal when comparing infants with and without a rib head cleft of greater than 1.00 mm in length, it can be argued that because the presence of a remote fracture of the lateral shaft of the rib influenced the decision as to cause and manner of death, that subsequent testing of the distribution of clefts with a length of greater than 1.00 mm among the various subgroups of each group representing general categories of cause and manner of death determination is biased.

Spearman’s correlation, using the 10R+ sample and all subgroups, comparing number of clefts per child and number of clefts with a length of greater than 1.00 mm per child to the number of the various gross fractures, age at death, and number of Salter-Harris fractures, CMLs (with scant or no hemorrhage and with cleft material), and microscopic clefts, generated p-values of <0.05 when comparing number of remote fractures of the rib head/neck, number of remote fractures of the lateral shaft, number of CMLs with cleft material each to both the number of clefts per child and the number of clefts with a length of greater than 1.00 mm per child; however, when the Spearman correlation is performed using the 10R+ sample, but without
children who died from NAI or under suspicious circumstances, there is no significant
correlation between the number of clefts or number of clefts greater than 1.00 mm in length and
the number of remote fractures of the rib head/neck, and only the correlation of number of clefts
per child with the number of CMLs with cleft material has a p-value of near significance (p-
value: 0.03882), and not the correlation with number of clefts with a length greater than 1.00 mm
(p-value: 0.5691). As clefts with a length of greater than 1.00 mm and CMLs are associated with
the category that was removed (i.e., children who died under suspicious circumstances), the
results are not surprising. In both analyses (i.e., both with suspicious deaths and without
suspicious deaths), the number of clefts per child correlates significantly with age at death (p-
values of 0.0004473 and 0.001076) and number of acute microscopic clefts per child (p-values of
0.009055 and 0.01574).

Although review of the literature indicates a possible link between prematurity and a risk
of fracture (Dahlenburg et al., 1989; Dabenzies et al., 1997), no consistent association between
estimated gestational age of the child and the presence or size of clefts was found. Statistical
analysis using a limited number of children (only those who died from SIDS or other natural
causes, and the 10R+ dataset, resulting in an \( n \) value of 30), indicated a significant correlation
between the number of clefts per child and the sample of circumstances of death used (p-value of
0.006648, and a correlation coefficient of -0.500); however, all other statistical analyses, using
other groupings of the sample, and various analyses found no statistically significant association
between EGA and number of clefts per child, or number of clefts of a certain length per child.

However, statistical tests did indicate an association between age of the child at death and
number of clefts per child. While the initial impression may be that younger children are more
prone to develop these clefts because their bone is more immature, the opposite association was identified. Eleven of the children were 2 weeks of age or less, and of 105 rib heads available for analysis in these eleven children, only three clefts were identified and all within one child who was 2 weeks of age. The three rib head clefts identified are those with eosinophilic material within the cleft space, acute clefts were also identified. In contrast to this apparent lack of clefts in children less than or equal to 2 weeks of age, eight of the children were greater than 9 months of age and had 77 rib heads available for analysis, among which were found 42 clefts. A test of equal proportions, using the prop.test function in R, comparing the proportion of rib heads with a cleft between the two age groups indicated a significant p-value of 5.647^{-15}; therefore, the age at death is a significant factor in determining the number of clefts found in a child; however age at death was not associated with the number of clefts with a length of greater than 1.00 mm. To highlight this, one child, who was older than 9 months of age had 10 clefts (found in 10 rib heads), and none were greater than 1.00 mm length.

In the review of the literature, risk factors for rib fractures were discussed. The most common situations that children are exposed to which are a risk fracture for fractures are CPR, prematurity, birth, and, albeit, not very common, other metabolic bone disease (i.e., other than prematurity). In this study, 83 of 90 children received CPR and 10 of 90 children had acute anterior rib fractures. Of the children who had acute anterior rib fractures, two died under suspicious circumstances, so, reasonably, 8 of 83 children who received CPR developed acute anterior rib fractures, which is in agreement with Dolinak (2007). However, when the Wilcoxon Rank Sum test and Welch two-sample t-test were used to compare the number of clefts per child to children receiving CPR versus those that did not receive CPR, no statistical significance was
identified. Also, when a Chi-square and Fisher’s exact test were used to compare children who had or had not received CPR to those who had or did not have acute clefts of the anterior rib head, CMLs with no or scant hemorrhage, or CMLs with cleft material, no significant p-values were identified; therefore, the distribution of children among these categories is similar. The proportion of the sample population that did not have CPR was small ($n=6$); however, these results indicate that CPR is not a contributor to the development of the clefts (either acute or with cleft material) or CMLs (either with scant or no hemorrhage or with cleft material).

In the same manner as just described for those infants who had or did not have CPR, comparisons of number of clefts per child and number of clefts per child that were greater than 1.00 mm in length each against children who were born vaginally versus those born via Cesarean section were conducted using Chi-square, Fisher’s exact test, Wilcoxon Rank Sum, Welch’s two-sample t-test, and Poisson regression analysis. Almost no p-values of $<0.05$ were obtained. Only when the mean number of clefts in children in Subgroups A, B, and D from Group 3, using the 10R+ dataset, were compared between vaginal and Cesarean section deliveries with the Welch’s two-sample t-test was a borderline insignificant p-value obtained (0.03749). As the preference of the author is to favor non-parametric testing in these analysis, and as no combination revealed a significant p-value, these results indicate that the method by which a child is born (vaginal versus Cesarean) is not a significant contributor to the number of clefts in the anterior portion of the rib head, or to the production of clefts with a length of greater than 1.00 mm. Using a Chi-square and Fisher’s exact test, the presence or absence of the three types of remote fractures identified grossly was compared to birth method (vaginal versus Cesarean) and presence or absence of a difficult delivery. Once again, no p-value of less than 0.05 was
obtained in any test. However, the number of children who had a difficult delivery (i.e., either because of shoulder dystocia, the need for repositioning or the need for assistance in delivery such as forceps or vacuum extraction) was small ($n=4$).

Although these results indicate that method of delivery (vaginal versus Cesarean) and difficulty of delivery are not significant contributors to the number of clefts per child or the size of clefts, at least two cases in the sample question this conclusion. One child, who died from SIDS, had shoulder dystocia at delivery, with several remote fractures being identified at autopsy, including a right rib head, three rib ribs, and the right clavicle. Although rib fractures are not strongly associated with birth trauma, fractures of the clavicle are, and given that all fractures are on the right side, birth trauma secondary to shoulder dystocia, causing fracture of the right clavicle and right ribs, would appear to be a reasonable explanation. One child, who was one-month-old who died while bed-sharing with adults had (among nine rib heads) one rib head cleft that was healing and had a length greater than 1.00 mm. Given the young age of the infant, and the healing/healed nature of the rib head cleft, the possibility that this cleft is associated with delivery is good. Finally, one child who died at birth had four acute clefts. If an acute cleft is not an artifact, but instead a precursor to the clefts filled with eosinophilic amorphous material, the only two possible causes were birth and CPR. Also, although vaginal versus Cesarean section methods of delivery were compared as to their possible effects in producing these clefts, birth versus no birth could not, obviously, be compared. Perhaps both vaginal and Cesarean section deliveries have an equal chance of producing these rib head clefts, and thus, no statistically significant difference between the two methods was identified.
Regarding an association between prematurity and the presence of clefts, a Chi square and Fisher’s exact test were conducted using the 10R+ sample, and comparing children with and without an estimated gestational age (EGA) of less than 37 weeks and less than 33 weeks with no clefts and one or more clefts, and with no to two clefts and greater than two clefts. Of these 12 tests, only the comparison of EGA of less than 37 weeks and presence or absence of clefts indicated p-values of less than 0.10, with the Chi-square and Fisher’s exact test indicating insignificant p-values of 0.09692 and 0.05543 respectively. However, as described above, a Spearman’s correlation comparing the number of clefts per child, number of clefts greater than 1.00 mm in length per child, and number of clefts greater than 0.501 mm in length per child against various combinations of Groups and Subgroups, and the 10R and 10R+ datasets, indicated p-values of <0.05 for several correlations, with one p-value being <0.01. Poisson regression analysis did not indicate that estimated gestational age was a significant factor in determining the number of clefts per child, or the number of clefts greater than 1.00 mm in length per child. These results indicate that the prematurity may or may not play a role in the production of the clefts, and does not offer strong support either way, although the results would appear to favor that prematurity is not a significant risk for the clefts.

The effects of various socio-economic and demographic factors on the number of clefts per child and number of clefts greater than 1.00 mm in length per child were assessed. These factors included sex of child (male versus female), ancestry of child (only white versus Native American, as the population of Montana is so homogeneous, and these two ancestry groups represent 99% of the population), marital status of mother, age of mother, and the role of the male figure (biological father versus boyfriend). Using Wilcoxon Rank Sum testing, no
significant differences were identified. As statistical analysis did indicate clefts with a length of 1.00 mm or greater were more likely to be found in children who died under suspicious circumstances and, thus, the possibility of abuse, the above results, showing no association of these various factors with the number of clefts greater than 1.00 mm in length per child, would appear to be at odds with Lauer et al. (1974), Schnitzer and Ewigman (2005), and Zhou et al. (2006) who opined or demonstrated a relationship between the young age of parents and abuse, un-married mothers and abuse, and the presence of an unrelated adult in the household (e.g., boyfriend) and abuse. However, given the relatively small sample population in this study, the retrospective nature of the review, and the incompleteness of investigative records available as well as the subjective nature of interpretation of some of the above factors, the presence or absence of an association between the number of clefts per child and the number of clefts per child greater than 1.00 mm in length and marital status of the mother, age of the parents, and role of father-figure in household, should be reserved until further studies have been performed.

Although Zumwalt and Fanizza-Orphanos (1990) have provided a guide to the histologic examination of fractures, the lack of subperiosteal bone formation (e.g., callus) in metaphyseal lesions as described by O’Conner and Cohen (1987) and Kleinman (2008) causes a different progression of healing changes to occur in these rib head clefts, and this study, with its large collection of clefts, provides guidance in understanding the progression of such healing changes. In examining the clefts, the various stages of healing could be approximated. The acute cleft was seen as a break in the primary spongiosa at the anterior edge of the growth plate oriented oblique to the plane of the growth plate. This cleft becomes filled with an amorphous, somewhat granular, acellular, eosinophilic material. Review of the literature does not provide an answer as
to what this material may be; however, most likely it represents by-products of the traumatic injuries and inflammatory response that would be elicited by a fracture of the bone, and may be a combination of fibrin, necrotic bone and cartilage, and necrotic cells. After the cleft becomes filled with this amorphous eosinophilic material, osteoclasts migrate to the site, often forming a small rim between the cleft material and the periosteum. Healing is a variable combination of fibrosis, osteoclast and osteoblast proliferation, and production of woven bone and cartilage. The healing appears to start at the distal aspect of the cleft, producing infilling of the cleft. The timing of the various transitions is not determined, and, given that there was a statistically significant correlation between the number of large clefts and the number of clefts that were healing, indicating that large clefts may heal more rapidly than small clefts, any determination of timing of healing of these clefts based solely upon histologic features is unreliable.
CHAPTER 6: CONCLUSION

Although it is doubtful that the hypotheses have a name, in the medical and forensic literature, rib fractures, especially posterior rib fractures, are strongly associated with child abuse. A related hypothesis upon which this association of infant rib fractures and abuse is most likely based is the belief that infant ribs are difficult if not nearly impossible to break, and would require great force, such as could occur during an abusive acts directed at the child, but not during normal play. Unfortunately, instead of rigorous testing of these hypotheses, relatively untested dogma has apparently developed, and, unfortunately, this dogma can guide the practice of forensic anthropologists, pediatricians, forensic pathologists, and any others who deal with child abuse, whether the victim is living or deceased. The results of this current research indicate that infant ribs are actually much easier to fracture than thought, although the result may not always be grossly or radiologically visible. Caffey (1972), Knight (1996), and Miller (1999) have suggested that rough and even routine handling of an infant or child has the potential to produce fractures under the right situations. One child of greater than 12 months of age in this study had 10 clefts identified in the 10 rib heads examined. Only one cleft had evidence of significant healing, indicating that the other nine most likely occurred relatively recently, or at least, developed in the interim since birth. Scene investigation and autopsy identified nothing suspicious for abuse. Unless abuse of children is so prevalent and so subtle that much is being missed, this single case lends support to the idea that non-abusive rough-handling occurring during playing with children may cause microscopic fractures. For example, it is not uncommon for parents to throw infants into the air and catch them, potentially compressing the chest in the process. Acts such as this, deemed socially acceptable, may actually constitute the rough-
handling causing microscopic fractures. While the importance of the identification of microscopic fractures of the rib head and neck may not currently be deemed of importance (largely because an understanding of their cause is not apparent), as medical imaging techniques improve and smaller defects are able to be identified, the need to understand the etiology of and interpret the significance of these findings will increase.

The practical applications of this study are currently limited but important. Prior to this current study, only Kleinman et al. (1992) had studied rib head fractures, and even then, only essentially as they were found in abused children. However, the current research has shown how microscopic examination of the rib heads in children at the time of autopsy can contribute information that may help the medical examiner better understand the case. A forensic anthropologist consultant with such knowledge can offer insight as to how better to examine for skeletal injuries at the time of autopsy. First of all, the removal of the rib heads and microscopic examination allows for detection of abnormalities not found by gross inspection or postmortem imaging. For example, the finding of a cleft with a length greater than 0.501 mm is potentially significant; however, in the sample employed here, two children had a postmortem CT scan, each of which missed clefts, with lengths of 0.662 mm and 0.760 mm, and two acute anterior shaft fractures. Also, four infants had postmortem skeletal surveys in the hospital prior to being brought for examination by the forensic pathologist, which missed clefts of length 1.003 mm, 1.124 mm, 1.315 mm, and 3.417 mm (one each in the four children), and four acute anterior shaft fractures and one acute rib head fracture in one of the children. If it is important to identify as many injuries as possible, histologic examination in addition to gross examination is important. In one child, five remote fractures of the rib head/neck were identified grossly but
seven were found microscopically, including one healing CML. And, one child had eight acute CMLs with cleft material found microscopically and none grossly. This knowledge may help encourage forensic anthropologists, who might otherwise be opposed to destruction of the bone, to suggest histologic examination. In addition, CMLs with cleft material, not suspected upon gross examination, were found in six other infants at the time of histologic examination. Although metaphyseal lesions in the ribs have not been specifically described, these CMLs with cleft material look exactly like metaphyseal lesions, and, if metaphyseal lesions are highly associated with child abuse, the importance of their identification in the ribs is important. Although Kleinman et al. (1992) advocated caution when interpreting isolated rib head fractures, the same authors, among others, highlight the importance of identification of metaphyseal lesions in the long bones. Their caution should be interpreted not as a lack of belief in isolated rib head fractures as a sign of abuse, but instead as a call for further investigation. In contrast to Kleinman et al. (1992), Malcolm (2008) illustrated a metaphyseal lesion of the rib, indicating it was a sign of non-accidental injury. Second, this current research serves as a basis for further investigation. Although 90 children is a large sample, the subjective nature of cause and manner of death determination and the lack of complete history on all children impairs interpretations; however, certain conclusions can be drawn.

In summary, based upon the review of the literature in combination with the results of this research, several points can be made. 1) In the literature, rib fractures, especially those of the posterior region of the rib, which would include the rib head, are strongly associated with inflicted injury. 2) Cartilage outgrowths are associated with past trauma, but, at the very anterior edge of the rib head, cartilage outgrowths can be present at birth. 3) The clefts identified at the
anterior portion of the metaphysis are not described in the literature as a normal morphologic finding or as a variant of normal, and appear as fractures histologically. Given these two points, the clefts represent fractures and not an anatomic or developmental variant. 4) The clefts are common, with 66 of 90 children in the sample having at least one cleft. 5) Statistical analysis indicates the numbers of clefts per child are unequally distributed amongst subgroup categories of the cause and manner of death. The statistical significance depends upon several factors; however, when comparing suspicious and non-suspicious deaths, there is a statistically significant (p-value of <0.01) difference in the distribution of number of clefts per child between the two groups. 6) Statistical analysis also indicates that the number of clefts with a length of greater than 1.00 mm, and even a length of greater than 0.501 mm per child is unequally distributed amongst subgroup categories of the cause and manner of death, being more common in the group of children represented by the combined total of a) those children who died of known inflicted trauma or b) those children who died from an undetermined cause and manner of death that was regarded as suspicious. Although the difference between some subgroups is significant (e.g., suspicious vs. SIDS), when just comparing the number of rib head clefts with a length of greater than 1.00 mm per child between suspicious deaths (including NAI) and non-suspicious deaths, there was a statistically significant (p-value of <0.01) difference. Note that, as the number of clefts with a length of greater than 1.00 mm present per child and the number of remote fractures of the lateral shaft are correlated, and as the presence of a remote fracture of the lateral shaft identified grossly at autopsy can influence the determination of the cause and manner of death, this conclusion could be interpreted as being biased, alternatively, as the microscopic rib head clefts are associated with suspicious deaths, it can lend support to the idea
that lateral rib fractures, if not otherwise explained, are associated with suspicious deaths. Important is that children with a remote fracture of the lateral portion of the rib shaft were more likely to have a rib head cleft of greater than 1.00 mm in length. 7) Children with more clefts are more likely to have a cleft with a length of greater than 1.00 mm. 8) There is no compelling statistical evidence that the clefts are associated with either CPR or birth method; however, based upon review of individual cases, an argument can be made that the birth method and possibly CPR may cause clefts. 9) An association with EGA, with a negative correlation between EGA and number of rib head clefts per child, was identified in an analysis of a limited portion of the overall sample, but not via analysis of other limited portions of the sample or via Poisson regression analysis. The fact that a negative correlation was present would support the idea that prematurity is a risk factor for these rib head clefts, as, as EGA goes up (i.e., child becomes less premature), the correlation is with fewer rib head fractures. 10) No association between number of clefts or number of clefts greater than 1.00 mm in length per child and socio-economic factors, including presence of biological father versus boyfriend in child’s life, was identified. In addition, the presence of the rib head clefts, either by number or by size, had no statistical association with sex of the child, marital status of the mother, ancestry of the child, or with the use of drugs by the parents; however, a prospective review of these associations instead of a retrospective review, and its inherent limitations in data collection, may alter such conclusions. 11) The number of clefts correlated with the age of the child, and with the number of acute microscopic clefts per child. 12) Based upon their location, the clefts are similar to an incomplete metaphyseal lesion, a finding that is commonly associated with child abuse. Like metaphyseal lesions, the clefts are fractures occurring in the metaphysis; however, they are very
common. Metaphyseal lesions with cleft material were much less common in the sample population. And, finally, 13) the statistical analysis of the data was complex, given the relative subjectivity of cause and manner of death determination, and unequal numbers of rib heads available for histologic analysis per child, which could not be adequately corrected by weighting the sample.

Based upon these findings, it can be said that microscopic review of the rib head and neck of left and right ribs #5-9 can provide useful information, and allow for detection of abnormalities that would otherwise be missed. The finding of a cleft with a length of greater than 1.00 mm is cause for concern, and without a documented cause, is suspicious. Whether or not a difficult vaginal delivery can cause such a fracture is not certain, although, based upon single cases within the sample, it seems possible; however, with a history of a routine delivery (i.e., no shoulder dystocia, or re-positioning, or forceps needed), and without history of other trauma, the finding of microscopic rib head fractures may assist investigators in questioning caretakers regarding the handling of the child, as they likely indicate a past or relatively recent episode of forceful compression of the chest. This information is of importance to practicing forensic anthropologists, to help in the evaluation of skeletal trauma, as well as to forensic pathologists; however, any professional who requires knowledge of patterns of child abuse in the skeletal system may benefit. Symes et al. (1996) advocated and demonstrated the benefits of close working relationships between medical examiners and forensic anthropologists in the evaluation of skeletal trauma. Recent articles by Love and Sanchez (2009), Love et al. (2013), Backo and Love (2013), Pinto et al. (2013), and Love et al. (2014) indicate that, at least in some jurisdictions, such relationships are working. Microscopic analysis of rib heads, because of the
necessary preparation, including decalcification, is relatively time consuming, and something that many forensic pathologists may not be willing to add to their busy schedule; however, consulting forensic anthropologists may be able to provide such a service.

Based upon the data collected, no association between the various socio-economic factors examined and the number of clefts per child, or number of clefts with a length of greater than 1.00 mm per child could be substantiated, despite evidence in the literature of the links between unmarried females, unrelated adults (e.g., boyfriend), and drug use with child abuse (three factors addressed in this current research) (Lauer et al., 1974; Schnitzer and Ewigman, 2005; Zhou et al. 2006). The relatively limited sample may be responsible for any lack of association identified with these factors in this study. Also, with some factors (e.g., drug use, cleanliness of house, living quarters, and even relationship of male to child), subjective interpretation by me when collecting data, or by the coroner when reporting the data, or incomplete or inconsistent collection of data, may be responsible. However, another possibility worthy of discussion is that certain risk factors for child abuse, when viewed from a global or societal perspective, such as in a study of 100,000 individuals, will become apparent; however, when viewed from the perspective of one individual, there is so much variation within a group regarding that risk factor and its association with abuse, that any affects are not apparent in a smaller sample, and, especially not based upon any one child.

Although an understanding of social and economic factors contributing to the development of a disease is important, forensic anthropologists and forensic pathologists as well as other physicians must always remember that they work with individuals, whether skeletal remains requiring interpretation of the circumstances of their arrival at that state, the body of a
deceased requiring an autopsy, or a patient requiring a diagnosis and treatment. In this regard, theoretical applications and applied knowledge are sometimes at odds. Turshen (1977:49) states, “The overwhelming concern with the individual is a major limitation of the paradigm of clinical medicine.” However, one must only understand the concept of slow and rapid metabolization of medication to understand that statement is unwarranted. Slow and rapid metabolizers break down a medication either more slowly or more rapidly, respectively, than normal. Knowledge of an individual’s state is of importance when prescribing them certain medications as a rapid metabolizer will clear a medication from their blood sooner than normal and may require a larger dose to achieve therapeutic effect, and a slow metabolizer will clear a medication from their blood slower than normal and may require a smaller dose to achieve therapeutic effect. A patient’s response to proton pump inhibitors or opiate therapy are only two examples of this important need to treat patient’s as individuals (Dickson and Stuart, 2003; Matin et al., 2007)

This difference between theoretical applications and applied knowledge may also been seen in the relative importance of biological reductionism as perceived by cultural anthropologists and physicians. Singer (1998:95) states that “political economy is both old and new. The term is of fresh vintage in the sense that it constitutes an emergent approach that is looked to as a needed corrective to the reductionism of the recent past, especially within anthropology.” However, Malkin (1993:2), a clinical pathologist, states

It is the thesis of the author that progress in biology and medicine and the refutation of a “vital force” has occurred because scientists since 1800 have been able to explain normal and abnormal living phenomena at lower and lower levels of organisms, starting with organ systems and ultimately being able to discuss life in terms of cell membranes and even molecules themselves. This method has been called reductionism by philosophers of science, and this approach has been eminently successful.
To appreciate the differences between an ecological approach and an individualized approach, one must merely examine the goals of the scientist involved. The goal of Zhou et al. (2006:54), with their ecological approach to identifying perinatal risk factors, was “to develop a population-based model that enables public health agencies to identify areas at high risk for infant maltreatment.” Zhou et al. (2006:54) also stated, “Many studies fail to consider an ecological, population-based method that explores why these individual risk factors occur in the larger context of the environment in which they are found.” Connell-Carrick and Scannapieco (2006) describe that “the ultimate purpose of neglect research is to improve the quality of the lives of those whom it affects.” Both authors address the need to improve society at large, an admirable and desirable position; however, neither necessarily advocates nor addresses the importance of one child, or the likelihood of any one single child to sustain injuries.

The importance of one’s position in this regard is obvious—i.e., is one concerned with society or with the individual? For example, just because in society in general a young, unmarried mother with less than a high school education, who is on Medicaid and living with her boyfriend is more commonly associated with child abuse does not mean that in a given case that a young, unmarried mother with less than a high school education, who is on Medicaid and living with her boyfriend has a deceased child who has sustained fatal abuse. So, while an ecological approach to disease certainly benefits public health policy and other aspects, it would fail when applied at the individual level. In other words, generalizations can work well when applied to populations at large; however, generalizations when applied to an individual can cause more harm than good. For example, society as a whole can appreciate the benefits in vaccination of its children, with the result being a great decrease in some infectious diseases (e.g., measles)
to eradication of others (e.g., small pox); however, for the few parents who have a lost a child to a post-vaccination encephalitis, the benefits of vaccination to the individual is not always apparent. Forensic anthropologists and forensic pathologists understand this fact, and realize that each case they investigate must be evaluated as a unique situation, and they must not fall victim to using generalizations to dictate their final decision.

In conclusion, this research identified that microscopic rib head fractures of greater than 0.501 mm in length are statistically significantly associated with a combined group of children representing either diagnosed fatal non-accidental injury or for whom there was a strong suspicion of non-accidental injury being the cause of death. Assuming that chest compression can cause rib head fractures, and that chest compression is a relatively common component of the various episodes of abuse inflicted upon an infant or young child, identification of microscopic rib head fractures can indicate the strong possibility of past or recent episodes of abuse. No definitive association was identified with CPR, birth method, or estimated gestational age. Without examination of this critical area of the body at autopsy, these findings would otherwise be missed. And, given the utility they may have when used by law enforcement to question caretakers of the child at the time of death, a reasonable search for them at the time of autopsy by forensic pathologists or their forensic anthropologist consultants would be prudent. The finding of a rib head fracture of greater than 0.501 mm in length, or being more conservative, a length of greater than 1.00 mm, or multiple such fractures, without a reasonable explanation, such as past compressive chest trauma or difficult delivery, is cause for concern, as it may indicate past abusive chest compression.
BIBLIOGRAPHY


US Department of Health & Human Services. 2006. Does the HIPAA Privacy rule require documentation of Institutional Review Board (IRB) or Privacy Board approval of an alteration or waiver of individual authorization before a covered entity may use or disclose protected health information for any of the following provisions: (1) for preparatory research at 45 CFR 164.512 (i)(1)(ii), (2) for research on the protected health information of decedents at 45 CFR 164.512(i)(1)(iii), or (3) a limited data set with a data use agreement as stipulated at 45 CFR 164.51. Washington, DC.[2013 Feb 24]. Available from: http://www.dhhs.gov/hipaafaq/permitted/research/318.html


### Appendix A

**Appendix A1: Information collected from review of tissue slides.**

<table>
<thead>
<tr>
<th>Category</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of left and right ribs sampled</td>
<td></td>
</tr>
<tr>
<td>Number of left and right rib heads available for analysis</td>
<td></td>
</tr>
<tr>
<td>Number of left and right transverse process regions available for analysis</td>
<td></td>
</tr>
<tr>
<td>Number of microscopic acute fractures of the rib head/neck, shaft of the rib, or clavicle</td>
<td></td>
</tr>
<tr>
<td>Number of microscopic remote fractures of the rib head/neck, shaft of the rib, or clavicle</td>
<td></td>
</tr>
<tr>
<td>Number of clefts of the rib head (with amorphous eosinophilic material)</td>
<td></td>
</tr>
<tr>
<td>Extent of healing of clefts</td>
<td>- Cleft extends to periosteum</td>
</tr>
<tr>
<td></td>
<td>- Thin rim of osteoclasts, or other cellular or extracellular material between cleft and periosteum</td>
</tr>
<tr>
<td></td>
<td>- Pronounced healing with woven bone, cartilage, or fibrosis</td>
</tr>
<tr>
<td></td>
<td>- Number of osteoclasts present</td>
</tr>
<tr>
<td>Measurements of the cleft</td>
<td>- Distance of tip of cleft at growth plate to the anterior edge of the growth plate</td>
</tr>
<tr>
<td></td>
<td>- Distance from anterior edge of growth plate to proximal end of base of cleft at periosteum</td>
</tr>
<tr>
<td></td>
<td>- Distance from anterior edge of growth plate to distal end of base of cleft at periosteum</td>
</tr>
<tr>
<td></td>
<td>- Length of cleft (from tip at growth plate to periosteum)</td>
</tr>
<tr>
<td></td>
<td>- Area of cleft</td>
</tr>
<tr>
<td>Presence of cartilage outgrowths</td>
<td>- Size and shape</td>
</tr>
<tr>
<td></td>
<td>- Location</td>
</tr>
<tr>
<td>Number of acute clefts (with no amorphous eosinophilic material)</td>
<td></td>
</tr>
<tr>
<td>Number of metaphyseal lesions</td>
<td>- With no or only scant hemorrhage</td>
</tr>
<tr>
<td></td>
<td>- With amorphous eosinophilic material</td>
</tr>
<tr>
<td>Number of Salter-Harris fractures</td>
<td></td>
</tr>
<tr>
<td>Presence of reduced cellularity</td>
<td></td>
</tr>
</tbody>
</table>
Appendix A2. Information collected from autopsy file.
Age (in months) at death
Birth type (vaginal or Cesarean)
Estimated gestational age at birth
Was CPR performed at time of death (yes/no)?
Who was CPR performed by?
Cause of death
Manner of death
If cause and manner of death were both undetermined, was death suspicious for inflicted injury?
Number of gross acute fractures of the
rib head/neck, lateral shaft, or anterior shaft
Presence of acute fractures of a clavicle
Number of gross remote fractures of the
rib head/neck, lateral shaft, or anterior shaft
Presence of remote fractures of a clavicle
Socio-economic factors
- Sex and ancestry of child
- Marital status and age of mother
- Relationship of male to child, and his age
- Type of residence and cleanliness of residence
- Were parents foster parents
- Was there history of parental drug use
Appendix B1. Data collection sheet--original

<table>
<thead>
<tr>
<th>Age (months):</th>
<th>Cause of death:</th>
</tr>
</thead>
<tbody>
<tr>
<td>Delivery:</td>
<td>Manner of death:</td>
</tr>
<tr>
<td>CPR (Y/N):</td>
<td>If undetermined, suspicious:</td>
</tr>
</tbody>
</table>

### Type of CPR:

<table>
<thead>
<tr>
<th># of ribs</th>
<th>Le</th>
<th>Right</th>
<th>Le</th>
<th>Right</th>
<th>No side</th>
</tr>
</thead>
<tbody>
<tr>
<td># of rib heads</td>
<td>Le</td>
<td>Right</td>
<td>Le</td>
<td>Right</td>
<td>No side</td>
</tr>
<tr>
<td># of TP regions</td>
<td>Le</td>
<td>Right</td>
<td>Le</td>
<td>Right</td>
<td>No side</td>
</tr>
</tbody>
</table>

X = None

<table>
<thead>
<tr>
<th>Rib head or neck</th>
<th>Shaft of rib</th>
<th>Clavicle</th>
</tr>
</thead>
</table>

Number of gross acute fractures

Number of microscopic acute fractures

Number of gross remote fractures

Number of microscopic remote fractures

<table>
<thead>
<tr>
<th>Left</th>
<th>Right</th>
<th>No side</th>
</tr>
</thead>
</table>

Number of rib heads with fractured spongiosa and no hemorrhage

Number of rib heads with fractured spongiosa and hemorrhage

### Rib head clefts

<table>
<thead>
<tr>
<th>Side</th>
<th>C/S</th>
<th>Inner tip</th>
<th>Prox</th>
<th>Distal</th>
<th>Length</th>
<th>Area</th>
<th>OC</th>
<th>Woven</th>
</tr>
</thead>
</table>

**Key**

C/S = Cartilage or spongiosa; Inner tip = inner tip to end of cartilage; prox = proximal corner of base of cleft to cartilage; distal = distal corner of base of cleft to cartilage; OC = # of osteoclasts; Woven = woven bone Y/N

### Cartilage outgrowths

Left | Right | Size | Location

### Reduced cellularity

Anterior versus posterior
<table>
<thead>
<tr>
<th>Age in months:</th>
<th>Cause of death:</th>
</tr>
</thead>
<tbody>
<tr>
<td>Delivery method:</td>
<td></td>
</tr>
<tr>
<td>EGA:</td>
<td>Manner of death:</td>
</tr>
<tr>
<td>CPR (Yes/No):</td>
<td>Suspicious (Yes/No):</td>
</tr>
<tr>
<td>At least EMTs (Yes/No):</td>
<td>Explain:</td>
</tr>
<tr>
<td>Number of ribs:</td>
<td>Number of rib heads if 6 randomized:</td>
</tr>
<tr>
<td>Number of rib heads:</td>
<td>Number of rib heads if 10 randomized:</td>
</tr>
<tr>
<td>Healing? OC</td>
<td>To periosteum Length Tip Incl 6 rand? Incl 10 rand? Photos</td>
</tr>
</tbody>
</table>

Additional findings:
APPENDIX C

Example of randomization procedure

Infant #52 had 17 ribs removed at autopsy (nine from left side and eight from right side), and of these ribs, 14 rib heads were available for microscopic analysis (six from left side and eight from right side). Clefts were identified on four left ribs and on three right ribs.

To randomize for six ribs, the R function, sample (1:9,3, replace=False), was used to determine which of the 3 of the 9 left ribs available would be in the random sample, e.g., #1, 4, and 8, and again for which 3 of the 8 right ribs available would be in the random sample, e.g., #1, 3, 6. However, of the nine left ribs and eight right ribs in the original sample, the rib head was not available for analysis on all; so, to determine whether or not, the random rib sampled also had a rib head available for analysis, sample (1:9,6,replace=False) was used to determine which 6 of the original 9 left ribs had a rib head available for analysis, e.g, #1, 3, 4, 6, 7, 9. So, since the 3 ribs in the randomized sample are #1, 4, 8, but rib #8 is not among the group of six that had a rib head available for analysis, the random sample only has two left sided ribs with a rib head available for analysis. This process was not needed on the right side since all eight ribs sampled had a rib head available for analysis. Therefore, in the six ribs random sample, five rib heads are available for analysis.

The six left sided rib heads yielded four clefts, so sample(1:6,4,replace=False) was used to determine which—e.g., if the four numbers were 2, 3, 5, 6, then the 2nd, 3rd, 5th, and 6th left sided rib with a rib head available for analysis had a cleft. In this case left ribs #3, 4, 7, and 9. Since the two ribs in the sample were #1 and 4, only the cleft on the left 4th rib could be included in the statistical analyses that followed.

The eight right sided rib heads yielded three clefts, so sample (1:8,3,replace=False) was used to determine which—e.g., if the three numbers were #3, 4, and 7; then the 3rd, 4th, and 7th right sided rib with a rib head available for analysis had a cleft. In this case, since all eight ribs originally sampled had a rib head available for analysis, the 3rd, 4th and 7th ribs directly correspond to the right ribs #3, 4, and 7. Since the three ribs sampled were # 1, 3, and 6, only the cleft on the right 3rd rib could be included in the statistical analyses that followed.

In this case, if infant #52 would have only six of the original 17 ribs removed at autopsy available for examination, there would have been five rib heads instead of 14, and two clefts identified instead of seven.

The general procedure for producing a 10 rib randomized sample was the same, but using five ribs on each side instead of three.
APPENDIX D

R script

#Script to read in general overall data
ribs<-read.csv("FinalSpreadsheet3A.csv",header=T)
hist(ribs$Z,main="Number of clefts per child",xlab="Number of clefts", breaks=0:11-0.5)

#Script to extract general numbers, means, etc from overall data
agemonths<-(ribs$B)
agemonths
sd(agemonths)
mean(agemonths)
summary(agemonths)
hist(agemonths, xlab="Age in months", main="Age In Months", breaks=0:21-0.5)
ega<-(ribs$AG)
sd(ega,na.rm=T)
mean(ega,na.rm=T)
summary(ega,na.rm=T)
hist(ega, xlab="Estimated gestational age",ylab="Frequency", main="EGA histogram",breaks=25:45-0.5)

#Testing of SIDS versus bed-sharing
egasids<-ribs$AG[ribs$E=="SIDS"]
egasids
mean(egasids)
egabedshare<-ribs$AG[ribs$E=="BedShare"]
egabedshare
mean(egabedshare,na.rm=T)
agemonthssids<-ribs$B[ribs$E=="SIDS"]
mean(agemonthssids)
sd(agemonthssids)
agemonthsbedshare<-ribs$B[ribs$E=="BedShare"]
mean(agemonthsbedshare)
sd(agemonthsbedshare)
t.test(agemonthssids,agemonthsbedshare, paired=F)
agemonthsbedsharecond<-ribs$B[ribs$E=="BedShare"&ribs$B<10]
mean(agemonthsbedsharecond)
sd(agemonthsbedsharecond)
t.test(agemonthssids,agemonthsbedsharecond,paired=F)
par(mfrow=c(1,2))
boxplot(agemonthssids, main="Age in months-SIDS deaths")
boxplot(agemonthsbedshare, main="Age in months-bed share deaths ")
par(mfrow=c(1,1))
wilcox.test(agemonthssids,agemonthsbedshare,paired=F)

#Figure38a histogram
allribheadsfreq<-sort(ribs$I)
allribheadsfreq
hist(allribheadsfreq,xlab="Number of rib heads per child", main="Number of rib heads to evaluate", breaks=0:25-0.5)

#Table 4 calculations
grp1clfts<-ribs$J[ribs$E4==1]
grp1clfts
length(grp1clfts)
mean(grp1clfts)
sd(grp1clfts)
hist(grp1clfts, xlab="Number of clefts", main="COD=SIDS")
boxplot(grp1clfts)
grp2clfts<-ribs$J[ribs$E4==2]
grp2clfts
length(grp2clfts)
mean(grp2clfts)
sd(grp2clfts)
hist(grp2clfts, xlab="Number of clefts", main="COD=Bed Share")
boxplot(grp2clfts)
grp3clfts<-ribs$J[ribs$E4==3]
grp3clfts
length(grp3clfts)
mean(grp3clfts)
sd(grp3clfts)
hist(grp3clfts, xlab="Number of clefts", main="COD=NAI and suspicious")
boxplot(grp3clfts)
grp4clfts<-ribs$J[ribs$E4==4]
grp4clfts
length(grp4clfts)
mean(grp4clfts)
sd(grp4clfts)
hist(grp4clfts, xlab="Number of clefts", main="COD=Natural, not SIDS")
boxplot(grp4clfts)
grp5clfts<-ribs$J[ribs$E4==5]
grp5clfts
length(grp5clfts)
mean(grp5clfts)
sd(grp5clfts)
hist(grp5clfts, xlab="Number of clefts", main="COD=Other")
boxplot(grp5clfts)

# Frequency of clefts, with 6 ribs randomized; Table 4 calculations
sixribheadsfreq<-sort(ribs$Q)
sixribheadsfreq
hist(sixribheadsfreq, xlab="Number of rib heads per infant", main="Number of rib heads to evaluate")
grp1clfts<-ribs$R[ribs$E4==1]
grp1clfts
length(grp1clfts)
mean(grp1clfts)
sd(grp1clfts)
hist(grp1clfts, xlab="Number of clefts", main="COD=SIDS")
boxplot(grp1clfts)
grp2clfts<-ribs$R[ribs$E4==2]
grp2clfts
length(grp2clfts)
mean(grp2clfts)
sd(grp2clfts)
hist(grp2clfts, xlab="Number of clefts", main="COD=Bed Share")
boxplot(grp2clfts)
grp3clfts<-ribs$R[ribs$E4==3]
grp3clfts
length(grp3clfts)
mean(grp3clfts)
sd(grp3clfts)
hist(grp3clfts, xlab="Number of clefts", main="COD=NAI and suspicious")
boxplot(grp3clfts)
grp4clfts<-ribs$R[ribs$E4==4]
grp4clfts
length(grp4clfts)
mean(grp4clfts)
sd(grp4clfts)
hist(grp4clfts, xlab="Number of clefts", main="COD=Natural, not SIDS")
boxplot(grp4clfts)
grp5clfts<-ribs$R[ribs$E4==5]
grp5clfts
length(grp5clfts)
mean(grp5clfts)
sd(grp5clfts)
hist(grp5clfts, xlab="Number of clefts", main="COD=Other")
boxplot(grp5clfts)
tenribheadsfreq<-sort(ribs$Y)
tenribheadsfreq
hist(tenribheadsfreq,xlab="Number of rib heads per infant", main="Number of rib heads to evaluate")
grp1clfts<-ribs$Z[ribs$E3==1]
grp1clfts
length(grp1clfts)
mean(grp1clfts)
sd(grp1clfts)
hist(grp1clfts, xlab="Number of clefts", main="COD=SIDS")
boxplot(grp1clfts)
grp2clfts<-ribs$Z[ribs$E3==2]
grp2clfts
length(grp2clfts)
mean(grp2clfts)
sd(grp2clfts)
hist(grp2clfts, xlab="Number of clefts", main="COD=Bed Share")
boxplot(grp2clfts)
grp3clfts<-ribs$Z[ribs$E3==3]
grp3clfts
length(grp3clfts)
mean(grp3clfts)
sd(grp3clfts)
hist(grp3clfts, xlab="Number of clefts", main="COD=NAI and suspicious")
boxplot(grp3clfts)
grp4clfts<-ribs$Z[ribs$E4==4]
grp4clfts
length(grp4clfts)
mean(grp4clfts)
sd(grp4clfts)
hist(grp4clfts, xlab="Number of clefts", main="COD=Known natural")
boxplot(grp4clfts)
grp5clfts<-ribs$Z[ribs$E4==5]
grp5clfts
length(grp5clfts)
mean(grp5clfts)
sd(grp5clfts)
hist(grp5clfts, xlab="Number of clefts", main="COD=Other")
boxplot(grp5clfts)

#Frequency of clefts, with 10 ribs randomized and >7 rib heads; Table 4 calculations
tenribheadsfreq<-sort(ribs$Y)
tenribheadsfreq
hist(tenribheadsfreq,xlab="Number of rib heads per infant", main="Number of rib heads to evaluate")
grp1clfts<-ribs$Z[ribs$E3==1&ribs$Y>7]
grp1clfts
length(grp1clfts)
mean(grp1clfts)
sd(grp1clfts)
hist(grp1clfts, xlab="Number of clefts", main="COD=SIDS")
boxplot(grp1clfts)
grp2clfts<-ribs$Z[ribs$E1==2&ribs$Y>7]
grp2clfts
length(grp2clfts)
mean(grp2clfts)
sd(grp2clfts)
hist(grp2clfts, xlab="Number of clefts", main="COD=Bed Share")
boxplot(grp2clfts)
grp3clfts<-ribs$Z[ribs$E3==3&ribs$Y>7]
grp3clfts
length(grp3clfts)
mean(grp3clfts)
sd(grp3clfts)
hist(grp3clfts, xlab="Number of clefts", main="COD=NAI and suspicious")
boxplot(grp3clfts)
grp4clfts<-ribs$Z[ribs$E4==4&ribs$Y>7]
grp4clfts
length(grp4clfts)
mean(grp4clfts)
sd(grp4clfts)
hist(grp4clfts, xlab="Number of clefts", main="COD=Known natural")
boxplot(grp4clfts)
grp5clfts<-ribs$Z[ribs$E4==5&ribs$Y>7]
grp5clfts
length(grp5clfts)
mean(grp5clfts)
sd(grp5clfts)
hist(grp5clfts, xlab="Number of clefts", main="COD=Other")
boxplot(grp5clfts)

#Script to extract certain cases--specifically groups 1-4
ribs<-read.csv("FinalSpreadsheet3A.csv",header=T)
testgrps<-ribs$E5<5
testgrps
testgrps1<-ribs[testgrps,]
testgrps1
length(testgrps1$Z)

#KruskalWallisTestComparingNumbersOfClefts

#KruskalWallisComparing all groups and groups 1-4 across all ribs
kruskal.test(ribs$J~ribs$E5)
kruskal.test(testgrps1$J~testgrps1$E5)
#KruskalWallisComparing all groups and groups 1-4 using 6 random ribs
kruskal.test(ribs$R~ribs$E5)
kruskal.test(testgrps1R~testgrps1$E5)
#KruskalWallisComparing all groups and Groups1-4 using 10 random ribs
kruskal.test(ribs$Z~ribs$E5)
kruskal.test(testgrps1Z~testgrps1$E5)
#KruskalWallisComparing all groups and Groups1-4 using 10 random ribs and 8-11 heads
testgrps2<-ribs$Y>7
testgrps3<-ribs[testgrps2,]
testgrps2a<-ribs$E5<5&ribs$Y>7
testgrps3a<-ribs[testgrps2a,]
kruskal.test(testgrps3$Z~testgrps3$E5)
kruskal.test(testgrps3a$Z~testgrps3a$E5)

#KruskalWallisComparing groups 1-4 across all ribs for number of clefts <1 mm in length
kruskal.test(ribs$K~ribs$E5)
kruskal.test(testgrps1$K~testgrps1$E1)
#KruskalWallisComparing groups 1-4 using 6 random ribs
kruskal.test(ribs$S~ribs$E5)
kruskal.test(testgrps1$S~testgrps1$E1)
#KruskalWallisComparing Groups1-4 using 10 random ribs
kruskal.test(ribs$AA~ribs$E5)
kruskal.test(testgrps1AA~testgrps1$E1)
#KruskalWallisComparing Groups1-4 using 10 random ribs and 8-11 heads
testgrps2<-ribs$E1<5&ribs$Y>7
testgrps3<-ribs[testgrps2,]
testgrps3
length(testgrps3)
length(testgrps3$B)
kruskal.test(testgrps3$AA~testgrps3$E1)

#KruskalWallisComparing all groups and groups 1-4 across all ribs for number of clefts >1 mm in length
kruskal.test(ribs$L~ribs$E5)
kru skal.test(testgrps1$L~testgrps1$E5)
#KruskalWallisComparing groups 1-4 using 6 random ribs
kruskal.test(ribs$T~ribs$E5)
kru skal.test(testgrps1$T~testgrps1$E5)
#KruskalWallisComparingGroups1-4 using 10 random ribs
kruskal.test(ribs$AB2~ribs$E1)
kru skal.test(testgrps1$AB2~testgrps1$E1)

#KruskalWallisComparing all groups and Groups1-4 using 10 random ribs and 8-11 heads and number of clefts >1 mm in length
testgrps2<-ribs$Y>7
testgrps3<-ribs[testgrps2,]
testgrps2a<-ribs$E5<5&ribs$Y>7
testgrps3a<-ribs[testgrps2a,]
kruskal.test(testgrps3$AB2~testgrps3$E5)
kruskal.test(testgrps3a$AB2~testgrps3a$E5)

#Script to read in general overall data
ribs<-read.csv("FinalSpreadsheet2.csv",header=T)

#T Test comparing number of clefts vaginal and Cesarean deliveries in 10 rib head sample, and 8-11 rib heads
cleftsvag<-ribs$Z[ribs$C=="Vag"&ribs$Y>7]
cleftscesar<-ribs$Z[ribs$C=="Cesar"&ribs$Y>7]
length(cleftsvag)
length(cleftscesar)
wilcox.test (cleftsvag,cleftscesar, paired=F)
t.test(cleftsvag,cleftscesar, paired=F)

#T Test comparing number of clefts >1 mm between vaginal and Cesarean deliveries in 10 rib head sample, and 8-11 rib heads
cleftsvag<-ribs$AB[ribs$C=="Vag"&ribs$Y>7]
cleftscesar<-ribs$AB[ribs$C=="Cesar"&ribs$Y>7]
wilcox.test (cleftsvag,cleftscesar, paired=F)
t.test(cleftsvag,cleftscesar, paired=F)

#T Test comparing number of clefts >.501 mm between vaginal and Cesarean deliveries in 10 rib head sample, and 8-11 rib heads
cleftsvag<-ribs$AB2[ribs$C=="Vag"&ribs$Y>7]
cleftscesar<-ribs$AB2[ribs$C=="Cesar"&ribs$Y>7]
wilcox.test (cleftsvag,cleftscesar, paired=F)
t.test(cleftsvag,cleftscesar, paired=F)
#T test comparing CPR and no CPR

cleftsCPR <- ribs$Z[(ribs$D == "Yes" & ribs$Y > 7)]
cleftsnoCPR <- ribs$Z[(ribs$D == "No" & ribs$Y > 7)]
length(cleftsCPR)
length(cleftsnoCPR)
t.test(cleftsCPR, cleftsnoCPR)

# cannot do t test--not enough infants with no CPR
# Wilcoxon
wilcox.test(cleftsCPR, cleftsnoCPR, paired = F)

## Above comparing clefts and such in non-suspicious deaths using Groups 1, 2, and 4 in E1, E3-E5 Groups

# Extract cases with 8-11 ribs
ribs <- read.csv("FinalSpreadsheet2.csv", header = T)

# T Test comparing number of clefts vaginal and Cesarean deliveries in 10 rib head sample, and 8-11 rib heads

cleftsvag <- ribs$Z[(ribs$E5 == 1 | ribs$E5 == 2 | ribs$E5 == 4) & ribs$C == "Vag" & ribs$Y > 7]
cleftscesar <- ribs$Z[(ribs$E5 == 1 | ribs$E5 == 2 | ribs$E5 == 4) & ribs$C == "Cesar" & ribs$Y > 7]
length(cleftsvag)
length(cleftscesar)
wilcox.test(cleftsvag, cleftscesar, paired = F)
t.test(cleftsvag, cleftscesar, paired = F)

# T Test comparing number of clefts >1 mm between vaginal and Cesarean deliveries in 10 rib head sample, and 8-11 rib heads

cleftsvag <- ribs$AB[(ribs$E5 == 1 | ribs$E5 == 2 | ribs$E5 == 4) & ribs$C == "Vag" & ribs$Y > 7]
cleftscesar <- ribs$AB[(ribs$E5 == 1 | ribs$E5 == 2 | ribs$E5 == 4) & ribs$C == "Cesar" & ribs$Y > 7]
wilcox.test(cleftsvag, cleftscesar, paired = F)
t.test(cleftsvag, cleftscesar, paired = F)

# T Test comparing number of clefts >0.501 mm between vaginal and Cesarean deliveries in 10 rib head sample, and 8-11 rib heads

cleftsvag <- ribs$AB2[(ribs$E5 == 1 | ribs$E5 == 2 | ribs$E5 == 4) & ribs$C == "Vag" & ribs$Y > 7]
cleftscesar <- ribs$AB2[(ribs$E5 == 1 | ribs$E5 == 2 | ribs$E5 == 4) & ribs$C == "Cesar" & ribs$Y > 7]
wilcox.test(cleftsvag, cleftscesar, paired = F)
t.test(cleftsvag, cleftscesar, paired = F)

# T test comparing CPR and no CPR

cleftsCPR <- ribs$Z[(ribs$E5 == 1 | ribs$E5 == 2 | ribs$E5 == 4) & ribs$D == "Yes" & ribs$Y > 7]
cleftsnoCPR <- ribs$Z[(ribs$E5 == 1 | ribs$E5 == 2 | ribs$E5 == 4) & ribs$D == "No" & ribs$Y > 7]
length(cleftsCPR)
length(cleftsnoCPR)
t.test(cleftsCPR,cleftsnoCPR)
#cannot do t test--not enough infants with no CPR
#Wilcoxon
wilcox.test(cleftsCPR,cleftsnoCPR,paired=F)

#comparing EGA and clefts in 10 ribs randomized and 10 ribs randomized with 8-11 ribs for all infants
ega<-ribs$AG
clefts<-ribs$AB2
length(ega)
cor.test(clefts,ega,method="spearman")
ega8to11<-ribs$AG[ribs$Y>7]
clefts8to11<-ribs$AB2[ribs$Y>7]
length(ega8to11)
cor.test(clefts8to11,ega8to11,method="spearman")

#comparing EGA and clefts in 10 ribs randomized and 10 ribs randomized with 8-11 ribs in various groups
ribs<-read.csv("FinalSpreadsheet2.csv",header=T)
natega<-ribs$AG[ribs$E5==1|ribs$E5==4]
natclefts<-ribs$AB2[ribs$E5==1|ribs$E5==4]
length(natega)
cor.test(natclefts,natega,method="spearman")
ega8to11<-ribs$AG[(ribs$E5==1|ribs$E5==4)&ribs$Y>7]
clefts8to11<-ribs$AB2[(ribs$E5==1|ribs$E5==4)&ribs$Y>7]
length(ega8to11)
cor.test(clefts8to11,ega8to11,method="spearman")

#ExtractingInfants who are non-suspicious and have 8-11 ribs
NonSusp<-!(ribs$E1==1|ribs$E1==2|ribs$E1==4)&ribs$Y>7
NonSusp1<-ribs[NonSusp,]
length(NonSusp1$Z)
cor.test(NonSusp1$Z,NonSusp1$AG,method="spearman")

#TestingForEachIndividualRib
indrib<-read.csv("EachRibIndividual.csv",header=T)
mean(indrib$B)
sd(indrib$B)
range(indrib$B)
hist(indrib$B, xlab="Age in months", main="Age in months of infant with rib with fractures")
length(indrib$B)
healrib<-((indrib$G=="Yes")
healrib1<-indrib[healrib,]
healrib1
length(healrib1$A)
range(healrib1$B)
rimrib<-((indrib$H=="Yes")
rimrib1<-indrib[rimrib,]
rimrib1
length(rimrib1$A)
range(rimrib1$B)
periostrib<-((indrib$I=="Yes")
periostrib1<-indrib[periostrib,]
periostrib1
length(periostrib1$A)
range(periostrib1$B)

# Size of rib clefts
mean(indrib$E)
sd(indrib$E)
range(indrib$E)
hist(indrib$E, xlab="Length of cleft (in mm)", main="Size of clefts")
boxplot(indrib$E)
mean(indrib$F)
sd(indrib$F)
range(indrib$F)
hist(indrib$F, xlab="Distance of cleft from growth plate (in mm)", main="Location of clefts")
boxplot(indrib$F)

# Bedshare intoxicated versus not-intoxicated
ribs<-read.csv("FinalSpreadsheet2.csv",header=T)
bedshare1<-ribs$E=="BedShare"
bedshare2<-ribs[bedshare1,]
length(bedshare2$A)
b edsharenotintox<-((ribs$E=="BedShare"&ribs$E2=="No"&ribs$Y>7)
bedshareintox<-((ribs$E=="BedShare"&ribs$E2=="Yes"&ribs$Y>7)
bedsharenotintox1<-ribs[bedsharenotintox,]
bedshareintox1<-ribs[bedshareintox,]
length(bedsharenotintox1$A)
length(bedshareintox1$A)
wilcox.test(bedsharenotintox1$B,bedshareintox1$B,paired=F)
wilcox.test(bedsharenotintox1$J,bedshareintox1$J,paired=F)
wilcox.test(bedsharenotintox1$L,bedshareintox1$L,paired=F)
wilcox.test(bedsharenotintox1$Z,bedshareintox1$Z,paired=F)
wilcox.test(bedsharenointox1$AB, bedsharetintox1$AB, paired=F)

# Comparing the individual groups
# Code for E1 groups
ribs <- read.csv("FinalSpreadsheet3A.csv", header=T)
sidstest <- (ribs$E1 == 1)
sidstest1 <- ribs[sidstest,]
besharetest <- (ribs$E1 == 2)
besharetest1 <- ribs[besharetest,]
susptest <- (ribs$E1 == 3)
susptest1 <- ribs[susptest,]
natetest <- (ribs$E1 == 4)
natetest1 <- ribs[natetest,]
othertest <- (ribs$E1 == 5)
othertest1 <- ribs[othertest,]

# Code for E3 groups
ribs <- read.csv("FinalSpreadsheet2.csv", header=T)
sidstest <- (ribs$E3 == 1)
sidstest1 <- ribs[sidstest,]
besharetest <- (ribs$E3 == 2)
besharetest1 <- ribs[besharetest,]
susptest <- (ribs$E3 == 3)
susptest1 <- ribs[susptest,]
natetest <- (ribs$E3 == 4)
natetest1 <- ribs[natetest,]
othertest <- (ribs$E3 == 5)
othertest1 <- ribs[othertest,]

# Code for E4 groups
ribs <- read.csv("FinalSpreadsheet2.csv", header=T)
sidstest <- (ribs$E4 == 1)
sidstest1 <- ribs[sidstest,]
besharetest <- (ribs$E4 == 2)
besharetest1 <- ribs[besharetest,]
susptest <- (ribs$E4 == 3)
susptest1 <- ribs[susptest,]
natetest <- (ribs$E4 == 4)
natetest1 <- ribs[natetest,]
othertest <- (ribs$E4 == 5)
othertest1 <- ribs[othertest,]

# Code for E5 groups
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
sidstest<-(ribs$E5==1)
sidstest1<-ribs[sidstest,]
bedsharetest<-(ribs$E5==2)
bedsharetest1<-ribs[bedsharetest,]
susptest<-(ribs$E5==3)
susptest1<-ribs[susptest,]
nattest<-(ribs$E5==4)
nattest1<-ribs[nattest,]
othertest<-(ribs$E5==5)
othertest1<-ribs[othertest,]

#Code for comparing number of clefts
wilcox.test(sidstest1$Z,bedsharetest1$Z,paired=F)
wilcox.test(sidstest1$Z,conf.int=TRUE)
wilcox.test(bedsharetest1$Z,conf.int=TRUE)
wilcox.test(sidstest1$Z,susptest1$Z,paired=F)
wilcox.test(sidstest1$Z,nattest1$Z,paired=F)
wilcox.test(sidstest1$Z,othertest1$Z,paired=F)
wilcox.test(bedsharetest1$Z,susptest1$Z,paired=F)
wilcox.test(bedsharetest1$Z,nattest1$Z,paired=F)
wilcox.test(bedsharetest1$Z,othertest1$Z,paired=F)
wilcox.test(nattest1$Z,susptest1$Z,paired=F)
wilcox.test(nattest1$Z,othertest1$Z,paired=F)
wilcox.test(susptest1$Z,othertest1$Z,paired=F)

pval1<-c(0.7545,0.08275,0.3965,0.05431,0.1105,0.2498,0.03054,0.06283,0.2559, 0.01685)
p.adjust(pval1,method="bonferroni",n=10)

#Code for comparing number of clefts >1.0 mm
wilcox.test(sidstest1$AB,bedsharetest1$AB,paired=F)
wilcox.test(sidstest1$AB,susptest1$AB,paired=F)
wilcox.test(sidstest1$AB,nattest1$AB,paired=F)
wilcox.test(sidstest1$AB,othertest1$AB,paired=F)
wilcox.test(bedsharetest1$AB,susptest1$AB,paired=F)
wilcox.test(bedsharetest1$AB,nattest1$AB,paired=F)
wilcox.test(bedsharetest1$AB,othertest1$AB,paired=F)
wilcox.test(nattest1$AB,susptest1$AB,paired=F)
wilcox.test(nattest1$AB,othertest1$AB,paired=F)
wilcox.test(susptest1$AB,othertest1$AB,paired=F)

#Code for comparing number of clefts >0.501 mm
wilcox.test(sidstest1$AB2,bedsharetest1$AB2,paired=F)
wilcox.test(sidstest1$AB2,susptest1$AB2,paired=F)
wilcox.test(sidstest1$AB2,nattest1$AB2,paired=F)
wilcox.test(sidstest1$AB2,othertest1$AB2,paired=F)
wilcox.test(bedsharetest1$AB2,susptest1$AB2,paired=F)
wilcox.test(bedsharetest1$AB2,nattest1$AB2,paired=F)
wilcox.test(bedsharetest1$AB2,othertest1$AB2,paired=F)
wilcox.test(nattest1$AB2,susptest1$AB2,paired=F)
wilcox.test(nattest1$AB2,othertest1$AB2,paired=F)
wilcox.test(susptest1$AB2,othertest1$AB2,paired=F)

#Correlations

#Extract data
ribs<-read.csv("FinalSpreadsheet3A.csv",header=T)
EightTo11<-ribs$Y>7&(ribs$E5==1|ribs$E5==2|ribs$E5==4|ribs$E5==5)
EightTo11a<-ribs[EightTo11,]
length(EightTo11a$B)
cor.test(EightTo11a$AD,EightTo11a$AR,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$AT,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$AV,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$AG,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$AL,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$AN,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$AP,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$AD,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$AJ,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$AX1,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$AZ1,method="spearman")
cor.test(EightTo11a$AD,EightTo11a$BB1,method="spearman")

#ProportionTestForShoulderDystociaVsSIDS,BedShare,andNatural
ribs<-read.csv("FinalSpreadsheet2.csv",header=T)
cleftswithsd<-ribs$Z[(ribs$E1==1|ribs$E1==2|ribs$E1==4)&ribs$C1=="Yes"&ribs$Y>7]
cleftswithoutsd<-ribs$Z[(ribs$E1==1|ribs$E1==2|ribs$E1==4)&ribs$C1=="No"&ribs$Y>7]
ribheadswithsd<-ribs$Y[(ribs$E1==1|ribs$E1==2|ribs$E1==4)&ribs$C1=="Yes"&ribs$Y>7]
ribheadswithoutsd<-ribs$Y[(ribs$E1==1|ribs$E1==2|ribs$E1==4)&ribs$C1=="No"&ribs$Y>7]
cleftsyes<-sum(cleftswithsd)
cleftsno<-sum(cleftswithoutsd)
clefts<-c(cleftsyes,cleftsno)
ribheadyes<-sum(ribheadswithsd)
ribheadno<-sum(ribheadswithoutsd)
ribheads<-c(ribheadyes,ribheadno)
#Histograms

hist(ribs$I, main="Total number of rib heads for analysis per child", xlab="Number of rib heads per child", breaks=0:25-0.5)

hist(ribs$Y, main="Total number of rib heads for analysis in 10 rib sample", xlab="Number of rib heads per child", breaks=0:15-0.5)

hist(ribs$J, main="Total number of clefts per child to evaluate", xlab="Number of clefts per child", breaks=0:15-0.5)

hist(ribs$Z, main="Total number of clefts in 10 rib sample per child", xlab="Number of clefts per child", breaks=0:15-0.5)

#Determinations of >1 mm and >.501 mm clefts

ribs<-read.csv("FinalSpreadsheet3A.csv", header=T)

length(ribs$L)

sum(ribs$L)

hist(ribs$L, main="Number of clefts >1.00 mm per child in All ribs sample", xlab="Number of clefts >1.00 mm per child", breaks=0:10-0.5)

length(ribs$AB)

sum(ribs$AB)

hist(ribs$AB, main="Number of clefts >1.00 mm per child in 10 ribs randomized sample", xlab="Number of clefts >1.00 mm per child", breaks=0:10-0.5)

length(ribs$AB2)

sum(ribs$AB2)

hist(ribs$AB2, main="Number of clefts >.501 mm per child in 10 ribs randomized sample", xlab="Number of clefts >.501 mm per child", breaks=0:10-0.5)

#PoissonRegression

fit<-glm(ribs$Z~ribs$B+ribs$C+ribs$D+ribs$E1, family=poisson)

summary(fit)

#Comparison of mean number of clefts between Groups 1-5 in All ribs sample and 10 ribs randomized, 8-11 ribs sample

ribs<-read.csv("FinalSpreadsheet2.csv", header=T)

sidstestE5all<-(ribs$E5==1)

sidstestE5all1<-ribs[sidstestE5all,]

sidstestE5and8<-(ribs$E5==1&ribs$Y>7)

sidstestE5and81<-ribs[sidstestE5and8,]

bedsharetestE5all<-(ribs$E5==2)

bedsharetestE5all1<-ribs[bedsharetestE5all,]

bedsharetestE5and8<-(ribs$E5==2&ribs$Y>7)

bedsharetestE5and81<-ribs[bedsharetestE5and8,]

susptestE5all<-(ribs$E5==3)
susptestE5all1<-ribs[susptestE5all,]
susptestE5and8<-(ribs$E5==3&ribs$Y>7)
susptestE5and81<-ribs[susptestE5and8,]
nattestE5all1<-ribs[nattestE5all,]
nattestE5and8<-(ribs$E5==4&ribs$Y>7)
nattestE5and81<-ribs[nattestE5and8,]
othertestE5all1<-ribs[othertestE5all,]
othertestE5and8<-(ribs$E5==5&ribs$Y>7)
othertestE5and81<-ribs[othertestE5and8,]
wilcox.test(sidstestE5all1$J,sidstestE5and81$Z,paired=F)
wilcox.test(bedsharetestE5all1$J,bedsharetestE5and81$Z,paired=F)
wilcox.test(susptestE5all1$J,susptestE5and81$Z,paired=F)
wilcox.test(nattestE5all1$J,nattestE5and81$Z,paired=F)
wilcox.test(othertestE5all1$J,othertestE5and81$Z,paired=F)
wilcox.test(sidstestE5all1$L,sidstestE5and81$AB,paired=F)
wilcox.test(bedsharetestE5all1$L,bedsharetestE5and81$AB,paired=F)
wilcox.test(susptestE5all1$L,susptestE5and81$AB,paired=F)
wilcox.test(nattestE5all1$L,nattestE5and81$AB,paired=F)
wilcox.test(othertestE5all1$L,othertestE5and81$AB,paired=F)

#Comparison of mean number of clefts all ribs vs 6 and 10 randomized
ribs<-read.csv("FinalSpreadsheet2.csv",header=T)
sidstestE5all<-(ribs$E5==1)
sidstestE5all1<-ribs[sidstestE5all,]
sidstestE5and8<-(ribs$E5==1)
sidstestE5and81<-ribs[sidstestE5and8,]
bedsharetestE5all<-(ribs$E5==2)
bedsharetestE5all1<-ribs[bedsharetestE5all,]
bedsharetestE5and8<-(ribs$E5==2)
bedsharetestE5and81<-ribs[bedsharetestE5and8,]
susptestE5all<-(ribs$E5==3)
susptestE5all1<-ribs[susptestE5all,]
susptestE5and8<-(ribs$E5==3)
susptestE5and81<-ribs[susptestE5and8,]
nattestE5all<-(ribs$E5==4)
nattestE5all1<-ribs[nattestE5all,]
nattestE5and8<-(ribs$E5==4)
nattestE5and81<-ribs[nattestE5and8,]
othertestE5all<-(ribs$E5==5)
othertestE5all1<-ribs[othertestE5all,]
othertestE5and8<-(ribs$E5==5)
othertestE5and81<-ribs[othertestE5and81]

wilcox.test(sidstestE5all1$J,sidstestE5and81$R,paired=F)
wilcox.test(bedsharetestE5all1$J,bedsharetestE5and81$R,paired=F)
wilcox.test(susptestE5all1$J,susptestE5and81$R,paired=F)
wilcox.test(nattestE5all1$J,nattestE5and81$R,paired=F)
wilcox.test(othertestE5all1$J,othertestE5and81$R,paired=F)
wilcox.test(sidstestE5all1$L,sidstestE5and81$T,paired=F)
wilcox.test(bedsharetestE5all1$L,bedsharetestE5and81$T,paired=F)
wilcox.test(susptestE5all1$L,susptestE5and81$T,paired=F)
wilcox.test(nattestE5all1$L,nattestE5and81$T,paired=F)
wilcox.test(othertestE5all1$L,othertestE5and81$T,paired=F)

#SelectionOfInfantsWithVariousGrossFractures
ribs<-read.csv("FinalSpreadsheet2.csv",header=T)
fracturesGARH<-(ribs$AK=="Yes")
fracturesGARH1<-ribs[fracturesGARH,]
fracturesGARH

fracturesGAAS<-(ribs$AM=="Yes")
fracturesGAAS1<-ribs[fracturesGAAS,]
fracturesGAAS

fracturesGALS<-(ribs$AO=="Yes")
fracturesGALS1<-ribs[fracturesGALS,]
fracturesGALS

fracturesGRRH<-(ribs$AQ=="Yes")
fracturesGRRH1<-ribs[fracturesGRRH,]
fracturesGRRH

fracturesGRAS<-(ribs$AS=="Yes")
fracturesGRAS1<-ribs[fracturesGRAS,]
fracturesGRAS

fracturesGRLS<-(ribs$AU=="Yes")
fracturesGRLS1<-ribs[fracturesGRLS,]
fracturesGRLS

mean(fracturesGRLS1$AG)
sd(fracturesGRLS1$AG)

#FisherExactTestsForCounts
ribs<-read.csv("FinalSpreadsheet2.csv",header=T)
#Number of infants in 5 E1 groups (10 ribs randomized) with clefts >1mm
largeclefts<-matrix(c(14,18,6,15,10,5,12,8,1,1),nr=5)
chisq.test(largeclefts)
fisher.test(largeclefts)
#Number of infants in 5 E1 groups (10 ribs randomized) with clefts
clefts<-matrix(c(4,6,3,5,6,15,24,11,11,5),nr=5)
chisq.test(clefts)
fisher.test(clefts)
# Number of clefts between vag and Cesarean

cleftsbirth<-matrix(c(7,14,18,44),nr=2)
chisq.test(cleftsbirth)
fisher.test(cleftsbirth)

# Number of clefts >1.00 mm between vag and Cesarean

largecleftsbirth<-matrix(c(20,38,5,20),nr=2)
chisq.test(largecleftsbirth)
fisher.test(largecleftsbirth)

# Number of acute clefts between 5 E1 groups 10 randomized

aclefts<-matrix(c(16,23,13,6,3,7,1,3,5),nr=5)
chisq.test(aclefts)
fisher.test(aclefts)

# Number of acute clefts between CPR and no CPR

acprclefts<-matrix(c(5,65,1,18),nr=2)
chisq.test(acprclefts)
fisher.test(acprclefts)

# Number of infants E1 groups all 5, 10 rib randomized with CMLs with scant or no hemorrhage

cmlsscant<-matrix(c(19,26,12,15,9,0,4,2,1,2),nr=5)
chisq.test(cmlsscant)
fisher.test(cmlsscant)

# Number of infants with CPR or no CPR and with CMLs with scant or no hemorrhage

cmlsscantCPR<-matrix(c(6,74,0,9),nr=2)
chisq.test(cmlsscantCPR)
fisher.test(cmlsscantCPR)

# Number of infants E1 groups all 5, 10 rib randomized with CMLs with cleft material

cmls<-matrix(c(17,29,10,16,11,2,14,0,0),nr=5)
chisq.test(cmls)
fisher.test(cmls)

# Number of infants with CPR or no CPR and with CMLs with cleft material

cmlsCPR<-matrix(c(76,6,7,0),nr=2)
chisq.test(cmlsCPR)
fisher.test(cmlsCPR)

# All infants and chi square comparing E5 groups and number of infants with one cleft or more, 10 ribs randomized

ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum<-matrix(c(4,6,3,3,8,16,24,10,7,9),nr=5)
chisq.test(cleftsnum)
fisher.test(cleftsnum)
#All infants and chi square comparing E5 groups and number of infants with more than 2 clefts
10 ribs randomized
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum2<-matrix(c(11,17,4,7,14,9,13,9,3,3),nr=5)
chisq.test(cleftsnum2)
fisher.test(cleftsnum2)

#All infants and chi square comparing E5 groups and number of infants with clefts >1.00 mm 10
ribs randomized
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum3<-matrix(c(14,18,6,10,15,6,12,7,0,2),nr=5)
chisq.test(cleftsnum3)
fisher.test(cleftsnum3)

#Chi square comparing E5 groups and number of infants with clefts, using 10 ribs randomized, 8-11 ribs
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum4<-matrix(c(3,5,1,3,5,16,22,10,4,8),nr=5)
chisq.test(cleftsnum4)
fisher.test(cleftsnum4)

#Chi square comparing E5 groups and number of infants with >2 clefts, using 10 ribs randomized, 8-11 ribs
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum5<-matrix(c(10,16,2,4,10,9,11,9,3,3),nr=5)
chisq.test(cleftsnum5)
fisher.test(cleftsnum5)

#Chi square comparing E5 groups and number of infants with cleft >1.00mm in length, using 10
ribs randomized, 8-11 ribs
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum6<-matrix(c(13,16,4,7,12,6,11,7,0,1),nr=5)
chisq.test(cleftsnum6)
fisher.test(cleftsnum6)

#Chi square comparing E5 groups, using 8-11 ribs, and comparing gross acute fractures of rib
head/neck
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum7<-matrix(c(19,25,7,7,13,0,2,4,0,0),nr=5)
chisq.test(cleftsnum7)
fisher.test(cleftsnum7)
Chi square comparing E5 groups, using 8-11 ribs, and comparing gross acute fractures of anterior shaft
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum8<-matrix(c(15,23,9,7,13,4,4,2,0,0),nr=5)
chisq.test(cleftsnum8)
fisher.test(cleftsnum8)

Chi square comparing E5 groups, using 8-11 ribs, and comparing gross acute fractures of lateral ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum9<-matrix(c(19,27,10,7,13,0,0,1,0,0),nr=5)
chisq.test(cleftsnum9)
fisher.test(cleftsnum9)

Chi square comparing E5 groups, using 8-11 ribs, and comparing gross remote fractures of rib head/neck
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum10<-matrix(c(18,24,8,7,12,1,3,3,0,1),nr=5)
chisq.test(cleftsnum10)
fisher.test(cleftsnum10)

Chi square comparing E5 groups, using 8-11 ribs, and comparing gross remote fractures of anterior shaft
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum11<-matrix(c(18,26,10,7,13,1,1,1,0,0),nr=5)
chisq.test(cleftsnum11)
fisher.test(cleftsnum11)

Chi square comparing E5 groups, using 8-11 ribs, and comparing gross remote fractures of lateral shaft
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
cleftsnum12<-matrix(c(19,27,7,7,13,0,0,4,0,0),nr=5)
chisq.test(cleftsnum12)
fisher.test(cleftsnum12)

Chi square comparing (using 8-11 ribs sample, n=77), infants with >2 clefts, and infants with at least one cleft >1.00 mm in length
cleftssize<-matrix(c(34,8,18,17),nr=2)
chisq.test(cleftssize)
fisher.test(cleftssize)

Chi square comparing (using 8-11 ribs sample, n=77), infants with a clefts >1.00 mm and with gross acute fracture of rib head/neck
cleftssize2<-matrix(c(50,21,2,4),nr=2)
chisq.test(cleftssize2)
fisher.test(cleftssize2)

# Chi square comparing (using 8-11 ribs sample, n=77), infants with a clefts >1.00 mm and with
gross acute fracture of anterior shaft of rib
cleftssize3<-matrix(c(46,21,6,4),nr=2)
chisq.test(cleftssize3)
fisher.test(cleftssize3)

# Chi square comparing (using 8-11 ribs sample, n=77), infants with a clefts >1.00 mm and with
gross acute fracture of lateral shaft of rib
cleftssize4<-matrix(c(52,24,0,1),nr=2)
chisq.test(cleftssize4)
fisher.test(cleftssize4)

# Chi square comparing (using 8-11 ribs sample, n=77), infants with a clefts >1.00 mm and with
gross remote fracture of rib head/neck
cleftssize5<-matrix(c(49,20,3,5),nr=2)
chisq.test(cleftssize5)
fisher.test(cleftssize5)

# Chi square comparing (using 8-11 ribs sample, n=77), infants with a clefts >1.00 mm and with
gross remote fracture of anterior shaft
cleftssize6<-matrix(c(50,24,2,1),nr=2)
chisq.test(cleftssize6)
fisher.test(cleftssize6)

# Chi square comparing (using 8-11 ribs sample, n=77), infants with a clefts >1.00 mm and with
gross remote fracture of lateral shaft
cleftssize7<-matrix(c(52,21,0,4),nr=2)
chisq.test(cleftssize7)
fisher.test(cleftssize7)

# Chi square comparing (n=83) infants with birth type to gross remote fracture of rib head/neck
cleftpreg<-matrix(c(23,52,2,6),nr=2)
chisq.test(cleftpreg)
fisher.test(cleftpreg)

# Chi square comparing (n=83) infants with shoulder dystocia to gross remote fracture of rib
head/neck
cleftpreg2<-matrix(c(72,3,7,1),nr=2)
chisq.test(cleftpreg2)
fisher.test(cleftpreg2)
#Chi square comparing (n=83) infants with birth type to gross remote fracture of anterior shaft
cleftpreg3<-matrix(c(25,55,0,3),nr=2)
chisq.test(cleftpreg3)
fisher.test(cleftpreg3)

#Chi square comparing (n=83) infants with shoulder dystocia to gross remote fracture of anterior shaft
cleftpreg4<-matrix(c(77,3,2,1),nr=2)
chisq.test(cleftpreg4)
fisher.test(cleftpreg4)

#Chi square comparing (n=83) infants with birth type to gross remote fracture of lateral shaft
cleftpreg5<-matrix(c(25,54,0,4),nr=2)
chisq.test(cleftpreg5)
fisher.test(cleftpreg5)

#Chi square comparing (n=83) infants with shoulder dystocia to gross remote fracture of lateral shaft
cleftpreg6<-matrix(c(75,4,4,0),nr=2)
chisq.test(cleftpreg6)
fisher.test(cleftpreg6)

#Chi square comparing infants with 8-11 ribs in 10 randomized EGA<37 weeks or >or=37 weeks and presence of cleft
cleftEGA<-matrix(c(14,41,0,13),nr=2)
chisq.test(cleftEGA)
fisher.test(cleftEGA)

#Chi square comparing infants with 8-11 ribs in 10 randomized EGA<37 weeks or >or=37 weeks and presence of > 2 clefts
cleftEGA1<-matrix(c(30,25,4,9),nr=2)
chisq.test(cleftEGA1)
fisher.test(cleftEGA1)

#Chi square comparing infants with 8-11 ribs in 10 randomized EGA<37 weeks or >or=37 weeks and presence of cleft >1.00mm
cleftEGA2<-matrix(c(36,19,8,5),nr=2)
chisq.test(cleftEGA2)
fisher.test(cleftEGA2)

#Chi square comparing infants with 8-11 ribs in 10 randomized EGA<33 weeks or >or=34 weeks and presence of cleft
cleftEGA3<-matrix(c(14,53,0,1),nr=2)
chisq.test(cleftEGA3)
fisher.test(cleftEGA3)

#Chi square comparing infants with 8-11 ribs in 10 randomized EGA<33 weeks or >or=34 weeks and presence of > 2 clefts
cleftEGA4<-matrix(c(30,25,4,9),nr=2)
chisq.test(cleftEGA4)
fisher.test(cleftEGA4)

#Chi square comparing infants with 8-11 ribs in 10 randomized EGA<33 weeks or >or=34 weeks and presence of cleft >1.00mm
cleftEGA5<-matrix(c(33,34,1,0),nr=2)
chisq.test(cleftEGA5)
fisher.test(cleftEGA5)

#Differences between 1-4.0 and 4.5-12 months of age
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
ribyoung<-(ribs$B>0.999&ribs$B<4.001&ribs$Y>7)
ribyoung1<-ribs[ribyoung,]
length(ribyoung1$B)
ribold<-(ribs$B>4.001&ribs$B<12.001&ribs$Y>7)
ribold1<-ribs[ribold,]
length(ribold1$B)
wilcox.test(ribold1$AB,ribyoung1$AB,paired=F)

#Proportion test healing and non-healing 1-4.0 months and 4.5-12 months
healing<-c(38,10)
total<-c(105,50)
prop.test(healing,total)

#Proportion test number of clefts in <2 weeks and >9 months
clefts<-c(3,42)
total<-c(105,77)
prop.test(clefts,total)

#Numbers of healing ribs
heal<-read.csv("EachRibIndividual.csv",header=T)
smallheal<-(heal$F<.999&heal$G="Yes")
smallheal1<-heal[smallheal,]
length(smallheal1$B)
smallnotheal<-(heal$F<.999&heal$G="No")
smallnotheal1<-heal[smallnotheal,]
length(smallnotheal1$B)
largeheal<-!(heal$F>.999&heal$G=="Yes")
largeheal1<-heal[largeheal,]
length(largeheal1$B)
largenonheal<-(heal$F>.999&heal$G=="No")
largenonheal1<-heal[largenonheal,]
length(largenonheal1$B)

# Fisher exact test comparing large and small healed and not healed, dividing at .501 mm
healed<-matrix(c(8,60,93,62),nr=2)
chisq.test(healed)
fisher.test(healed)

# Fisher exact test comparing large and small healed and not healed, dividing at 1 mm
healed<-matrix(c(34,34,144,11),nr=2)
chisq.test(healed)
fisher.test(healed)

# Wilcoxon Rank Sum and t-test to compare age of children with acute fractures to age of children without acute fractures and number of clefts in those with acute fractures to those without
ribs<-read.csv("FinalSpreadsheet3.csv",header=T)
acute<-ribs$AM=="Yes"
acute1<-ribs[acute,]
noacute<-ribs$AM=="No"
noacute1<-ribs[noacute,]
wilcox.test(acute1$B,noacute1$B,paired=F)
t.test(acute1$B,noacute1$B)
wilcox.test(acute1$AG,noacute1$AG,paired=F)
t.test(acute1$AG,noacute1$AG)
wilcox.test(acute1$Z,noacute1$Z,paired=F)
t.test(acute1$Z,noacute1$Z)
# Chi-square acute rib head vs acute shaft and remote
frac<-matrix(c(4,2,6,78),nr=2)
chisq.test(frac)
fisher.test(frac)
fracrem<-matrix(c(2,6,1,81),nr=2)
chisq.test(fracrem)
fisher.test(fracrem)

# Poisson Regression
ribs<-read.csv("FinalSpreadsheet3A.csv",header=T)
ribs1<-(ribs$C=="Vag"|ribs$C=="Cesar")&(ribs$D=="Yes"|ribs$D=="No")&(ribs$Y>7))
ribs2<-ribs[ribs1,]
fit<-glm(ribs2$AB~ribs2$B+ribs2$C+ribs2$D+ribs2$AG,family=gaussian)
summary(fit)
fit1<-glm(ribs2$AB~ribs2$E1G,family=poisson)
summary(fit1)

##Final analysis of 8-11+ rib sample with overall calculation
ribs<-read.csv("FinalSpreadsheet3ATest.csv",header=T)
ribs1<-ribs$Y>7
ribs2<-ribs[ribs1,]
length(ribs2$A)

#Delivery method
ribsvag<-ribs2$C=="Vag"
ribsvag1<-ribs2[ribsvag,]
length(ribsvag1$A)
ribscesar<-ribs2$C=="Cesar"
ribscesar1<-ribs2[ribscesar,]
length(ribscesar1$A)

#CPR
ribsyes<-ribs2$D=="Yes"
ribsyes1<-ribs2[ribsyes,]
length(ribsyes1$A)
ribsno<-ribs2$D=="No"
ribsno1<-ribs2[ribsno,]
length(ribsno1$A)

#Age at death and EGA
range(ribs2$B)
median(ribs2$B)
mean(ribs2$B)
wilcox.test(ribs2$B,conf.int=T)
SIGN.test(ribs2$B)
range(ribs2$AG,na.rm=T)
median(ribs2$AG, na.rm=T)
mean(ribs2$AG, na.rm=T)
wilcox.test(ribs2$AG,conf.int=T)
SIGN.test(ribs2$AG)

#Number of clefts and sizes
hist(ribs2$Z, main="Number of rib clefts per child", ylab="Frequency", xlab="Number of clefts", breaks=0:20-0.5)
range(ribs2$Z)
mean(ribs2$Z)
median(ribs2$Z)
wilcox.test(ribs2$Z, conf.int=T)
SIGN.test(ribs2$Z)

hist(ribs2$AB, main="Number of rib clefts with length of >1.00 mm per child", ylab="Frequency", xlab="Number of clefts", breaks=0:20-0.5)
range(ribs2$AB)
mean(ribs2$AB)
median(ribs2$AB)
wilcox.test(ribs2$AB, conf.int=T)
SIGN.test(ribs2$AB)

hist(ribs2$AB2, main="Number of rib clefts with length of >.501 mm per child", ylab="Frequency", xlab="Number of clefts", breaks=0:20-0.5)
range(ribs2$AB2)
mean(ribs2$AB2)
median(ribs2$AB2)
wilcox.test(ribs2$AB2, conf.int=T)
SIGN.test(ribs2$AB2)

# Numbers for Group 3
ribs2SIDS <- ribs2$E4 == "1"
ribs2SIDS2 <- ribs2[ribs2SIDS,]
length(ribs2SIDS2$A)
median(ribs2SIDS2$AG)
mean(ribs2SIDS2$AG)
wilcox.test(ribs2SIDS2$AG, conf.int=T)

hist(ribs2SIDS2$Z, main="Number of rib clefts per child", ylab="Frequency", xlab="Number of clefts", breaks=0:20-0.5)
mean(ribs2SIDS2$Z)
median(ribs2SIDS2$Z)
wilcox.test(ribs2SIDS2$Z, conf.int=T)
sign.test(ribs2SIDS2$Z)

hist(ribs2SIDS2$AB, main="Number of rib clefts with length of >1.00 mm per child", ylab="Frequency", xlab="Number of clefts", breaks=0:20-0.5)
mean(ribs2SIDS2$AB)
median(ribs2SIDS2$AB)
wilcox.test(ribs2SIDS2$AB, conf.int=T)
sign.test(ribs2SIDS2$AB)

hist(ribs2SIDS2$AB2, main="Number of rib clefts with length of >.501 mm per child", ylab="Frequency", xlab="Number of clefts", breaks=0:20-0.5)
mean(ribs2SIDS2$AB2)
median(ribs2SIDS2$AB2)
wilcox.test(ribs2SIDS2$AB2,conf.int=T)
sign.test(ribs2SIDS2$AB2)

ribs2Bed<-ribs2$E4=="2"
ribs2Bed2<-ribs2[ribs2Bed,]
length(ribs2Bed2$A)
median(ribs2Bed2$AG)
mean(ribs2Bed2$AG)
wilcox.test(ribs2Bed2$AG,conf.int=T)
hist(ribs2Bed2$Z,main="Number of rib clefts per child", xlab="Frequency", ylab="Number of clefts",breaks=0:20-0.5)
mean(ribs2Bed2$Z)
median(ribs2Bed2$Z)
wilcox.test(ribs2Bed2$Z,conf.int=T)
sign.test(ribs2Bed2$Z)

ribs2Bed2$AB
hist(ribs2Bed2$AB,main="Number of rib clefts with length of >1.00 mm per child", xlab="Frequency", ylab="Number of clefts",breaks=0:20-0.5)
mean(ribs2Bed2$AB)
median(ribs2Bed2$AB)
wilcox.test(ribs2$AB,conf.int=T)
sign.test(ribs2$AB)

ribs2Susp<-ribs2$E4=="3"
ribs2Susp2<-ribs2[ribs2Susp,]
length(ribs2Susp2$A)
median(ribs2Susp2$AG)
mean(ribs2Susp2$AG)
wilcox.test(ribs2Susp2$AG,conf.int=T)
hist(ribs2Susp2$Z,main="Number of rib clefts per child (Group 3)", ylab="Frequency", xlab="Number of clefts",breaks=0:20-0.5)
range(ribs2Susp2$Z)
mean(ribs2Susp2$Z)
median(ribs2Susp2$Z)
wilcox.test(ribs2Susp2$Z,conf.int=T)
t.test(ribs2Susp2$Z,conf.int=T)
SIGN.test(ribs2Susp2$Z)
hist(ribs2Susp2$AB,main="Number of rib clefts with length of >1.00 mm per child (Group 3)",
ylab="Frequency", xlab="Number of clefts",breaks=0:20-0.5)
range(ribs2Susp2$AB)
mean(ribs2Susp2$AB)
median(ribs2Susp2$AB)
wilcox.test(ribs2Susp2$AB,conf.int=T)
t.test(ribs2Susp2$AB,conf.int=T)
SIGN.test(ribs2Susp2$AB)

hist(ribs2Susp2$AB2,main="Number of rib clefts with length of >.501 mm per child (Group 3)",
xlab="Frequency", ylab="Number of clefts",breaks=0:20-0.5)
range(ribs2Susp2$AB2)
mean(ribs2Susp2$AB2)
median(ribs2Susp2$AB2)
wilcox.test(ribs2Susp2$AB2,conf.int=T)
t.test(ribs2Susp2$AB2,conf.int=T)
SIGN.test(ribs2Susp2$AB2)

hist(ribs2Susp2$AE1,main="Proportion of healing ribs",xlab="Proportion that are healing",ylab="Frequency of children with")
mean(ribs2Susp2$AE1)
median(ribs2Susp2$AE1)
wilcox.test(ribs2Susp2$AE1,conf.int=T)
t.test(ribs2Susp2$AE1,conf.int=T)
SIGN.test(ribs2Susp2$AE1)

ribs2Nat<-ribs2$E4=="4"
ribs2Nat2<-ribs2[ribs2Nat,]
length(ribs2Nat2$A)
median(ribs2Nat2$AG)
mean(ribs2Nat2$AG)
wilcox.test(ribs2Nat2$AG,conf.int=T)
hist(ribs2Nat2$Z,main="Number of rib clefts per child", xlab="Frequency", ylab="Number of clefts",breaks=0:20-0.5)
mean(ribs2Nat2$Z)
median(ribs2Nat2$Z)
wilcox.test(ribs2Nat2$Z,conf.int=T)
sign.test(ribs2Nat2$Z)
hist(ribs2Nat2$AB,main="Number of rib clefts with length of >1.00 mm per child",
xlab="Frequency", ylab="Number of clefts",breaks=0:20-0.5)
mean(ribs2Nat2$AB)
median(ribs2Nat2$AB)
wilcox.test(ribs2Nat2$AB,conf.int=T)
sign.test(ribs2Nat2$AB)
hist(ribs2Nat2$AB2, main="Number of rib clefts with length of >.501 mm per child", xlab="Frequency", ylab="Number of clefts", breaks=0:20-0.5)
mean(ribs2Nat2$AB2)
median(ribs2Nat2$AB2)
wilcoxon.test(ribs2Nat2$AB2, conf.int=T)
sign.test(ribs2Nat2$AB2)

ribs2Other<-ribs2$E4=="5"
ribs2Other2<-ribs2[ribs2Other,]
length(ribs2Other2$A)
median(ribs2Other2$AG)
mean(ribs2Other2$AG)
wilcoxon.test(ribs2Other2$AG, conf.int=T)

hist(ribs2Other2$Z, main="Number of rib clefts per child", xlab="Frequency", ylab="Number of clefts", breaks=0:20-0.5)
mean(ribs2Other2$Z)
median(ribs2Other2$Z)
wilcoxon.test(ribs2Other2$Z, conf.int=T)
sign.test(ribs2Other2$Z)

hist(ribs2Other2$AB, main="Number of rib clefts with length of >1.00 mm per child", xlab="Frequency", ylab="Number of clefts", breaks=0:20-0.5)
mean(ribs2Other2$AB)
median(ribs2Other2$AB)
wilcoxon.test(ribs2Other2$AB, conf.int=T)
sign.test(ribs2Other2$AB)

hist(ribs2Other2$AB2, main="Number of rib clefts with length of >.501 mm per child", xlab="Frequency", ylab="Number of clefts", breaks=0:20-0.5)
mean(ribs2Other2$AB2)
median(ribs2Other2$AB2)
wilcoxon.test(ribs2Other2$AB2, conf.int=T)
sign.test(ribs2Other2$AB2)

ribs2NoSusp<-ribs2$E4B=="NoSusp"
ribs2NoSusp2<-ribs2[ribs2NoSusp,]
length(ribs2NoSusp2$A)
median(ribs2NoSusp2$AG)
mean(ribs2NoSusp2$AG)
wilcoxon.test(ribs2NoSusp2$AG, conf.int=T)

hist(ribs2NoSusp2$Z, main="Number of rib clefts per child (Group 3)", xlab="Frequency", ylab="Number of clefts", breaks=0:20-0.5)
range(ribs2NoSusp2$Z)
mean(ribs2NoSusp2$Z)
median(ribs2NoSusp2$Z)
wilcox.test(ribs2NoSusp2$Z,conf.int=T)
t.test(ribs2NoSusp2$Z,conf.int=T)
SIGN.test(ribs2NoSusp2$Z)
hist(ribs2NoSusp2$AB,main="Number of rib clefts with length of >1.00 mm per child (Group 3)", ylab="Frequency", xlab="Number of clefts", breaks=0:20-0.5)
range(ribs2NoSusp2$AB)
mean(ribs2NoSusp2$AB)
median(ribs2NoSusp2$AB)
wilcox.test(ribs2NoSusp2$AB,conf.int=T)
t.test(ribs2NoSusp2$AB,conf.int=T)
SIGN.test(ribs2NoSusp2$AB)
hist(ribs2NoSusp2$AB2,main="Number of rib clefts with length of >.501 mm per child (Group 3)", ylab="Frequency", xlab="Number of clefts", breaks=0:20-0.5)
range(ribs2NoSusp2$AB2)
mean(ribs2NoSusp2$AB2)
median(ribs2NoSusp2$AB2)
wilcox.test(ribs2NoSusp2$AB2,conf.int=T)
t.test(ribs2NoSusp2$AB2,conf.int=T)
SIGN.test(ribs2NoSusp2$AB2)

# Boxplots
lmts<-range(0,10)
par(mfrow=c(1,5))
boxplot(ribs2SIDS2$AB2,main="SIDS", ylim=lmts)
boxplot(ribs2Bed2$AB2,main="Bed-share", ylim=lmts)
boxplot(ribs2Susp2$AB2,main="Suspicious", ylim=lmts)
boxplot(ribs2Nat2$AB2,main="Natural", ylim=lmts)
boxplot(ribs2Other2$AB2,main="Other", ylim=lmts)
par(mfrow=c(1,1))

## Non-suspicious deaths
ribs2NonSusp<-(ribs2$E1=="1"|ribs2$E1=="2"|ribs2$E1=="4"|ribs2$E1=="5"
ribs2NonSusp2<-ribs2[ribs2NonSusp,]
length(ribs2NonSusp2$A)
median(ribs2NonSusp2$AG)
mean(ribs2NonSusp2$AG)
wilcox.test(ribs2NonSusp2$AG,conf.int=T)
hist(ribs2NonSusp2$Z,main="Number of rib clefts per child", xlab="Frequency", ylab="Number of clefts", breaks=0:20-0.5)
mean(ribs2NonSusp2$Z)
median(ribs2NonSusp2$Z)
wilcox.test(ribs2NonSusp2$Z,conf.int=T)
sign.test(ribs2NonSusp2$Z)
hist(ribs2NonSusp2$AB, main="Number of rib clefts with length of >1.00 mm per child",
xlab="Frequency", ylab="Number of clefts", breaks=0:20-0.5)
mean(ribs2NonSusp2$AB)
median(ribs2NonSusp2$AB)
wilcoxon.test(ribs2NonSusp2$AB, conf.int=T)
sign.test(ribs2NonSusp2$AB)
hist(ribs2NonSusp2$AB2, main="Number of rib clefts with length of >.501 mm per child",
xlab="Frequency", ylab="Number of clefts", breaks=0:20-0.5)
mean(ribs2NonSusp2$AB2)
median(ribs2NonSusp2$AB2)
wilcoxon.test(ribs2NonSusp2$AB2, conf.int=T)
sign.test(ribs2NonSusp2$AB2)

hist(ribs2NoSusp2$AE1, main="Proportion of healing ribs", xlab="Proportion that are healing",
 ylab="Frequency of children with")
mean(ribs2NoSusp2$AE1)
median(ribs2NoSusp2$AE1)
wilcoxon.test(ribs2NoSusp2$AE1, conf.int=T)
t.test(ribs2NoSusp2$AE1, conf.int=T)
SIGN.test(ribs2NoSusp2$AE1)

kruskal.test(ribs2$Z ~ ribs2$E4A)
clefts<-aov(ribs2$Z ~ ribs2$E4A)
summary(clefts)
coefficients(clefts)
shapiro.test(residuals(clefts))
leveneTest (ribs2$Z, ribs2$E4A)
bartlett.test(ribs2$Z ~ ribs2$E4A)
TukeyHSD(clefts)
TransformedAB<-sqrt(ribs2$AB)
kruskal.test(ribs2$AB ~ ribs2$E4A)
clefts2<-aov(ribs2$AB ~ ribs2$E4A)
summary(clefts2)
coefficients(clefts2)
shapiro.test(residuals(clefts2))
leveneTest (ribs2$AB, ribs2$E4A)
bartlett.test(ribs2$AB ~ ribs2$E4A)
TukeyHSD(clefts2)
kruskal.test(ribs2$AB2 ~ ribs2$E4A)
clefts3<-aov(ribs2$AB2 ~ ribs2$E4A)
summary(clefts3)
coefficients(clefts3)
shapiro.test(residuals(clefts3))
leveneTest (ribs2$AB2,ribs2$E4A)
bartlett.test(ribs2$AB2~ribs2$E4A)
TukeyHSD(clefts3)
pairwise.wilcox.test(ribs2$Z,ribs2$E5A,p.adj="bonferroni",exact=F,paired=F)
pairwise.wilcox.test(ribs2$AB,ribs2$E5A,p.adj="bonferroni",exact=F,paired=F)
pairwise.wilcox.test(ribs2$AB2,ribs2$E5A,p.adj="bonferroni",exact=F,paired=F)

wilcox.test(ribs2$Z,ribs2$Z,paired=F,conf.int=TRUE)
wilcox.test(ribs2$AB,ribs2$AB,paired=F,conf.int=TRUE)
wilcox.test(ribs2$Z,ribs2$Z,paired=F,conf.int=TRUE)
wilcox.test(ribs2$AB,ribs2$Z,paired=F,conf.int=TRUE)
wilcox.test(ribs2$AB,ribs2$AB,paired=F,conf.int=TRUE)
cor(ribs2$AE1,ribs2$AB,method="spearman")

cor(ribs2$B,ribs2$AE1,method="spearman")
plot(ribs2$B,ribs2$AE1, xlab="Age at death(in months)", ylab="Proportion of clefts that are healing")

# Model fitting
# Poisson regression

ribregclefts<-glm(ribs2$AB~ribs2$E4B+ribs2$B+ribs2$C+ribs2$D+ribs2$AG, family=poisson(), na.action=na.omit)
ribregclefts<-zeroinfl(ribs2$AB~ribs2$B+ribs2$C+ribs2$D+ribs2$E4B+ribs2$AG, dist="negbin")

summary(ribregclefts)
anova(ribregclefts)
Anova(ribregclefts)

# testing of model fit
# the lower AIC is, the better
# next test should be 1
ribregclefts$deviance/ribregclefts$df.residual

# next test, substitute residual deviance score for residDev
pchiq(residDev,df of residual deviance,lower=F)
# or... when significance indicates lack of fit
pchiq(deviance(ribregclefts),df.residual(ribregclefts),lower=F)

# compare difference in size of residuals between models
1-pchiq(deviance(ribregclefts)-deviance(ribregclefts1),df.residual(ribregclefts)-df.residual(ribregclefts1))
#Another test
res<-residuals(ribregclefts,type="deviance")
plot(predict(ribregclefts),res,xlab="Fitted values",ylab="Residuals",ylim=max(abs(res))*c(-1,1))
abline(h=0,lty=2)

#Logistic regression
odds.susp<-glm(ribs2$E4B~ribs2$AB,family=binomial)
odds.reduced<-glm(ribs2$E4B~1,family=binomial)
anova(odds.reduced,odds.susp,test="Chisq")
summary(odds.susp)
exp(coef(odds.susp))
exp(cbind(OR=coef(odds.susp),confint(odds.susp)))
fit<-odds.susp$fitted
hist(fit)
r<-(ribs2$sta-fit)/(sqrt(fit*(1-fit)))
sumsq<-sum(r^2)
1-pchisq(sumsq,df=73)
anova(odds.susp,test="Chisq")

#Hosmer-Lemeshow test
index<-sort.list(fit)
index[1:10]
observed<-rep(NA,10)
for (i in 1:10) {observed[i] <- sum(hosmer[(8*(i-1)+1):(8*i),1])/8}
observed
predicted<-rep(NA,10)
for (i in 1:10) {predicted[i]<-sum(hosmer[(8*(i-1)+1):(8*i),2])/8}
predicted
plot(predicted,observed,type="b")
abline(a=0,b=1)

##Social factors
doctor<-table(ribs2$Aa)
doctor

sex<-table(ribs2$A1)
sex
ribsMale<-ribs2$A1="Male"
ribsMale1<-ribs2[ribsMale,]
ribsFemale<-ribs2$A1="Female"
ribsFemale1<-ribs2[ribsFemale,]
length(ribsMale1$A)
length(ribsFemale1$A)
wilcox.test(ribsMale1$Z,ribsFemale1$Z, paired=F, conf.int=T)
wilcox.test(ribsMale1$AB,ribsFemale1$AB, paired=F, conf.int=T)
wilcox.test(ribsMale1$AB2,ribsFemale1$AB2, paired=F, conf.int=T)

ancestry<-table(ribs2$A1a)
ancestry
kruskal.test(ribs2$AB~ribs2$A1a)
ribsWhite<-ribs2[ribsWhite,]
ribsNative<-ribs2[ribsNative,]
length(ribsWhite1$A)
length(ribsNative1$A)
wilcox.test(ribsWhite1$Z,ribsNative1$Z, paired=F, conf.int=T)
wilcox.test(ribsWhite1$AB,ribsNative1$AB, paired=F, conf.int=T)
wilcox.test(ribsWhite1$AB2,ribsNative1$AB2, paired=F, conf.int=T)

marriage<-table(ribs2$A2)
marrage
ribsMar<-(ribs2$A2=="Married")
ribsMar1<-ribs2[ribsMar,]
ribsUnmar<-(ribs2$A2=="Unmarried")
ribsUnmar1<-ribs2[ribsUnmar,]
length(ribsMar1$A)
length(ribsUnmar1$A)
wilcox.test(ribsMar1$Z,ribsUnmar1$Z, paired=F, na.rm=T, conf.int=T)
wilcox.test(ribsMar1$AB,ribsUnmar1$AB, paired=F, na.rm=T, conf.int=T)
wilcox.test(ribsMar1$AB2,ribsUnmar1$AB2, paired=F, na.rm=T, conf.int=T)

male<-table(ribs2$A2b)
male
kruskal.test(ribs2$AB~ribs2$A2b, na.action=na.omit)
kruskal.test(ribs2$Z~ribs2$A2b2,na.action=na.omit)
kruskal.test(ribs2$AB~ribs2$A2b2,na.action=na.omit)
kruskal.test(ribs2$AB2~ribs2$A2b2,na.action=na.omit)
ribsBoy<-(ribs2$A2b=="Boyfriend")
ribsBoy1<-ribs2[ribsBoy,]
ribsFath<-(ribs2$A2b=="Father")
ribsFath1<-ribs2[ribsFath,]
length(ribsBoy1$A)
length(ribsFath1$A)
wilcox.test(ribsBoy1$Z,ribsFath1$Z, paired=F, na.rm=T, conf.int=T)
wilcox.test(ribsBoy1$AB,ribsFath1$AB, paired=F, na.rm=T, conf.int=T)
wilcox.test(ribsBoy1$AB2,ribsFath1$AB2, paired=F, na.rm=T, conf.int=T)

living<-table(ribs2$A3)
living
ribsClean<-(ribs2$A3=="Clean")
ribsClean1<-ribs2[ribsClean,]
ribsMessy<-(ribs2$A3=="Messy")
ribsMessy1<-ribs2[ribsMessy,]
length(ribsClean1$A)
length(ribsMessy1$A)
wilcox.test(ribsClean1$Z,ribsMessy1$Z, paired=F, na.rm=T, conf.int=T)
wilcox.test(ribsClean1$AB,ribsMessy1$AB, paired=F, na.rm=T, conf.int=T)
wilcox.test(ribsClean1$AB2,ribsMessy1$AB2, paired=F, na.rm=T, conf.int=T)

housing<-table(ribs2$A3a)
housing
foster<-table(ribs2$A5)
foster
drug<-table(ribs2$A6)
drug
ribsYes<-(ribs2$A6=="Yes")
ribsYes1<-ribs2[ribsYes,]
ribsNo<-(ribs2$A6=="No")
ribsNo1<-ribs2[ribsNo,]
length(ribsYes1$A)
length(ribsNo1$A)
wilcox.test(ribsYes1$Z,ribsNo1$Z, paired=F, na.rm=T, conf.int=T)
wilcox.test(ribsYes1$AB,ribsNo1$AB, paired=F, na.rm=T, conf.int=T)
wilcox.test(ribsYes1$AB2,ribsNo1$AB2, paired=F, na.rm=T, conf.int=T)