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Classical metaphyseal lesions thought to be pathognomonic of child abuse are often artifacts or indicative of metabolic bone disease

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ABSTRACT

Objective: The objective of the present study was to review the histopathology in the original articles by authors Kleinman and Marks that described the specificity of the classical metaphyseal lesion for child abuse and to determine if there were any oversights in the authors' analysis.

Methods: We reviewed the histopathology of the original studies that equated the classical metaphyseal lesion with child abuse. We compared this with the histopathology of metaphyseal fractures caused by known accidental, severe trauma in children and reviewed the histopathology of artifacts that can sometimes be produced in bone histology preparations.

Results: Acute classical metaphyseal lesions showed no hemorrhage, and the chronic classical metaphyseal showed islands of cartilage proliferation at the metaphyses and growth plate, findings consistent with rickets and other metabolic bone disorders. Some of the acute metaphyseal lesions were consistent with artifacts.

Conclusion: We believe the original studies that equate the classical metaphyseal lesion with child abuse are flawed. The most compelling observation that challenges the histopathology of the classical metaphyseal lesion as being a fracture is the absence of hemorrhage in the acute classical metaphyseal lesion. We hypothesize that some of the classical metaphyseal lesions were artifacts or represent metabolic bone disorders that were not considered and that these two non-traumatic explanations may have been the basis of the abnormal bone findings.

Introduction

In the mid-1990s Kleinman and Marks published a series of 4 articles describing the classical metaphyseal lesion (CML) in 4 different long bone metaphyses and concluded that the CML was a highly specific type of fracture that is found almost exclusively in child abuse [1–4].

The authors had previously reported the radiologic and histopathology findings from 31 cases of infants who died and were allegedly abused in which radiographs and postmortem bone specimens were available for their evaluation [5]. It was from these 31 allegedly abused infants that the authors studied these 4 long bone metaphyses. These 5 studies of Kleinman and Marks were, in part, based on the work of Caffey who described metaphyseal lesions in association with subdural hematomas [6,7].

These publications have greatly influenced how child abuse is diagnosed in the infant with multiple unexplained fractures (MUF). The report of a pediatric radiologist in which a CML is found on a skeletal survey will almost always mention child abuse as a major concern as the cause of the CML. The finding of a CML or multiple CMLs will often drive the case of an infant with MUF to be called child abuse, although this dogma has been recently challenged [8–12].

Over the past 22 years author pediatrician Marvin Miller has been involved in both clinically evaluating infants with MUF and in reviewing cases of infants with MUF as a potential expert witness in which child abuse has been diagnosed and parents/caregivers deny wrongdoing. Many of these infants with MUF had CMLs which often did not behave like a true fracture in their clinical presentation and/or in their radiographic progression.

Clinical presentation

It has been reported that CMLs are found in approximately 50–77% of infants with MUF in which child abuse is alleged [13–15]. Noteworthy is that in most of these cases the CMLs were not associated with bruising, pain, swelling, or functional impairment of the affected long bone [13–15]. The CMLs were usually incidental findings that were noted on the skeletal survey. Since it is alleged that these fractures are typically caused by violent shaking or handling of the infant, it seemed

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unusual that these types of fractures behaved differently from a diaphyseal fracture of the long bone in which bruising, swelling, pain, and functional impairment are often found.

Radiographic progression

The progression of CMLs on serial skeletal surveys was also atypical in that the usual exuberant callus that is characteristic of a rib fracture or a diaphyseal fracture of a long bone is often absent in a CML. Interestingly, pediatric radiologists often comment in their reports that it is difficult to date the age of the CML because of this finding [16]. Flaherty comments about the CML: "CMLs commonly heal without subperiosteal new bone formation or marginal sclerosis. They can heal quickly and be undetectable on plain radiographs in 4 to 8 weeks" [10]. However, intraosseous and subperiosteal bleeding are essential parts of bone healing. That the CML is reported to occur in the most vascular part of the growth plate without pain, subperiosteal bleeding, or visible mineralization that accompanies healing leads one to question its classification as a fracture.

Evidence of low bone strength in infants with MUF

Infants with MUF associated with CMLs have been shown to have relatively lower bone strength, as studies have shown these infants to have lower bone density than normal controls and altered bone architecture. Fractures in such infants are thus likely fragility fractures, and risk factors for causing the metabolic bone disease in these infants have been described [14,15,17].

This skepticism about the CML and whether it was truly a fracture prompted a review of the histopathology in the original studies of Kleinman and Marks with a pediatric pathologist. We hypothesized that there may be non-traumatic explanations for the CMLs as described by Kleinman and Marks.

Methods

Review of the histology in the 4 Kleinman and Marks articles on regional CMLs

The original study of 31 deceased infants who were allegedly abused and which formed the basis of the 4 regional CML studies was reviewed [5]. The case information and histopathology of the CMLs in the 4 Kleinman and Marks articles were reviewed:

1. Proximal humerus [1]
2. Distal femur [2]
3. Proximal tibia [3]
4. Distal tibia [4]

Pediatric pathologist Dr. David Mirkin, with 50 years of experience, reviewed the histopathology photomicrographs in these 4 publications to see if he agreed with the conclusions of these 4 articles – namely, that the histopathology of the bone findings was consistent with a fracture, and that there was no other pathologic process that could explain the bone histology. Only low resolution print materials were available for review.

We also reviewed the correlation between the histology of the acute CMLs and the radiographs of the CMLs (both premortem and postmortem). Specifically, we evaluated how frequently the radiographs showed an acute CML when the bone histology showed an acute CML.

Review of medical literature that describes CMLs associated with known trauma

We reviewed the medical literature for other articles that described metaphyseal fractures and growth plate fractures in young infants to see how these findings compared with the histopathology findings of the 4 Kleinman and Marks articles.

Two PubMed searches were performed to find photomicrographs of the histopathology of metaphyseal fracture in infants who were not abused.

1. A PubMed search combining the terms "metaphyseal fracture", "pathology" and "infant" yielded 41 publications, and only one had relevant pathology of a metaphyseal fracture in an infant who had a fracture that was clearly not related to child abuse (Ref. [26]).
2. A PubMed search combining the terms "metaphyseal fracture", "histology" and "infant" yielded 45 publications and only one had relevant histology of a growth plate fracture immediately next to the metaphysis in an infant who had a fracture that was clearly not related to child abuse (Ref. [27]).

Review of medical literature that describes bone artifacts

We had concern that some of the CMLs that Kleinman and Marks described were bone artifacts. We therefore reviewed the literature looking for published photomicrographs of bone artifacts that might mimic a metaphyseal fracture. A Google search of images combining the terms "bone histology" or "bone histopathology" and "artifacts" showed several relevant images, one of which is illustrated in Fig. 1D.

Results

Review of 4 Kleinman and Marks articles on regional CMLs

Table 1 summarizes the case information of the deceased infants and CMLs as provided in the 4 articles.

Table 2 compares the findings of Kleinman and Marks with our findings in 6 acute CMLs described in these 4 papers (one acute CML is described in 3 of the papers and 3 acute CMLs are described in the proximal tibia paper which are listed as a, b, and c in Table 2).

Table 3 compares the findings of Kleinman and Marks with our findings in one representative healing CML from each of the 4 articles.

The major results from our review are the following:

a. Acute CMLs

The authors publish bone histology of one acute CML in 3 of these papers and 3 infants with acute CMLs in the proximal tibia paper. Fig. 1C shows an example of an acute CML from the distal tibia paper.

There was no hemorrhage in any of the following histopathology photomicrographs of the 4 acute CMLs presented in the 4 Kleinman and Marks articles (Figures refer to Kleinman and Marks articles):

1. Proximal humerus [1]: Fig. 2
2. Distal femur [2]: Fig. 2C

(Article states: "No evidence of repair or hemorrhage is found.")

3. Proximal tibia [3]: Fig. 3C

(Article states: "High power image shows trabecular disruption with minimal hemorrhage and no repair")

4. Distal tibia [4]: Fig. 1C

The histopathology in all 6 of the acute CMLs was also consistent with rickets.

b. Healing CMLs

In the 4 studies, bone histology is presented of 11 healing CMLs, and in all of them the Kleinman and Marks describe them as showing some degree of chondrocyte proliferation, findings similar to those described

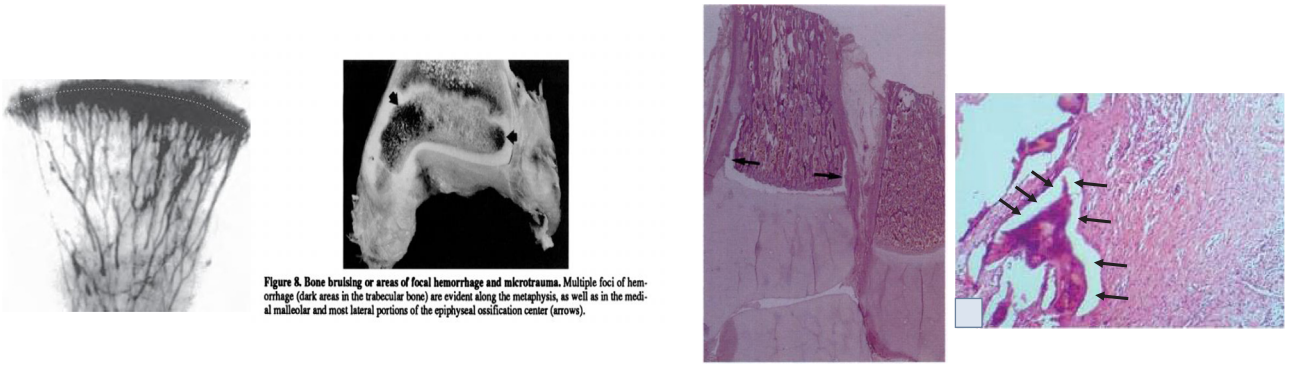


Figure 1A shows a microangiogram of the proximal metaphysis in human fetal femur. The metaphyseal blood supply is clearly highly vascular even though only the arterial components are shown. The primary spongiosa is estimated by dotted line.
[Figure 5 from reference 28; Reprinted with permission from the Journal of Anatomy]
Figure 1B shows the gross specimen from a child's long bone after a traumatic event which shows focal hemorrhage at the metaphysis [black arrows]
[Figure 8 from reference 26; Reprinted with permission from the Yale Journal of Biology and Medicine]
Figure 1C shows the microscopic section from an acute CML in the distal tibia as reported by Kleinman and Marks in which there is no apparent hemorrhage. Alleged fracture (black arrows) is likely an artifact of bone separation.
[Figure 1C from reference 4; Reprinted with permission from the American Journal of Roentgenology].
Figure 1D shows the microscopic section from a bone specimen in which there was artifactual separation of the bone during microtomy [black arrows]
[Figure 3a from reference 23; This open journal allows for reprinting of this Figure without written permission]
This separation artifact is similar to what we believe is an artifact in Figure 1C.

Fig. 1. Visual evidence of hemorrhage in a true metaphyseal fracture and none in an acute CML.

in rickets as illustrated in Fig. 2. [18–21]. Specific figures in these 4 articles that showed findings that are highly suggestive of rickets are the following:

(Figures refer to those in the Kleinman and Marks articles):

1. Proximal humerus [1]: Fig. 6C
2. Distal femur [2]: Fig. 4C
3. Proximal tibia [3]: Fig. 4C
4. Distal tibia [4]: Fig. 4C

c. Similarity of Acute CMLs to Artifacts
Several of the photomicrographs of acute CMLs had bone irregularities that appeared to be tissue separation artifacts that mimic a fracture (see section 2 of Discussion) [22–25].

d. Histology-Radiograph Correlations
The histology-radiograph correlation of the acute CMLs showed the following in each of the 4 studies.

1. Proximal humerus: There was one acute CML identified by histology, and this was not visible radiographically.
2. Distal femur: There were 3 acute CMLs identified by histology. The authors do not comment specifically if these were visible on the radiographs. They state: "The acute classical metaphyseal lesion is often quite subtle radiographically".
3. Proximal tibia: There were 16 CMLs identified by histology (both acute and chronic – the authors do not state how many of each). Radiographic abnormalities were noted in 14 of the 16. The authors state: "The acute fracture may not be visible on the standard lateral projection".
4. Distal tibia: There was 3 acute CMLs identified by histology, and one was visible on the premortem radiograph. The other 2 acute CMLs could only be identified on the postmortem radiographs.

Because the acute CMLs that were identified by bone histology were often not noted on the radiographs, it is likely that some of these acute CMLs were bone artifacts made during the preparation of the bone specimens.

Review of medical literature that describes CMLs associated with known trauma

The review of the medical literature for publications describing metaphyseal fractures and fractures near the growth plate revealed two relevant studies:

Table 1
Summary of infants and CMLs studied in 4 regional bone studies.

Study/Reference	# infants	# CMLs	Acute CML	Healing CML	Unilateral/Bilateral	Age Range/Age Average
Proximal Humerus [1]	4	7	1	6	NG	2–5 months 14 weeks
Distal Femur [2]	11	15[#]	3	9	4/7	1–5 months 3.5 months
Proximal Tibia [3]	NG	17[@]	NG	NG	5/6	NG
Distal Tibia [4]	12	16/14[*]	3[**]	11	8/4	NG

NG = Not Given.
[#] Medial involvement of the distal femur in all cases.
[@] Medial and posterior involvement in all specimen X-rays.
[*] Technical problems precluded evaluation of 2 instances; only 14 CMLs evaluated. Medial involvement of the distal tibia in all instances; lateral involvement in 4.
[**] Only 1 of 3 Acute CMLs were seen on skeletal X-ray, the other 2 were seen on specimen X-ray at autopsy.

Table 2
Comparison of 6 acute CMLs from four different long bone metaphyses: original authors findings versus our findings.

Bone/Reference	Age/Gender/Figure of ACUTE CML	Histology Findings by Original Authors of ACUTE CML	Our Histology Findings of ACUTE CML
1. Proximal Humerus [1]	2 month Female Fig. 2	"extension of fracture into zone of hypertrophic chondrocytes of physis"	1. No hemorrhage 2. No fracture line noted
2. Distal Femur [2]	1 month Male Fig. 1C	"No evidence of repair or hemorrhage is found"	1. No hemorrhage 2. Microtome artifact
3. Proximal Tibia [3] a	4 month NG Fig. 2C	"disruption of trabeculae of primary spongiosa with extension of fracture line into proliferative zone of physis"	1. No hemorrhage 2. No fracture line noted 3. Artifact present
4. Proximal Tibia [3] b	1 month NG Fig. 3C	"trabecular disruption with minimal hemorrhage and no evidence of bone repair"	1. No hemorrhage 2. No fracture line noted 3. Possible artifact
4. Proximal Tibia [3] c	4 month NG Fig. 4D	"extension of growth plate cartilage into physis"	1. No hemorrhage
4. Distal Tibia [4] NG = Not Given	3 month Female Fig. 1C	"Artifactual widening of plane of fracture is present"	1. No hemorrhage 2. Microtome artifact

A. Ogden [26]

The authors obtained 57 skeletal tissue samples from non-fatal accidents in which there were acute injuries to the developing skeleton in children ranging in age from 7 months to 15 years and performed histology on these bone specimens. They present 2 photomicrographs of bone metaphysis in infants/children who experienced trauma that clearly demonstrate metaphyseal fracture/injury in which there is hemorrhage at the metaphysis. An example is shown in Fig. 1B.

B. Rodriguez [27]

The authors studied 11 young infants who died within the first few weeks of life from various lethal congenital neuromuscular disorders including myotonic dystrophy, spinal muscular atrophy, and primary myopathies. These infants had severe bone demineralization associated with fractures that was related to the decreased fetal and postnatal bone loading. The authors did histology on the bones with fractures. In the only recent fracture which was of the proximal tibial epiphysis, there were necrotic chondrocytes with an organized blood clot, indicating such a fracture near the metaphysis again bleeds within the bone.

The combined results of these 2 studies indicate that metaphyseal fractures/growth plate fractures associated with trauma do result in hemorrhage, as would be expected from both:

1. the anatomy of the metaphysis, which has a rich vascular supply [28], and
2. the fact that hemorrhage is necessary to initiate bone healing [29,30]

Review of medical literature that describes bone artifacts

The pathology literature review of bone histology artifacts showed that it is well-known that bone separation artifacts can arise in the preparation of bone specimens during microtomy as illustrated in Fig. 1D [22–25]. Several bone irregularities which Kleinman and Marks called acute CMLs are similar to these bone artifacts that we found in the pathology literature. An example of an acute CML in the Kleinman and Marks article on the distal tibia is shown in Fig. 1C and is similar in appearance to the bone artifact from the pathology literature in Fig. 1D [[4,23] (Fig. 3a)]. Thus, we believe that it is likely that the histology of the acute CML described by Kleinman and Marks may represent artifact, especially given the absence of hemorrhage in the acute CML and the often normal radiographs that did not show the acute CMLs.

We believe that several of the other CML microscopy abnormalities are not true bone abnormalities, but rather are artifacts in the preparation of the slides including the Figures in the following Kleinman and Marks articles:

1. Proximal humerus [1]: Fig. 1C
2. Distal femur [2]: Fig. 2C
3. Proximal tibia [3]: Fig. 2C

Discussion

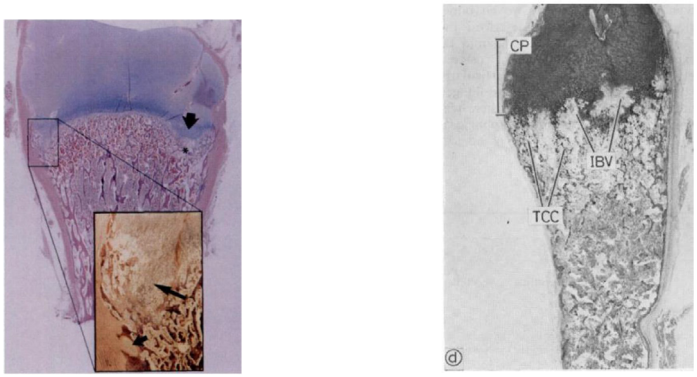
The findings of the above review lead to the following conclusions:

1. Lack of hemorrhage in the acute CML

The single most compelling observation that indicates the CMLs reported by Kleinman and Marks in these 4 articles are unlikely fractures is that there is no hemorrhage associated with any of the acute CMLs presented in the histopathology in these 4 articles, in spite of the metaphysis being a very vascular region of bone and hemorrhage being essential for bone repair [28–30]. The cytokines and cellular elements from the leaked blood of the traumatized vascular tree in the region of a fracture initiate bone healing which leads to complete repair [29,30]. Metaphyseal and growth plate fractures in young infants/children with known trauma or fragility fractures showed hemorrhage in the histopathology of the fractures [26,27]. Thus, fractures at or near the

Table 3
Comparison of 4 representative healing CMLs from 4 different long bone metaphyses: original authors findings versus our findings.

Bone/Reference	Age/Gender/Figure of HEALING CML	Histology Findings by Original Authors: HEALING CML	Our Histology Findings HEALING CML
1. Proximal Humerus [1]	5 month Male Fig. 4B	"Volume of hypertrophic zone is increased, which results in irregularity and loss of convex metaphyseal margin"	Compatible with rickets and other metabolic bone disorders
4. Distal Femur [2]	4 month Male Fig. 4C	"focal extension of hypertrophic chondrocytes into metaphysis"	Compatible with rickets and other metabolic bone disorders
3. Proximal Tibia [3]	3 month NG Fig. 6C	"Focal extension of physeal hypertrophic cartilage into fracture site is visible"	Compatible with rickets and other metabolic bone disorders
4. Distal Tibia [4] NG = Not Given	1 month NG Fig. 4C	"zone of extension of hypertrophic chondrocytes into primary spongiosa"	Compatible with rickets and other metabolic bone disorders



A B

Healing CML from alleged child abuse (Kleinman) **Neonatal rickets**

A. Histopathology of healing CML of distal tibia which is described in article as: "Note trabecular disorganization. Although no laterally situated injury was evident radiographically, histologic evidence of fracture is visible on inset. Note extension of hypertrophic chondrocytes into primary spongiosa".
[Figure 3B from reference 4; Reprinted with permission from the American Journal of Roentgenology]

B. Histopathology of rickets of rib in neonate which is described in article as: "There was irregular penetration of the cartilage by blood vessels leaving residual large tongues and islands of calcified cartilage".
[Figure 2 from reference 19; Reprinted with permission from the BMJ]
(CP = Cartilage Plate; TCC = Tongues of Calcified Cartilage; IBV= Invading Blood Vessels)

The histopathology of the healing CML is similar to that seen in healing neonatal rickets.

Fig. 2. Comparison of the healing CML with that of infant rickets.

metaphysis do produce hemorrhage. As noted in Robbins, a well-accepted pathology textbook: "Immediately after fracture, rupture of blood vessels results in a hematoma, which fills the fracture gap and surrounds the area of bone injury" [31].

It is noteworthy that Kleinman and Marks themselves state in their articles that hemorrhage is absent in acute CMLs [2,3]. In the proximal tibia paper they state: "Notable intramedullary hemorrhage and evidence of subperiosteal bleeding are seldom found. This correlates with the usual lack of hemorrhage visible at dissection". Thus, the lack of hemorrhage in the acute CML that we observed in our review of the 4 Kleinman and Marks CML articles is compelling evidence that these are not fractures, but represent some other phenomenon. We believe this phenomenon is a region of poorly mineralized bone that has not yet calcified or an artifact.

2. Similarity of histology of the acute CML to known bone artifacts

The histopathology published by Kleinman and Marks illustrating the acute CML lesions is strikingly similar to the well-known bone separation artifacts that can arise in the preparation of the bone specimens during microtomy [22–25]. Some of these alleged bone separation abnormalities that Kleinman reported as acute CMLs on microscopy were not noted on the radiographs which would be further evidence that some of these CMLS may not have been true fractures and may have been artifacts (2,3,4).

The authors state in the distal tibia CML paper that: "Artifactual widening of plane of fracture is present" in their description of an acute CML [4, Fig. 1C in this paper]. We question why they don't conclude that this CML is a complete artifact, and that there is truly no fracture (see Fig. 1C in the present paper).

3. Similarity of histology of the healing CML to other bone disorders

The histopathology that the authors present in the healing CMLs in all 4 articles are similar and show a pattern of chondrocyte proliferation in the absence of hemorrhage, subperiosteal new bone formation or marginal sclerosis. This finding is seen in disorders of poor bone mineralization including rickets, and other metabolic bone disorders such as hypophosphatasia, vitamin D deficiency rickets, vitamin C deficiency, inherited disorders of vitamin D metabolism, and renal tubular acidosis [18–21]. Cohen's recent publication of an autopsy series of 41 infants who died of sudden, unexpected death showed a high frequency of vitamin D deficiency as measured by 25 OH vitamin D levels with 26 of 41 (63%) subjects having 25 OH vitamin D levels less than 20 ng/ml (normal: > 30) [32]. Rib histology was abnormal in 69% of the infants with inadequate vitamin D levels. Noteworthy, the authors published bone histopathology findings of some of their cases with low 25 OH vitamin D levels which were consistent with rickets [32].

4. Our interpretation of the origin of the CML

We interpret the bone microscopy of the CMLs presented in the 4 Kleinman articles as showing poor bone mineralization of the metaphysis. We believe one of the disorders that may have been represented in the series of infants that Kleinman and Marks studied was Metabolic Bone Disease of Infancy (MBDI). MBDI is a relatively common disorder of poor bone mineralization that presents in the first 6 months of life. It is a multifactorial disorder that can arise from various influences that are mostly of fetal origin including maternal vitamin D deficiency - during pregnancy, inadequate maternal calcium and/or phosphate absorption during pregnancy, decreased fetal bone loading, and other possible factors [14,15,18–21,33–35]. Early infantile rickets can occur in the first few months of life after vitamin D stores gained in utero are depleted, a condition exacerbated by lack of supplementation and the demands of rapid bone growth. Such an explanation would be consistent with the following characteristics of many, but not all, CMLs:

1. Lack of swelling, bruising, and functional impairment of the affected bone
2. Difficulty in dating the CML
3. Filling in of the CML without exuberant callus on follow-up skeletal radiographs

Because the CML may represent a region of poor bone mineralization, some CMLs may fracture or cause growth plate slippage as Ogden and Salter/Harris have previously described, but in such instances there would be clinical signs of a fracture and evidence of a more exuberant healing in follow up skeletal x-rays [36,37].

Bone scans are often normal when the plain X-ray shows a CML, another observation that questions the validity that all CMLs are real

fractures [38]. Analysis of fracture patterns in infants with MUF and their specificity for child abuse by Kemp et al. discounted Kleinman's studies, as they did not meet inclusion criteria for their study, stating: "Historically metaphyseal fractures have been thought as strong predictors of abuse, but we could not find comparative studies to support or refute this hypothesis" [39].

5. Implications of our findings

It is estimated that there are some 1700 cases of infants under one year of age with MUF in the USA per year, and some 50–76% have CMLs [40]. The CML dogma of diagnosing an infant who has a CML with child abuse as a result of violent shaking has been accepted for almost 30 years [41]. Based on the findings presented herein, we believe that some of these cases of alleged child abuse in infants based on the finding of a CML may have been incorrectly diagnosed, and medical diagnoses have likely been missed with devastating consequences for families.

Kleinman and Marks obtained their material in these 4 studies from 31 cases of infants who had died in which child abuse had been diagnosed. While child abuse is in the differential diagnosis of these 31 cases, the fact that some of these infants likely had evidence of poor bone mineralization from our analysis raises the possibility that there may be alternative medical diagnoses that should also have been considered in these cases including hypophosphatasia, vitamin D deficiency, vitamin C deficiency, vitamin D resistant rickets, renal tubular acidosis, type 1 hereditary vitamin D dependent rickets, and MBDI. Some CMLs are found to eventually be normal variants [42].

The CML has often been associated with the triad of the shaken baby syndrome (aka abusive head trauma). The recent review of the shaken baby syndrome by the Swedish Council on Technology Assessment in Health Care raises serious concern that shaking can cause the triad [43]. The authors of this report also underscore the strong possibility that medical diagnoses, including some of the diagnoses mentioned above, may have been/may be missed in alleged cases of shaken baby syndrome. One specific diagnosis that could explain both fractures and the triad is hypocalcemic vitamin D deficient rickets [44,45].

Conclusion

The Kleinman and Marks regional metaphyses studies that proposed and popularized the concept that the CML is highly specific for child abuse are flawed. Without evidence of hemorrhage in the acute CML and with several of the acute CML photomicrographs being consistent with artifact, these studies lack the scientific foundation to conclude that a CML is indicative of child abuse. Many of their healing CMLs are consistent with rickets and other metabolic bone disorders. We do not believe a diagnosis of child abuse should be made based solely on the finding of a CML.

Acknowledgments

The authors are grateful to Shelley Miller and Eric Gershon for their critical review of the manuscript. This manuscript is dedicated to the memory of Dr. John Plunkett whose life's work greatly enhanced our understanding of infant injury.

Conflict of interest statement

The authors have no conflict of interest.

Dr. Miller discloses that he reviews cases of infants with unexplained fracture at the request of parents who are accused of child abuse in such cases or the parent's lawyers. Most of these cases are done pro bono.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at http://dx.doi.org/10.1016/j.mehy.2018.03.017.

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