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COMMENTARY

Rickets vs. abuse: a national and international epidemic

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In the May 2007 issue of *Pediatric Radiology*, the article "Can classic metaphyseal lesions follow uncomplicated caesarean section?" [1] suggested that enough trauma could occur under these circumstances to produce fractures previously described as "highly specific for child abuse" [2]. However, the question of whether the metaphyses were normal to begin with was not raised. Why should this be an issue?

Vitamin D deficiency (DD), initially believed to primarily affect the elderly and dark-skinned populations in the US, is now being demonstrated in otherwise healthy young adults, children, and infants of all races. In a review article on vitamin D published in the *New England Journal of Medicine* last year [3], Holick reviewed some of the recent literature, showing deficiency and insufficiency rates of 52% among Hispanic and African-American adolescents in Boston, 48% among white preadolescent females in Maine, 42% among African American females between 15 and 49 years of age, and 32% among healthy white men and women 18 to 29 years of age in Boston. A recent study of healthy infants and toddlers aged 8 to 24 months in Boston found an insufficiency rate of 40% and a deficiency rate of 12.1% [4].

In September 2007, a number of articles about congenital rickets were published in the *Archives of Diseases in Childhood* including an international perspective of mother and newborn DD reported from around the world [5]. Concentrations of 25-hydroxyvitamin D [25(OH)D] less than 25 nmol/l (10 ng/ml) were found in 18%, 25%, 80%, 42% and 61% of pregnant women in the UK, UAE, Iran, northern India and New Zealand, respectively, and in 60 to 84% of non-western women in the Netherlands. Currently, most experts in the US define DD as a 25(OH)D level less than 50 nmol/l (20 ng/ml). Levels between 20 and 30 ng/ml are considered to indicate insufficiency, reflecting increasing parathyroid hormone (PTH) levels and decreasing calcium absorption [3]. With such high prevalence of DD in our healthy young women, congenital deficiency is inevitable, since neonatal 25(OH)D concentrations are approximately two-thirds the maternal level [6].

Bodnar et al. [7] at the University of Pittsburgh, in the largest US study of mother and newborn infant vitamin D levels, found deficient or insufficient levels in 83% of black women and 92% of their newborns, as well as in 47% of white women and 66% of their newborns. The deficiencies were worse in the winter than in the summer. Over 90% of these women were on prenatal vitamins. Research is currently underway to formulate more appropriate recommendations for vitamin D supplementation during pregnancy (http://clinicaltrials.gov, ID: R01 HD043921).

The obvious question is, "Why has DD once again become so common?" Multiple events have led to the high rates of DD. In the past, many foods were fortified with

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vitamin D. At present, milk is the only product required to be fortified in the US, although studies have shown that up to 70% of sampled milk does not contain the required amount of vitamin D. There are few other natural food sources of vitamin D. While exposure to the sun's UVB rays remains the best natural source of vitamin D, during the winter months, above the 35° latitude (at the level of Atlanta, Georgia) there is a loss of UVB rays that are necessary for skin synthesis of vitamin D. In addition, along with the concern for skin cancer and skin aging, there has been a significant increase in the use of sunscreen and protective clothing. A sun protection factor of 8 blocks about 95% of the UVB rays, and with regard to vitamin D, sunscreen use produces the same effect as a burka worn by certain Islamic women, who, even in the sunny climate of the Middle East, continue to have high rates of DD. The increase in obesity is another important factor since fat storage of this vitamin decreases its availability to the rest of the body. In a recently published study, the vitamin D status of neonates born to obese mothers was found to be poorer than the vitamin D status of neonates born to lean mothers [8].

Over the past few decades, there has been a significant increase in the frequency and the duration of breast-feeding, both of which are risk factors for infant nutritional DD. In 2003, the American Academy of Pediatrics published guidelines recommending that all breast-fed infants receive 100% of their daily vitamin D requirement from supplementation. Research has demonstrated breast milk to be so deficient in vitamin D that the average lactating woman must feed her infant 8 l (about 89 three-ounce bottles) of breast milk per day to provide the recommended daily vitamin D intake of 200 IU [9]. Even infants born to mothers who are replete with vitamin D demonstrate decreasing levels within 8 weeks if not supplemented [10]. Ziegler et al. [11] at the University of Iowa, during a study of iron levels in breast-fed infants, incidentally found that the majority of their unsupplemented infants had DD, with severe DD common in the winter. Recent literature suggests that supplementing lactating women with 2,000 to 4,000 IU per day (rather than the current recommendation of 400 IU per day) is required to provide both the mother and her breast-fed infant with adequate vitamin D [12].

What are the radiographic findings of congenital and nutritional rickets in infants younger than 6 months old, the age at which we typically first see the fraying and cupping associated with rickets? The appearance of DD in early infancy will vary with many factors, including age at presentation, prenatal maternal DD, nutritional history, geographic location, sun exposure, skin color, and season. Past case reports of congenital rickets range from normal-appearing bones to diffuse bone rarefaction, fractures at birth, and metaphyseal fraying and cupping [13–18]. The best location to search for radiographic evidence of congenital rickets and nutritional rickets in infants less than 3 months of age is in the cranium [19]. The skull, as a reflection of rapid brain growth in early life, is the most affected bone at this age. Softening of the skull (craniotabes), especially in the occipital region with palpable enlargement of the sutures and fontanelles, was a common clinical finding with congenital rickets in the past, and is once again being described in association with newborn DD [20]. Demineralized sutures create a diastatic appearance with poorly defined margins on radiographs (pseudodiastasis). The skull is better evaluated on the head CT scan, which should be done to exclude sutural diastasis from increased intracranial pressure. Indistinctness of the facial bones and the orbital roof make the teeth and petrous bones appear relatively dense on radiographs [21].

Metaphyseal changes of the long bones appear first at the most recently formed metaphyseal margin, the Laval-Jeantet ring. These metaphyseal changes are usually symmetric, and will occur first in the fastest-growing long bones (the femurs, tibiae and fibulae). The metaphyseal changes in the distal femurs and proximal tibiae occur first along the medial aspects [22], and the early metaphyseal changes in the distal ulnar can be found even when the distal radial metaphyses appear normal [23]. While the physeal and metaphyseal changes can be subtle in early infancy, with treatment, the metaphyses may become increasingly sclerotic and deformed [23]. While these early metaphyseal changes may mimic Salter-Harris II fractures, they are asymptomatic and, with treatment, usually resolve without the interval stages of fracture healing (subperiosteal new bone formation, callous, and remodeling) within a few weeks. When subperiosteal changes are present, particularly in the tibiae, a lateral view may reveal early bowing deformity. Bowing deformities of the long bones prior to weight bearing are not common, but when present, they reflect positional stresses or musculotendinous forces, such as the "saber shin deformity" where the pull of the Achilles tendon on the calcaneus causes the tibiae to bow anteriorly [19].

Rachitic flaring of the anterior rib ends will also be subtle on radiographs in the acute stage at this age, but may become more pronounced with healing. Loss of cortical distinction, subperiosteal new bone formation in the long bone diaphyses, insufficiency fractures and Looser zones (i.e. pseudofractures) of the ribs and forearms have all been described in infants [19, 23, 24]. It may be particularly difficult to distinguish rib fractures from Looser zones in infants on radiographs alone. Timing of fractures in children with rickets is not possible since published parameters assume normal underlying bone. The only assumption that can be made of fractures in an infant with DD is that the healing will be delayed.

It is also important to remember when searching for radiographic evidence of vitamin deficiencies that multiple deficiencies are the rule and may modify the radiographic appearance of the skeletal response. In addition, the appearance of rickets will be altered by factors such as seasonal and dietary changes that cause waxing and waning of the deficiency [23].

Radiographs alone may not give us enough information about the bone health in these infants. Prior studies on the accuracy of radiographs in determining bone mineralization have shown that adults with osteoporosis have 30–40% bone mineral loss before it becomes evident on radiographs. Unfortunately, the value of DXA bone density studies at this age may be limited. Bishop and Plotkin [25] studied the bone mineral density of the lumbar spine by DXA in infants with fractures due to osteogenesis imperfecta and in infants with fractures thought to be non-accidental, and found both groups within the reference range and not significantly different until after 6 months of age. However, differences in bone mineral density have

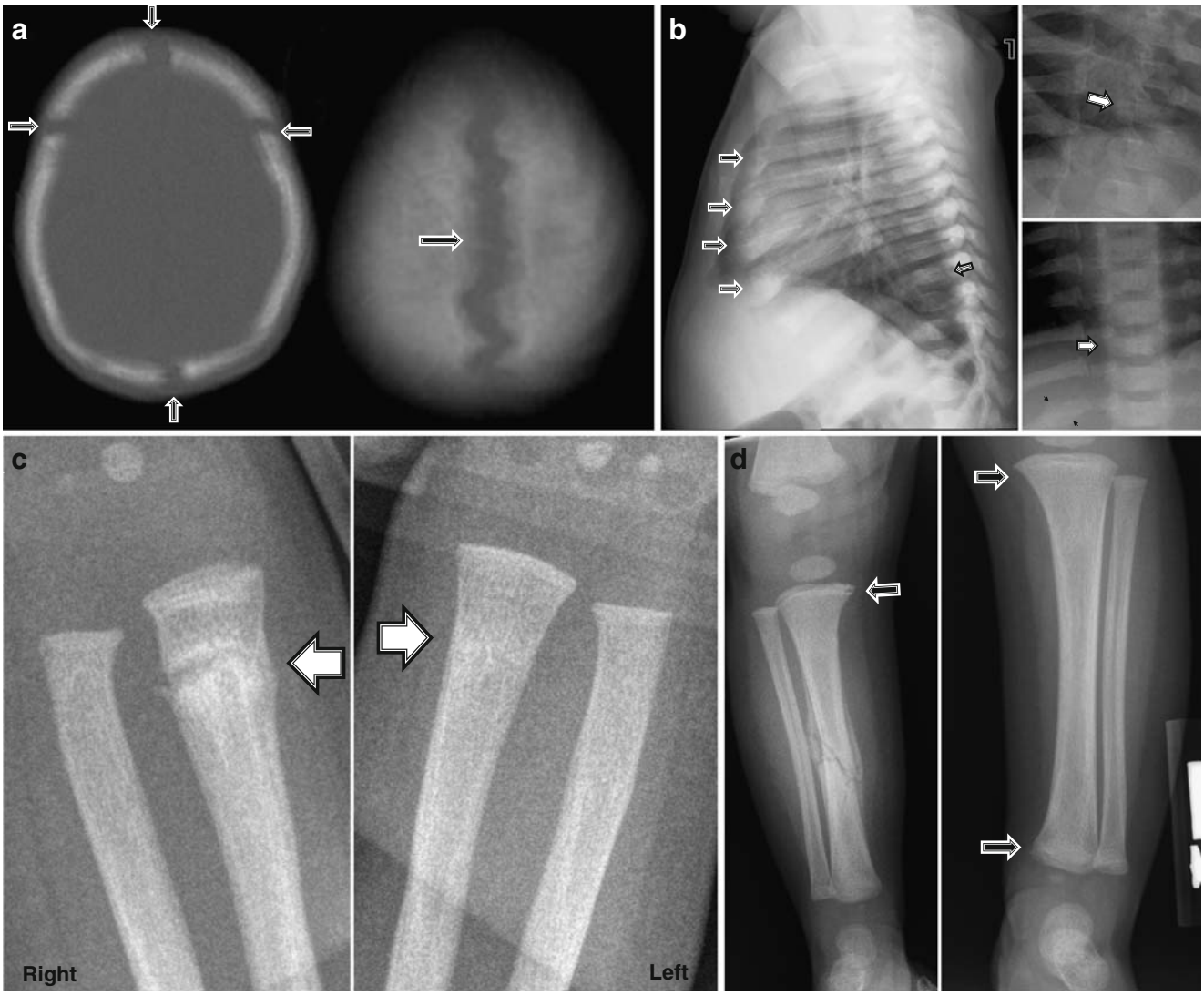

Fig. 1 Case 1, a 2-month-old Caucasian girl. **a** Axial CT images (bone algorithm) demonstrate diastatic-appearing coronal and sagittal sutures with poorly defined margins (*arrows*). Brain CT scan showed no intracranial abnormality, and there were no clinical findings for increased intracranial pressure, confirming the pseudodiastatic appearance of craniotabes. **b** Chest radiographs. The lateral view shows a flared appearance of the anterior rib ends (*white outlined arrows*) with a compression fracture of T8 that is confirmed on the AP and oblique views (*black outlined arrows*). Also, notice the healing right posterior fracture of the tenth rib vs. pseudofracture (Looser zone, *small black arrows*). **c** AP views of the forearms demonstrate transverse lucency with sclerotic borders of the right distal radial metadiaphysis and transverse sclerotic changes in the same location of the left distal radius (i.e. Looser zones-*arrows*). **d** AP views of the lower extremities show mild irregular sclerotic changes of the tibial and fibular metaphyses, distal more than proximal (*arrows*). The oblique fracture of the right tibia was asymptomatic on presentation and was never noted on multiple prior visits to the pediatrician

been documented in young infants with osteogenesis imperfecta using quantitative CT [26], which may be a more accurate modality in early infancy. Quantitative US for bone strength may be a useful adjunct to CT to give us a better understanding of bone health [27].

While MRI is unnecessary in detecting physeal widening in older infants and children, in younger infants, when the epiphyses are minimally, if at all, ossified, it is the best modality for evaluating physeal abnormalities. MRI has already been used to describe rickets in older children [28], with the failure of ossification in the hypertrophic zone and persistence of cartilage into the metaphysis. The physeal width of the distal femur showed little variability (0.9 to 1.9 mm) in a study of MRI in normal children, which correlates with a prior histologic study [29].

Case reports

Case 1 A 2-month-old Caucasian girl (Fig. 1) was brought to the pediatrician for an asymptomatic leg bump. A healing oblique right tibial fracture was found on the radiograph. The skeletal survey was interpreted as 16 fractures suspicious for inflicted injury. The mother's vitamin D level was 8.7 ng/ml (insufficiency 20–30 ng/ml, deficiency <20 ng/ml). The infant was not checked for DD.

Case 2 A 2-month-old Caucasian girl (Fig. 2) was brought to the pediatrician for her regular well-baby check and for right knee swelling. A metaphyseal fracture of the right distal femur was diagnosed by radiograph and a follow-up skeletal survey was interpreted as up to 28 fractures highly specific for nonaccidental trauma. The mother's vitamin D level, checked in the summer, was <4 ng/ml. Two months after being changed to formula feeding, the infant's vitamin D level, in the summer, was 17.8 ng/ml.

Case 3 A 4-month-old Caucasian boy (Fig. 3) had left lower leg swelling after an accident at day care. Radiographs demonstrated a fracture and follow-up skeletal surveys were interpreted as seven or eight fractures that were nonspecific. The mother was found to have a vitamin D level of 14 ng/ml and a PTH of 120 pg/ml (levels >72 pg/ml indicative of hyperparathyroidism). After 6 months of formula feeding, the infant's vitamin D level was 19.6 ng/ml, and the alkaline phosphatase level was 2,386 U/l (normal range 145–320 U/l).

Case 4 A 2-month-old girl (Fig. 4) with an African-American mother and Caucasian father presented with a viral respiratory illness. A chest radiograph demonstrated healing rib fractures. Skeletal survey was interpreted as six, possibly eight, fractures highly specific for nonaccidental trauma. The baby had skull and cervical spine fractures. MRI demonstrated small old subdural hemorrhages. She had no clinical or imaging findings to suggest increased intracranial pressure. While on prenatal vitamins during her second pregnancy, the mother's vitamin D level was 17 ng/ml

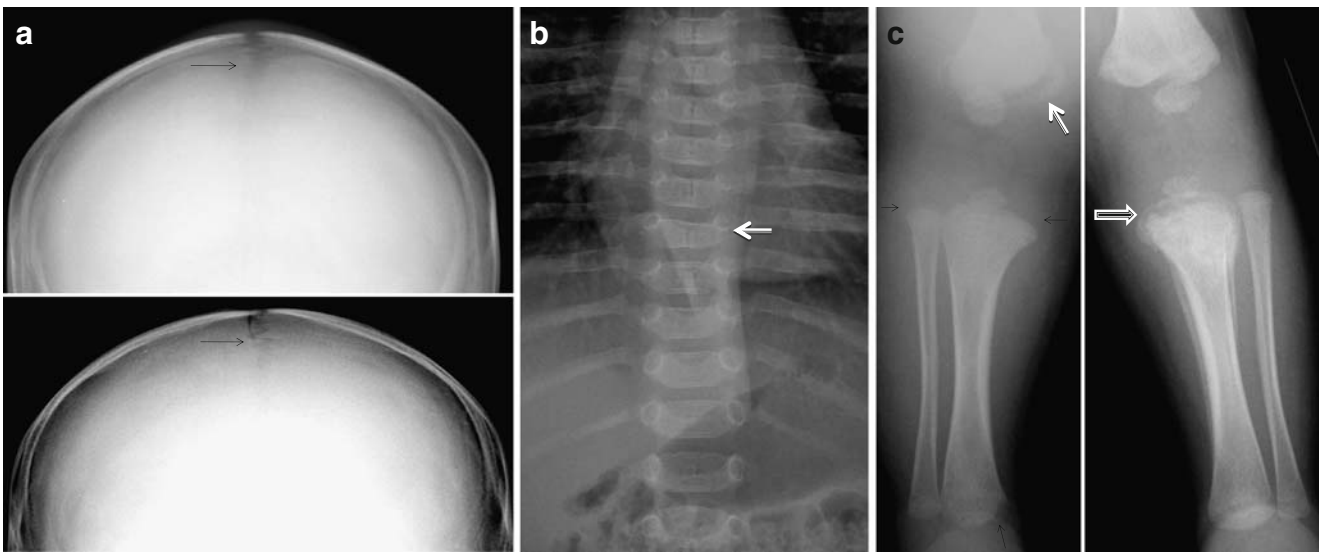

Fig. 2 Case 2, a 2-month-old Caucasian girl. **a** AP skull radiographs. On presentation the sagittal suture appears widened (*top image*, *arrow*) but this has resolved after 5 months of formula feeding (*bottom image*, *arrow*). Brain CT and MRI showed no intracranial abnormality and there were no clinical findings for increased intracranial pressure. **b** AP spine view shows asymmetric T8 vertebral body height deformity (*arrow*). **c** AP views of the lower extremities show mild fibular bowing deformities with a metaphyseal fracture of the right distal femoral metaphysis (*white arrow*) and left proximal tibia (*white outlined arrow*). Metaphyseal irregularities of the right proximal and distal tibia and fibula are also noted (*thin black arrows*). The left distal tibia and fibula are partially obscured

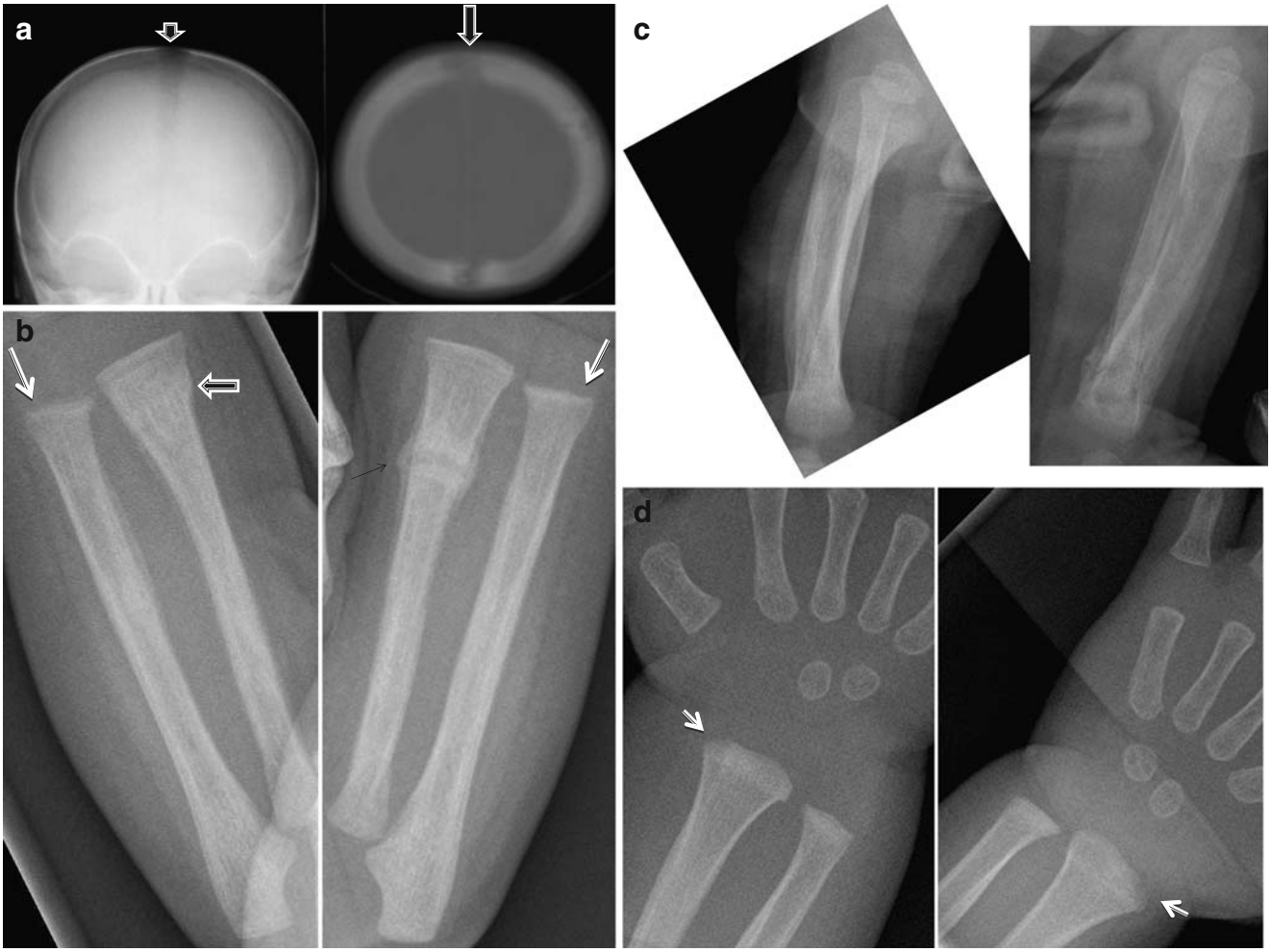

Fig. 3 Case 3, a 4-month-old Caucasian male. **a** Skull imaging. AP plain radiograph demonstrates a diastatic-appearing sagittal suture with poorly defined margins (*small arrow*). Axial CT image (bone algorithm) demonstrates that the demineralization along the sagittal suture (*large arrow*) and not diastasis is causing this radiographic appearance (i.e. pseudodiastasis). Brain CT scan showed no intracranial abnormality and there were no clinical signs of increased pressure. **b** AP views of the forearms demonstrate a transverse lucency through the distal radial diaphysis with surrounding sclerosis (*thin black arrow*). A lack of symptoms, angulation or displacement is important in differentiating fractures from Looser zones. There is a buckle fracture in the right distal radial metadiaphysis (*white outlined arrow*). Note the indistinct appearance of the distal ulnar metaphyses (*long white arrows*), which also show early cupping, while the distal radial metaphyses are still well defined. The early metaphyseal changes in the wrist typically present in the ulna before the radius. **c** Lateral views of the lower extremities show symmetric anterior bowing and broadening of the tibiae consistent with the saber shin deformity. This bowing occurs before weight bearing begins, and is due to the traction of the Achilles tendon on the calcaneus. **d** A rickets survey was performed 8 months after the initial presentation, when it was documented that the vitamin D level was 19.6 ng/ml and the alkaline phosphatase level was 2,386 U/l (normal range 145–320 U/l). During these 6 months, the infant had been formula fed. The radial metaphyses are deformed (*arrows*)

at the beginning of the winter. The infant was not checked for DD. Even her second child, born several months after the mother's DD was documented, was not checked for DD.

These were all term infants, with two born in the winter and two born in the summer (the African-American mother and the Caucasian girl with a vitamin D level of 8.7 ng/ml delivered in the summer). They were born north of the 35° latitude. There was no indication of suspected abuse in any of these infants prior to their first radiograph despite being seen by physicians, nurses, home health visitors, lactation consultants, day-care workers, audiologists, ultrasonographers, family and friends. At this level of inflicted injury, should there not be evidence of the battered infant syndrome? Should infants with so many inflicted fractures not be in severe pain or distress?

Recently published rickets data from Canada [30] reveal a similar pattern to the above cases. Of the 104 infants with confirmed rickets, 94% under 1 year of age were breast-fed without vitamin D supplementation (three were on formula, but developed hypocalcemic seizures within the first 3 weeks of life). No supplemented, breast-fed infant showed DD (Canadian recommendations are 400 IU/day).

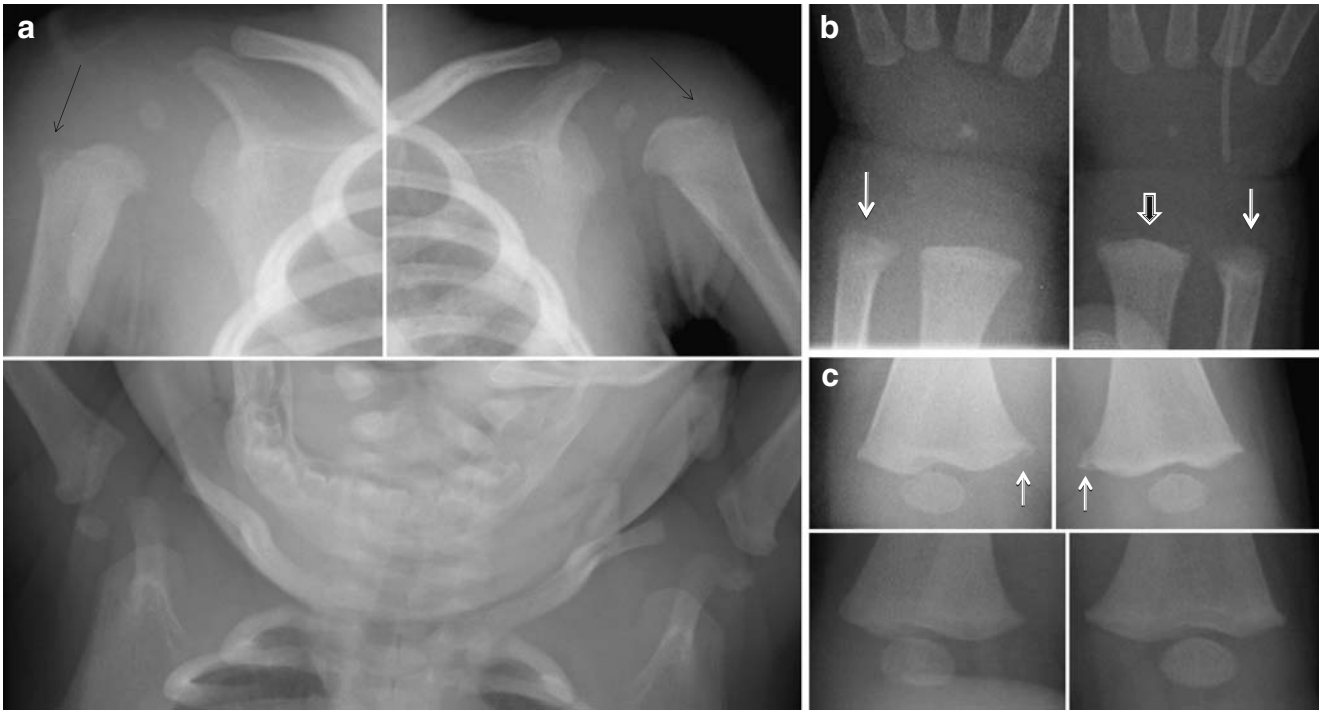

Fig. 4 Case 4, a 2-month-old Caucasian/African-American girl. **a** AP views of the shoulders demonstrate symmetric irregular metaphyseal mineralization along the lateral aspects that were asymptomatic (*top arrows*) and had resolved after a month of formula feeding without subperiosteal new bone formation or callus. **b** AP views of the wrists demonstrate the distal ulnar metaphyseal changes (*thin arrows*). While the right distal radial metaphysis remains normal in appearance, the left demonstrates early fraying (*thick arrow*). These were all asymptomatic on presentation and historically. **c** AP view of the knees demonstrate metaphyseal flaring and fragmentation that appear to represent classic metaphyseal lesions (*top*, *arrows*) although they were asymptomatic and resolved without subperiosteal new bone formation, callus, or remodeling after 16 days of formula feeding (*bottom*)

The number of cases reported per month from February to May was double the cases reported per month from June to December. The clinical and radiographic findings in the infants less than 1 year of age were skeletal deformities, fractures, hypocalcemic seizures, delayed development and failure to thrive. This is similar to the findings from the US in a review of rickets cases from 1986 to 2003, although the authors also reported three infants with rickets who were breast-fed with supplementation [31]. The current US recommendation of 200 IU/day is a recent reduction from the previous recommendation of 400 IU/day.

Early detection and treatment of DD is important as serious long-term effects are now being reported and adequate vitamin D levels are important beyond bone and dental health. DD has been linked to increased rates of type I diabetes (with a recent study suggesting a level relationship), schizophrenia, depression, autoimmune diseases, cancer, and asthma. In fact, nearly every tissue in the body has been found to have vitamin D receptors [3]. It is clear we are only beginning to understand what vitamin D does, and more importantly, what its deficiency implies.

Regarding the issue of classic metaphyseal lesions occurring in infants following uncomplicated cesarean section, these cases were diagnosed in the UK, where it has been demonstrated that 18% of pregnant women have vitamin D levels less than 10 ng/ml. In the UK food products are not required to be fortified, including milk. Should we assume that these deliveries were not as "uncomplicated" as reported, or should we consider the possible effects of the normal trauma of delivery (even in uncomplicated cesarean sections) on infants born with congenital rickets?

In summary, in infants less than 6 months of age with multiple asymptomatic metaphyseal lesions (particularly along the medial aspect of the distal femurs and proximal tibiae as well as in the distal tibiae and fibulae), pseudodiastasis of the sutures, transverse lucencies through the forearms and ribs, and compression fractures of the spine should alert the radiologist to the possibility of osteomalacia with early metaphyseal changes, insufficiency fractures and Looser zones. Clinical correlation is important, as most of these lesions are asymptomatic both at presentation and historically. With documentation of DD, 2 week follow-up radiographs may identify metaphyseal lesions that are resolving at an accelerated rate without interval subperiosteal new bone, callus or remodeling. While concomitant nonaccidental injury should never be excluded from the differential, it goes without saying that the suspicion or

presence of abuse should not preclude the diagnosis and treatment of DD. With subclinical DD being reported at such high prevalences, the radiologists' attention to these metabolic changes on radiographs will provide valuable input to help promote improvement in the vitamin D status of pregnant women and their newborn infants.

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EDITORIAL

Vitamin D insufficiency/deficiency – a conundrum

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In this issue of the journal, Drs. Kathy Keller and Patrick Barnes present a commentary on the nutritional state of children in different parts of the world. For radiologists, the vitamin D insufficiency/deficiency pandemic might come as a surprise; however, as Keller and Barnes point out, the actual vitamin D level in children throughout the world is both a nutritional and a cultural problem and is quite well reported: "… 52% of Hispanic and black adolescents in a study in Boston and 48% of white preadolescent girls in a study in Maine had 25-hydroxyvitamin D levels below 20 ng per milliliter" [1–3]. The accepted level for deficiency of vitamin D is less than 20 ng/ml and of insufficiency is less than 30 ng/ml. Holick's article [1] is quite informative and worth reading in its entirety. However, Keller and Barnes do not stop at informing us about this deficiency but go on to postulate that the lack of vitamin D in some children is responsible for skeletal lesions that are characteristic of child abuse. In a related commentary, Dr. Russell Chesney [4], noted nephrologist and chairman of the Department of Pediatrics at the University of Tennessee Health Science Center, helps us understand the pediatric view on both issues. A third commentary by Dr. Carole Jenny [5], head of the American Academy of Pediatrics Section on Child Abuse, discusses why the vitamin D problem and child abuse are clearly two separate entities and when they are, in fact, related and when they are not.

Have Keller and Barnes taken two separate entities and tried to connect them? Are they related, or is there another answer?

We believe that it is one of the responsibilities of a medical journal to publish articles that present data that force us to rethink our preconceived notions. We believe it is important that all pediatric radiologists understand this issue, as we play a focal role in the diagnosis of child abuse. We want our readership to digest these commentaries before reading the last commentary in this issue – the editors' point of view [6]. We would then like you to draw your own conclusions about this current conundrum.

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COMMENTARY

Rickets or abuse, or both?

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Pediatric radiologists are often the first physicians to make the diagnosis of either rickets or child abuse in infants and young children. In a busy pediatric radiology department, these diagnoses are not rare, and appear to be increasing in frequency [1, 2]. At least three issues are not always clear. First, the laboratory values of serum calcium, phosphate and alkaline phosphatase activity are broadly ranging in infants and children [3]. Hence, in an individual patient, the biochemical evidence of vitamin D-deficiency rickets can be problematic. Second, patients with metaphyseal lesions and/or rib fractures are sometimes said to have "pathognomonic findings" of either rickets or abuse [2, 4]. Fractures at these locations are found in a wide variety of inherited and metabolic disorders as well as in cases of trauma [5]. Third, these two conditions are sometimes found together, which I have personally seen.

The pathologic lesions in bone of a rachitic child include soft ribs, an enlarged costochondral junction, and irregularly thickened growth plates of long bones [4]. Histologically, islands of hypertrophic cartilage are evident within the metaphyseal trabeculae. The thickened, irregular trabeculae are often lined by wide osteoid seams. These findings account for the widening of the spaces between the physes and for fraying [4]. The pediatric radiologist must also be familiar with the constellation of skeletal findings in non-accidental trauma [2].

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We are in the midst of an epidemic of nutritional vitamin D-deficiency rickets that has been termed "the third wave of rickets" [6, 7]. The massive epidemic of rickets that occurred during the industrial revolution in northern cities in Asia, Europe and North America formed the first wave. It was due to the blocking of cutaneous ultraviolet irradiation by blackened skies due to universal coal burning. In this first wave, vitamin D deficiency with clinical evidence of rickets was found to affect up to 40–50% of children in various northern regions. The addition of irradiated ergosterol at 400 IU (a dose equivalent to 10 μg of vitamin D_2) to dairy products in the United States diminished the occurrence of rickets to the point that it became a curiosity [7]. Ironically, this quantity of vitamin D_2 was equal to that found in a teaspoonful of cod liver oil, a German remedy for rickets since the mid- to late 19th century [6, 7].

Factors contributing to the second wave were the universal use of breast milk by dark-skinned adherents of sects or religions that insisted that mothers wear covering robes and headdresses, which greatly limited sunlight exposure. Many of these individuals were Asians (Turks emigrating to Sweden or Germany and Indians to the United Kingdom), who moved to northern latitudes, and African-Americans living in northern United States cities [8]. This was a phenomenon of the 1980s. Pediatricians were reminded that human milk contained a suboptimal amount of vitamin D, and there were strong recommendations to supplement breast-fed infants with oral vitamin D [9].

The third wave has occurred since the mid-1990s and is largely found in breast-fed infants whose mothers are dark-skinned and remain indoors. This wave of rickets is very common in Canada, as well [10]. The finding of classic nutritional rickets in many infants has stimulated debate, and new American Academy of Pediatrics guidelines emphasize that all breast-fed American infants should receive an oral supplement of at least 200 IU (5 μg) of

vitamin D daily. The recommendation for Canadian infants is 400 IU [10, 11].

Inherent in each wave is that infants with rickets are born to mothers who are deficient or insufficient in vitamin D themselves [12]. Historically, several clinical factors combine to produce maternal vitamin D deficiency. During periods of industrialization, the smog of atmospheric particles from coal burning effectively blocked out ultraviolet light at the 288 nm wavelength necessary for vitamin D production. Many breast-feeding mothers are dark-skinned and wear robes or headdresses, such that the photocutaneous synthesis of vitamin D_3 from 7-dehydrocholesterol in skin cells is minimal or does not take place [8]. Currently, working mothers remain inside because of their jobs and the use of computers, and rarely go outside at a time of day when the incident angle of the sun is optimal for promotion of photocutaneous synthesis of the vitamin [13, 14]. Each of these factors can create a situation in which a woman who is vitamin D-deficient during pregnancy cannot transfer adequate stores of this prohormone to her fetus. Even with recommended doses of vitamin D in prenatal vitamins, this maternal vitamin D deficiency can be difficult to overcome [1].

The two forms of vitamin D—ergocalciferol, or vitamin D_2, and cholecalciferol, or vitamin D_3—are biologically equivalent in terms of healing rickets, but are derived from either dietary sources or supplements (D_2) or from sunlight (D_3). Each form of the vitamin must undergo further metabolic steps to produce the active hormone, 1,25 dihydroxyvitamin D [1]. This hormone enhances intestinal calcium and phosphate absorption from the gut. The importance of adequate circulating values of $1,25(OH)_2D$ is that it optimizes blood concentrations of calcium and phosphate, which allow mineralization of osteoid and suppression of excess parathyroid hormone synthesis and secretion. Since the zone of provisional calcification (mineralization) is between the metaphysis and the epiphysis, the infant or child with rickets has hypomineralization at the growth plate, producing the classic lesion of rickets. These undermineralized bones are soft ("osteomalacia"), can bend or bow and widen upon weight-bearing and are more liable to fracture [5].

A full and exhaustive discussion of metaphyseal lesions is beyond the scope of this article, but they are found both in rickets and in nonaccidental trauma [2, 13]. Of interest, metaphyseal lesions and fractures due to rough play or falls are found in the animal kingdom and have been reported recently in arctic foxes, alpacas, yearling steers and polar bear cubs [15–18].

The article by Keller and Barnes is highly informative with regard to the confusion concerning metaphyseal lesions in infants where abuse is considered. This paper points out how the differences between rickets and abuse are blurred. A partnership with the pediatrician is essential [19]. The child's history and environment are important. Historical information concerning feeding and sun exposure is also relevant. Laboratory studies in the child, including serum calcium and phosphate levels, and 25 hydroxyvitamin D and parathyroid hormone concentrations are indicated. Unfortunately, serum alkaline phosphatase activity is elevated both in rickets and in healing fractures. In the milieu surrounding suspected abuse cases, the full history can be difficult to obtain [19]. All of these factors are important to the radiologist when confronted with this scenario.

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COMMENTARY

Rickets or abuse?

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An article in this month's journal presents a thorough review of the long-recognized problem of vitamin D deficiency in pregnant women and young children. The authors then present several cases of infants with multiple bony lesions. In the case presentations they imply that these children were suffering from vitamin D deficiency rickets, although the diagnosis of rickets apparently was not made in any of the children.

The source of the cases was not mentioned in the article, although I suspect that these may have been cases sent to an expert by attorneys. With the exception of case 5, it also was not stated in the article if the children were diagnosed as having been abused.

The problem with such a series of cases is that it might leave the impression that children with metaphyseal lesions and fractures are likely to have vitamin D deficiency rickets. A "convenience sample" can be misleading because it exhibits the logic error embodied by the *availability heuristic* [1]. Our perception of the frequency of events can be skewed by the examples available to the observer. It is difficult to make generalizations from a series of extreme cases. A person looking down from an airplane at the tops of mountains poking through the clouds who never sees the valleys between them cannot describe the terrain in a meaningful way. The reporting of cases collected from a forensic practice (if this is, in fact, the case) might lead to a biased sample rather than a statistically valid sample. In my practice, a child protection program in a northern climate that evaluates over 1,800 children per year for alleged abuse or neglect, we have been checking every child with multiple fractures for metabolic bone diseases for several years and have not yet identified a single child with vitamin D deficiency. One of my colleagues, however, did find one child, a solely breast-fed 9-month-old with obviously demineralized bones.

Since I am not a radiologist, I cannot comment on the radiological interpretation of these cases. However, I would be quite surprised if some of these bony abnormalities were not related to maltreatment. It is unclear whether the authors of the paper are trying to redefine the previously described radiological characteristics of rickets. However, a careful correlation of radiographs and biochemical parameters in infants with proven vitamin D deficiency could be undertaken to examine the issue and address it as a valid research question. On the other hand, the careful clinical/pathological correlations of metaphyseal fractures that have been done by Dr. Paul Kleinman and his colleagues cannot be ignored and should continue to guide our practice until new discoveries are made using valid methodology [2–5].

Every case of multiple fractures or suspected child abuse should be carefully evaluated. The collaboration of pediatric radiologists and pediatricians is an important part of this evaluation. In the field of pediatrics, the development of the new subspecialty, Child Abuse Pediatrics, will set standards for pediatrics experts [6]. Three years of fellowship training (including research training), board examinations, and stringent professional standards for continuing education along with self and peer evaluation will certainly nurture experts in the field and promote excellence in clinical practice.

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COMMENTARY

Evaluating the data concerning vitamin D insufficiency/deficiency and child abuse

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Keywords Vitamin D · Rickets · Child abuse · Bone

There is absolutely no question that serum levels of vitamin D in children in sections of the population of the US, Canada and various parts of the world are lower than the accepted normal [1]. There are many reasons for this, and the American Academy of Pediatrics and others are addressing dosage requirement for basic supplementation of vitamin D [2, 3]. The connection, however, between vitamin D insufficiency/deficiency and fractures in children with otherwise normal radiographs is another issue. What is the evidence for fragility of bones in children with insufficient levels of vitamin D and even in those with deficiency levels if the radiographs are normal, that is, when there is no radiographic evidence of rickets?

The definition of rickets is "an interruption in the development and mineralization of the growth plate of bone, with radiographic abnormalities" [4]. Merely having insufficiency/deficiency of vitamin D levels in the blood *does not* constitute rickets. It is, therefore, incumbent to show radiographic changes in the 30–50% of infants and children with low vitamin D to claim that they have rickets.

What are the radiographic findings of rickets?

1. *Diminished bone mineralization*.

This is a difficult determination on plain radiographs except in the premature, very-low-birth-weight population. Our digital imaging makes it much more difficult to assess bone mineralization. Even further subjectivity goes into the face and skull evaluation in the neonate and young infant. Keller and Barnes [5] give one reference supporting the concept that the skull and facial bones have the earliest changes [6]. Let us look critically at this reference. There were a total of 25 patients said to have rickets although no data are given supporting this. Although 80% of their patients were said to have demineralization of the skull, there was no control group in that study to determine if they could, indeed, distinguish those patients from normal infants or show that "the best location to search for radiographic evidence of congenital rickets and nutritional rickets in infants less than 3 months of age is the cranium and facial bone" [5]. In the referenced article, only six patients were under 3 months of age. There is no mention of what, if any, other findings of rickets were present. Thus, a major point in the Keller and Barnes commentary in the diagnosis of radiographic findings of rickets is a very weakly supported one.

2. *Changes of growing bone found at the physis and metaphysis*.

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In growing patients, not sick prematures, these findings are most characteristic of rickets:

(a) Demineralization of the zone of provisional calcification. "The initial radiographic finding is rarefaction of the normally sharply defined zone of provisional calcification on the metaphyseal side of the growth plate so that the metaphyseal bone fades gradually into the lucent physeal and epiphyseal cartilage" [7].
(b) Metaphyseal cupping and fraying. The cartilage becomes disordered (from its normal columnar pattern) and the affected metaphysis becomes frayed and cupped. Because of the loss of mineralization in the zone of provisional calcification, the epiphysis and metaphysis are widely separated. This is the most valuable sign of rickets. If the metaphysis and epiphysis including the physeal lines appear normal, the patient does not have radiographic rickets (excluding prematures).

It is apparent in all the images of Keller and Barnes that the epiphysis and metaphysis are not separated and the physis is normal. There is no cupping and fraying. By definition, radiological rickets is *not* present in these images. Keller and Barnes cite cupping of the distal ulna metaphysis. This is well known to be a normal finding in young infants and *not* to be considered radiographic evidence of rickets when it is the *only* finding (no changes of the radius or changes at the knees) [8]. None of the infants described by Keller and Barnes as examples of "healing" have the expected pattern of mineralization of the zone of provisional calcification.

3. *Deformity from rickets (osteomalacia)*.

Vertebral compression fractures have been described in rickets in children with seizures and severe rachitic bone diseases. However, there have not yet been any reports of isolated vertebral compression fractures in patients with proven rickets that were believed to be due to vitamin D insufficiency. Pending such reports, the claim of Keller and Barnes of such a mechanism is questioned. Therefore, *isolated* vertebral axial load compression fractures *are not and cannot be* the result of rickets, as Keller and Barnes claim. There is *no* literature to support these claims.

Congenital rickets

Let us examine "congenital rickets." Keller and Barnes refer to patients with congenital rickets (infants less than 6 months of age) as having "normal-appearing bone to diffuse cortical rarefaction, fractures at birth, and metaphyseal fraying and cupping" [5]. These patients appear in Table 1 [9–14]. Three of the seven infants were premature and one was near term. Three of the mothers had renal failure or severe preeclampsia. Two other mothers had hypocalcemia or diminished vitamin D. All of the infants had abnormal physical examinations and all who had radiographs (six of seven) had abnormal findings; there were metaphyseal changes in all six infants.

Thus, it is not reasonable to assume that an infant with shaft fractures or vertebral fractures and no metaphyseal changes has congenital rickets or, without appropriate biochemical parameters, rickets of any sort. One additional point made by these cases shows that the disturbed maternal calcium homeostasis with a low exchangeable pool in mothers with osteomalacia is as important as vitamin D levels in causing the baby's problem.

The occurrence of fractures secondary to a metabolic disease is complex. In "congenital rickets," it is the maternal calcium homeostasis that probably plays a major role. In a recent case-control study by Olney et al. [15], 68 children with two or more incidences of low-energy fractures were compared with a control group (57 children) without fractures. Their ages ranged from 3 to 18 years. A significant number of children with fractures and control subjects had idiopathic hypercalciuria based on 24-h urine collection. These children (in both groups) had lower bone density. Though both groups (21% of the children with fractures and 18% of the controls) had insufficient vitamin D levels, this was not a significant factor in those who had recurrent fractures.

Where are we now in trying to connect vitamin D deficiency rickets and child abuse? Let us look once more at the cases Keller and Barnes submitted:

1. The authors do not give us their selection criteria for the patients presented, i.e. exclusion criteria and total pool from which they were selected. It appears that the patients were selected from among those involved in litigation concerning whether child abuse was present.
2. None of the children had vitamin D levels reported at the time they were supposed to have rickets.
3. None of the children had calcium phosphate, alkaline phosphatase, or parathyroid hormone values reported at the time they were supposed to have rickets.
4. All of the children were below 4 months of age. All of the "congenital rickets" reports summarized in Table 1 had abnormal radiographs in a manner described above for rickets. Among the patients presented by Keller and Barnes, however, there was not one who had a widened physis or, on the recovery films, had the characteristic appearance of healing rickets.

Table 1 Findings of infants with congenital rickets.

Reference	No. of patients	Maternal history	Baby's maturity	Baby's clinical status	Radiographic findings (as stated in article)
9	1	Renal failure	27 weeks/830 g	Hyaline membrane disease	Metaphyseal changes
10	1	Low vitamin D, 7.1 ng/ml	Term/2.75 kg	Craniotabes	Suture widening; metaphyseal changes at wrist
11	1	Severe preeclampsia; normal vitamin D	29 weeks/684 g; small for gestational age; premature	Normal at birth; respiratory distress at 2 weeks; elevated alkaline phosphatase; low calcium; died at 65 days	Day 1: fraying of metaphysis
12	2 A B	Normal calcium (6.5 mg/dl), phosphorus (5.5 mg/dl), PTH 32 pg/ml	Full term/3 kg; 36 weeks/2.4 kg	Hypocalcemic seizures on day 7: elevated alkaline phosphatase (52 KA units/100 ml); aminoaciduria at 2 weeks: craniotabes; calcium 6.8 mg/dl, phosphorus 5.5 mg/dl, alkaline phosphatase 70 KA units/100 ml	No radiographs at time of illness. At 2 weeks: normal skull; long bones acute rickets
13	1	Hypocalcemia 4.3 mE/l	Full term/2.5 kg	Craniotabes; prominence of costochondral junctions and widening of wrists	Metaphyseal changes: wrists and lower limbs generalized rarefaction, cupping and fraying
14	1	Renal failure, polyhydramnios	31 weeks/1.12 kg	Tetany at 3 days; low serum calcium phosphate; high parathyroid hormone	Fracture femur; fracture ribs; rickets long bones

5. The fractures shown are mainly that — fractures. The areas in which one expects to see signs of rickets in this age group are all normal.
6. The normal variant of a mildly cupped ulna with a normal radius is normal, and therefore not an example of rickets [8].

Are any, some, or all of these children abused? Diminished fractures and healing would not be expected until treatment was initiated. Did further fractures occur after initiation of child protection procedures as might be expected if vitamin D deficiency was present? The radiographic and limited clinical data of the cases presented suggest that a child protection team (or equivalent) needs to investigate the possibility of child abuse while continuing to consider other causes of injury. How many of these children had retinal hemorrhages or external signs of trauma? Were the fractures multiple and/or occurring at different times? What was the social situation? Was there any history to support accidental injury? While there are no data, in our opinion, to suggest any of the lesions described by Keller and Barnes are rachitic, we must keep an open mind until a full work-up, as described by the American Academy of Pediatrics [16], is fulfilled. Kleinman [17, 18] has enlightened us on the nature of the classic metaphyseal lesion, and over 15% of his text concerns the differential diagnosis of this lesion and the work-up of those diseases that may masquerade as child abuse.

The diagnosis of child abuse is a team effort. One must consider the *entire* situation. The entire clinical, laboratory, radiographic, and, most importantly, social evaluation must be taken into consideration before reaching a conclusion.

A final word about the vitamin D pandemic — the *denominator* is crucial. If vitamin D insufficiency/deficiency is so prevalent and this causes weakened bones, where are the increased cases of bone changes and fractures consistent with rickets? In particular, where are the birth-related fractures? With the accounts recording the low maternal vitamin D level, one would expect a much larger number of fractures, many of which should be clinically apparent. Perhaps other factors are necessary (disordered maternal calcium metabolism, increased urinary excretion of calcium, etc.) and are equal in importance for bones to be weakened [15].

In the article by O'Connell and Donoghue [19] that provides Keller and Barnes a foundation for their commentary, there were three classic metaphyseal lesions per 187,000 births or an incidence of 0.0016%. We are not given the denominator that is the total number of cesarean sections [20] but, in fact, even if cesarean sections accounted for one-third to one-half of all the deliveries, the incidence of classic metaphyseal lesions would only increase to 0.0048%. Perhaps O'Connell and Donoghue missed a clue as to why these babies were injured, such as the delivery technique or some unusual handling of the baby after delivery. While we do not know what caused the

babies' problems precisely, these lesions as shown by O'Connell and Donoghue are extremely rare and do not force us to postulate underlying abnormal bone.

In conclusion, the demonstration of vitamin D insufficiency/deficiency levels and the bone changes of rickets are not the same. Each must be considered separately. For these reasons and because of the other data described, we find that the connection made by Keller and Barnes between "rickets" and fractures they consider to be similar in appearance to those seen in child abuse is not based on any scientific data. Unfortunately, the current scenario is reminiscent of Paterson's "temporary brittle bone disease" [21]. This concept has remained without proof and has been discredited [22–25]. The work-up of child abuse considers a differential diagnosis including rickets but, unless there is reasonable evidence of rachitic bone disease, there is no scientific basis for confusing vitamin D insufficiency/deficiency alone with child abuse.

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