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COMMENTARY

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

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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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