Letters to the Editor

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Fractures: Abuse or Rickets?

From
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Editor:

I read with interest the article by Perez-Rossello and colleagues (1) in the January 2012 issue of *Radiology*. However, their data do not support their conclusion that rickets and vitamin D deficiency are unlikely explanations for fractures in infancy.

The age of their study population (8–24 months) is considerably different from that of the population at greatest risk for fractures from alleged child abuse and neonatal rickets (<6 months) (2,3). The authors failed to cite relevant literature reporting alarmingly high deficiency rates (29%–46%) in neonates and the strong association to maternal vitamin D status (4). Yorifuji et al (5) recently reported subclinical rachitic craniotabes in 22% of newborns in Kyoto, Japan. Because vitamin D levels rise substantially in the 1st year of life (6,7), even in breast-fed infants, the authors should have assessed for signs of healing rickets that are ignored by the narrow Thacher criteria (8). They failed to screen for numerous relevant radiographic signs (eg, rachitic rosary, subperiosteal resorption, intracortical tunneling).

It is improper to conclude that rickets is rare while reporting an alarming 5.0%–7.5% incidence (1,9) among deficient infants—nearly 1% of the original sample population of healthy infants in Boston, Mass. It is perplexing that they concluded rachitic fractures are "rare" with a sample size of two and without skeletal survey, a considerably discrepant observation from that of Chapman et al (10), who reported a 17.5% fracture prevalence in a larger study using more comprehensive imaging. Screening for fractures by parental questionnaire, the details of which were omitted by Perez-Rossello and colleagues, is a relatively insensitive screening tool among a population susceptible to subclinical insufficiency fractures and occult birth injuries.

Two of the three radiologists who reviewed the radiographs also read the same radiographs from the original study (9) and could have significantly biased interpretations. The considerable decreased prevalence of rickets (from 7.5% to 5.0%) and demineralization (from 37.5% to 5.0%–12.0%) in the original versus current report also undermine the validity of the methodology. The disagreement rate among radiologists was unacceptable, particularly when considering that such decisions can determine the outcomes of child abuse investigations. The striking similarity between some rachitic changes and the deformities attributed to physical abuse adds further credibility to the recent concerns about misdiagnosis (3).

Disclosures of Potential Conflicts of Interest: Financial activities related to the present article: none to disclose. Financial activities not related to the present article: institution received money for expert testimony. Other relationships: none to disclose.

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Editor:

I am a retired clinical professor of pediatrics and was the founder and director of a child abuse team at a major children's hospital. For the past decade I have been studying bone science and bone injury in infants. I am a sponsored member of the American Society for Bone and Mineral Research. In the January 2012 issue of *Radiology* (1), Perez-Rossello and colleagues reported on a study regarding the impact of vitamin D deficiency on bone quality. However, their selection criteria seem problematic.

First, although Perez-Rossello and colleagues studied children ranging in age from 8 to 24 months, they derived the radiographic criteria for rickets from the study by Thacher et al (2)—where the average patient age was 4.5 years.

Second, because most multiple unexplained fractures of infancy, including cases of vitamin D deficiency, occur primarily in the first 6 months of life (3), their study population may not be a valid comparison group. It is conceivable that there are variations in bone physiology specific for the first 6 months of life that can be risk factors for bone fragility.

Third, Perez-Rossello and colleagues used the subjective Likert scale, which is known to be prone to biases (4), to assess demineralization.

The authors based their assessment on the undefined term "definite demineralization." It is not clear whether they considered "definite" focal demineralization such as that of the parasutural, metaphyseal, and subperiosteal areas that can be seen with vitamin D deficiency as well as other mineralization problems (5).

Because of the short half-lives of common biomarkers used for assessing bone and other factors that can affect them, the absence of abnormalities in these tests cannot exclude rickets or other forms of bone fragility (6). Classic radiographic findings of rickets are not usually seen in the first 6 months of life. However, there are often more subtle changes (7).

The effect of vitamin D on bone strength cannot be assessed by evaluating conventional radiographs. Vitamin D deficiency is a risk factor for bone quality and, therefore, for bone failure. Vitamin D and its metabolites affect the growth plate cartilage, bone turnover, differentiation and survival of distinct cell populations, and maintenance of bone homeostasis through positive and negative control of gene expression and epigenetic mechanisms (8).

Bone failure can be explained by vitamin D deficiency in association with other bone risk factors in genetically predisposed individuals but not by vitamin D deficiency alone.

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Response

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We welcome the opportunity to respond, as space permits, to Drs Ayoub and Hyman. With regard to their criticism of the methodology used in our study, we have the following replies: