LETTER TO THE EDITOR

Reply regarding rickets vs. abuse: the evidence

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Editor's note: It is the policy of the journal to publish simultaneously, both online and in print, letters to the editor and the authors' reply. Because of the many revisions necessary before the authors' reply was accepted, Dr. Feldman's letter was mistakenly published online first. I apologize to Drs. Keller and Barnes for this inadvertent error in our editorial process. T.L. Slovis

Jenny [1], Chesney [2], Slovis and Chapman [3], and Feldman [4] seem to acknowledge the *evidence* that the highest rates of vitamin D deficiency (DD) are now being reported in undersupplemented breastfed infants younger than 6 months of age (including those born to mothers with DD). What is the *evidence* that DD can be congenital rickets? Greer [5] has concluded that "good evidence" exists that these infants are at *increased risk of rickets*. Jenny's article, referenced in Slovis and Chapman [3], on evaluating infants with multiple fractures states that a 25-OH-vitamin D level can be obtained "if rickets is suspected because of radiographic findings or *history*." We agree with her call for a careful correlation of radiographs and biomechanical parameters in infant DD as a valid research project. Chesney [2] states that he has witnessed DD in an

DD but provide no illustrations. In his recent review, Kleinman et al. [10] lists *rickets* at the top of the differential diagnosis for the classic metaphyseal lesion (CML). He states, "on occasion discrete osseous fragments resembling corner fractures may be identified in the absence of more dramatic signs of rickets." Is he not describing the early signs of "CML-like lesions" in rickets? In both his book, as referenced in Slovis and Chapman [3], and his recent review [10], Kleinman clearly identifies rickets (along with other conditions) as a mimic of abuse, including the metaphyseal

infant who was also "abused." This is not surprising since

the age range of infant DD overlaps the peak age range for

infant abuse [6]. In the face of this epidemic, why aren't

there more reports of radiographic evidence of rickets in

infants <6 months of age? Shouldn't these infants be at

increased risk for fractures? Are there really no radiographic

findings until the classic metaphyseal "cupping and fraying" occurs at 6 months or older? And do these classic changes

develop so rapidly that no one has ever identified them in

report radiographic abnormalities of rickets in infants with

Yorifuji et al. [7], Gordon et al. [8], and Ward et al. [9] all

their earlier stages?

In our case series, the radiologists originally described the bone findings as characteristic of abuse, often calling the bone mineralization normal, and provided no differential diagnosis to include bone fragility disorders. Yet, all these infants were asymptomatic and fit the classic demographic profile placing them in the highest-risk category for severe DD. This discrepancy between the radiographic findings and the clinical findings should not be ignored, particularly when the psychosocial evaluation of the caretaker shows no risk factors for abuse. Ruling out abnormal bone mineralization on radiography is

lesions, skull fractures, subperiosteal new bone formation, insufficiency fractures (e.g., Looser zones), and osteopenia.

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unscientific and introduces bias. We must develop a better way of testing bone mineral density (BMD) for osteopenia and demineralization in these infants [11].

By radiography and CT, in our cases, we observed parasutural demineralization (i.e. pseudodiastasis), as previously described by Resnick [12] and Swischuk and Hayden [13], which correlates clinically with craniotabes. We also question that an imaging finding of persistent lückenschädel (lacunar skull change) in these infants may also indicate craniotabes. Slovis and Chapman focused on the lack of physeal widening, no cupping and fraying, the lack of radial involvement, and no classic findings of healed rickets in our cases. Establishing the separation of the epiphyses from the metaphyses on radiographs when the epiphyses are either not ossified or in the early stages of ossification has not been reported as far as we know [14]. The classic changes of cupping and fraving are usually not identified until 6 months or older [3, 14]. Swischuk and Hayden [15] and Silverman and Kuhn [16] noted the involvement of the distal ulna out of proportion to the radius. The healing pattern that has been "classically" described in *infantile* rickets occurs with vitamin D therapy beyond formula feeding or vitamin D supplementation [14]. Would we not expect a difference in the radiographic appearance of healing rickets treated with 2,000-6,000 IU per day as compared with 200-400 IU/day (see our infant cases 3, 4)? Slovis and Chapman agree that compression fractures occur in rickets but state that an isolated vertebral compression fracture cannot result from rickets due to vitamin D insufficiency. It should be realized that such a child may have previously been D-deficient. Furthermore, Slovis and Chapman do not address the other classic deformities of rickets that are present in our series, e.g., anterior ribs ("rachitic rosary"), "saber shin" deformity of the tibia, bowed limbs, and Looser zones. They also avoid the issues of pathologic fractures and pseudofractures, including CMLs and Looser zones.

Our point regarding the O'Connell and Donoghue [17] article on CMLs occurring in uncomplicated cesarean sections in the UK was not to show whether this occurs frequently but to acknowledge that these authors (along with Slovis/Chapman) seem to make the inference that the injury was inflicted by the medical staff. Perhaps we should also consider that there may have been a predisposition in these infants? This is especially a concern in the UK, where 18% of pregnant women have vitamin D levels <10 ng/ml [14].

The four cases in our rickets vs. abuse commentary indeed represent alleged child abuse cases that we reviewed on behalf of the defense. In addition to our institutional work as radiologic consultants in child protection cases, including prosecution cases, we also volunteer our services in defense cases, as many other child protection physicians do. This is in compliance with the AMA and ACR codes of

ethics regarding medical expert testimony. In all four cases, no charges were submitted, but travel expenses were reimbursed (total \$2,500 for all four cases). In three of the four cases, we did contact and consult with the treating radiologists. To comply with the journal format of Pediatric Radiology, our original text, figures, and references were substantially reduced. We listed in every case the number and type of fractures each infant was alleged to have, but our commentary was focused on the bone findings that should alert radiologists to the possibility of DD (alone or coexisting with abuse). Also, none of these infants had retinal hemorrhages. In all four cases, the historical data fulfilled the criteria for congenital rickets due to maternalfetal and neonatal vitamin D deficiency plus radiographic findings previously published in the medical literature [14]. In all four cases, at presentation, infant vitamin D level determinations were recommended but not done.

Case 1 The clinical and radiographic information provided by Dr. Feldman for this case is incomplete. The diagnosis of abuse was initially made only from the radiographs. The forensic pediatrician recommended that no further bone fragility workup be performed. Social workers found absolutely no risk factors for abuse. The readers are invited to review Bodnar et al. articles [18, 19] and make their own judgment regarding misrepresentation of the literature. Dr. Feldman's comments regarding the literature about seasonal variations in maternal vitamin D levels must take into consideration sun exposure and dietary intake, both of which were deficient in this mother by history. Furthermore, both prenatal and neonatal vitamins have been shown to contain inadequate vitamin D. Dr. Feldman also misquotes the literature regarding vitamin D levels, including units of measurement, for both deficiency and insufficiency. The case was dismissed by the judge with the agreement of the attorney general.

Case 2 The judge ruled that the parents were responsible for the child's injuries. However, contrary to Dr. Feldman's account, the judge granted custody to the maternal grandparents, mandated further medical intervention for the child, and allowed parental visitation with the goal of eventual family reunification.

Case 3 Also contrary to Dr. Feldman's account, this case was dismissed including a full apology to the family from the state of jurisdiction.

Case 4 Dr. Feldman states that this infant had an "acute brainstem and cervical spinal cord injury," in addition to cervical vertebral and skull fractures. The infant also had extracerebral collections without signs of increased intracranial pressure or retinal hemorrhages. Dr. Feldman states that he published this case as an abusive cervical spinal



cord injury. In fact, on MRI the neuroradiologist was unable to date the injuries because they were not acute. Furthermore, the identification of a syrinx also indicated chronicity. The medical records showed that this infant was manually rotated during the delivery after the head had already presented. The head was twisted to bring the body into proper position for delivery. Maternal and infant vitamin D level determinations were not done. Despite the *history* of significant birth trauma, the judge found the parents responsible for the child's injuries.

In conclusion, we propose that: (1) Maternal-fetal and neonatal vitamin D deficiency (DD) exists and is increasing. (2) DD can produce imaging abnormalities prior to the classic changes of rickets and predispose the infant to "fractures." (3) The imaging findings of rickets are different in infants younger than 6 months of age compared to older infants. (4) These findings can mimic abuse (e.g., CMLs). (5) Infants with bone imaging findings suggestive of abuse should also have vitamin D level determinations. (6) Radiologists should provide a differential diagnosis when faced with bone imaging findings suggestive of abuse. (7) The significance of the imaging findings must always be considered in the context of the clinical, social, and biochemical aspects of the case. (8) It should be possible to collect reliable data (imaging findings, vitamin D levels, BMD) and test these hypotheses.

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