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Letter to the Editor

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European Society of Paediatric Radiology (ESPR) Child Abuse Taskforce Committee: a response to Miller et al.

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Dear Editor,

We write on behalf of the European Society of Paediatric Radiology (ESPR) Child Abuse Taskforce to respond to the article published by Miller, Stolfi and Ayoub (1). The diagnosis of physical child abuse in infants and young children is complex, and radiological imaging in suspected cases plays a pivotal role. The consequences of misinterpretation or misdiagnosis of the imaging obtained in these cases may have significant implications on the child and family unit. Both the medical and legal professions must rely on the extant robust and scientifically sound literature upon which to form opinions, both to appropriately prosecute suspected perpetrators and to defend those who have

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Rick van Rijn: Department of Radiology, Academic Medical Centre, University of Amsterdam, Amsterdam, The Netherlands. https://orcid.org/0000-0002-8876-6515 been falsely accused. The accuracy and legitimacy of both medical and legal decision-making in infant and child abuse cases can be compromised when reliance is instead placed on demonstrably scientifically unsound published work, such as this article (1).

Unexplained fractures in infants and young children, including classic metaphyseal lesions (CMLs) and posterior rib fractures, carry a high specificity for physical abuse. Several decades of well-established research exists, endorsed by specialist paediatric radiology organisations including the ESPR and the Society for Pediatric Radiology (SPR), and supported by recently published systematic reviews (2, 3). In contrast, in their article, Miller et al. speculate that unexplained fractures in infants and young children, including CMLs and posterior rib fractures, are the result of undiagnosed "metabolic bone disease of infancy" (MBDI). To support their outlier opinion, the authors have combined a multitude of maternal and infant risk factors to invent this new diagnostic entity, which (they say) has the radiographic signs of healing rickets. This is based solely on their own speculation.

Miller et al. opine that CMLs "often indicate an underlying bone mineralization disorder that would indicate bone fragility," which in their view proves that CMLs and posterior rib fractures do not have a high specificity for physical abuse. The authors' efforts to support this new argument are unavailing. They reference two of their own publications (4, 5) and two other articles wholly unrelated to metabolic bone disease: the first documenting rib fractures in infants after chest physiotherapy for bronchiolitis or pneumonia (6); and the second presenting three neonates with birth-related rib fractures (7). Leaving aside the authors' dubious terminology and unsubstantiated contentions, this article also includes several misconceptions, which if allowed to stand, may endanger vulnerable children.

Firstly, we address the fundamental issue of differentiating physiological from pathological radiographic appearances, overlooked by Miller et al. It is known that subperiosteal new bone formation (SPNBF) less than

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2 mm thick may be normal in the long bones of infants aged 1–4 months, in which case it is usually (but not invariably) symmetrical (8). Yet without disclosing their methods, principles, or research base, Miller et al. purport that they are able to distinguish this physiological SPNBF from pathological SPNBF, which they then assert is the first (of 7) radiographic findings in "undiagnosed MBDI".

In their Figure 3, Miller et al. present a radiograph of the distal radius and ulna in a 12-week-old infant and state that it shows an "abnormal growth plate of the distal forearm" with "significant ulna cupping" and "clubbing of the radius". This infant was also presented with "eight fractures including four rib fractures without internal thoracic injury and four CMLs". The authors have misinterpreted this radiograph which in fact is normal: cupping of the distal ulna is a well-known normal finding in this age group (9, 10). For a more detailed discussion on ulnar cupping and other normal variants, readers are directed to articles by Quigley and Stafrace (11) and Eide et al. (12).

In Figure 6 of their article, Miller et al. claim that a lateral spine radiograph in a 12-week (8-week corrected) female infant with, "risk factors for MBDI" is "abnormal" with "bone in bone in multiple vertebrae". This infant was also presented with "acute fractures of the distal left radius, left femur and left fifth rib" and "healing fractures of the right fifth rib, the right radius and ulna and two

parietal bone fractures". Readers are again cautioned that this "bone in bone" may be a normal finding on spine radiographs but only in neonates and infants up to 2 months of age (11, 13). We further note that the infant in their Figure 6 also had a vertebral compression fracture of L3 upon which the authors failed to comment.

Secondly, readers should beware the effects of postprocessing on image quality and the risk of exclusive reliance on such images for diagnostic purposes. Miller et al. also present a three-dimensional (3D) surface rendered head CT reconstruction for the same 12-week-old female infant as in their Figure 6 (lateral spine radiograph, discussed above), relying solely on this reconstruction which they claim, "shows the isolated posterior and lateral regions of brown and black ... indicating skull mineralization defects" and that "the brown edges of the widened sutures show parasutural hypomineralization". Miller et al. interpret these findings as being the result of hypomineralization. We caution the reader that, as with any 3D surface rendering, this appearance can be replicated simply by the operator manipulating the 3D surface rendering and selectively reducing the bony "thickness" of the skull, as we demonstrate in Figure 1. In practice, mineralization is never assessed with 3D surface rendering precisely because it is a thresholding-based technique: the threshold can approximate the attenuation of bone in a healthy



Figure 1: Left lateral 3D reconstruction of the skull and proximal cervical spine from a head CT examination in an infant. The "thickness" of the surface rendered bone has been manipulated by the operator at the imaging workstation with (A) demonstrating baseline bone thickness and (B) to (F) demonstrating a progressive decrease in bone thickness. This process of decreasing the bone thickness from images (A) to (F) replicates brown and black "defects", and sutural "widening", with an associated "brown edge": findings claimed by Miller et al. (1) to represent "mineralization defects" or "parasutural hypomineralization" in infants with "metabolic bone disease of infancy". Skull vault thickness/bone density should not be determined from 3D reconstructions. Note the presence of an orogastric tube.

patient, resulting in inaccuracies in the reconstructed image (14). The opposite is also true – healthy bone may appear undermineralized, depending on the threshold (Figure 1). This effect of surface rendering is more pronounced in infants and young children because of their thinner skull vault as compared to older children and adults. None of these well-known and widely accepted limitations to 3D surface rendering are disclosed by Miller et al. We must caution readers that 3D CT reconstructions should never be interpreted without concurrent evaluation of the native axial images, which have not been provided in Miller et al.'s article.

Finally, we raise awareness amongst readers that a family history of Ehlers–Danlos syndrome (EDS) in a (first degree) relative does not provide a scientifically robust explanation for fractures in an infant. In these cases, other causes of the fractures must be sought. Shur provides an excellent critique on the false association between EDS and fractures in infants (15).

For the reasons outlined above, caution is required by any professional seeking to cite this article by Miller et al. to argue that an infant or child who presents with fractures that have a high specificity for physical abuse is instead suffering from an "undiagnosed MBDI." In their article, Miller et al. promulgate a flawed ideology specifically rejected by the wider international paediatric radiology community: we wholly endorse the recently published letter by Brown et al. (16) written on behalf of the SPR Child Abuse Committee.

Given our assessment that the Miller et al. article has an entirely inadequate evidence base and that the authors are unable to substantiate their unique interpretation of their own images, we have not commented on the remaining issues such as pregnancy history, medical history, risk factors for the so-called "MBDI" or the reliability of results of infant and maternal blood studies related to bone physiology. However, as practicing paediatric radiologists, we are of course in favour of any rigorous scientific study with a control group that further elucidates the sensitivity and specificity of these important non-radiological parameters.

In conclusion, the article by Miller et al. is speculative, muddled in thinking, weak in methodology and worrisomely incorrect in image interpretation. If taken at face value, it risks encouraging poor science and could mislead and misinform clinicians and courts, whose decisions impact the infants and young children whom it is our collective duty to protect from harm.

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References

- Miller M, Stolfi A, Ayoub D. Findings of metabolic bone disease in infants with unexplained fractures in contested child abuse investigations: a case series of 75 infants. J Pediatr Endocrinol Metab 2019;32:1103–20.
- Cardiff University. 1.6 Metaphyseal fractures. Child protection evidence systematic review on fractures: Royal College of Paediatrics and Child Health; 2018 [updated April 2018; 11 March 2020]. Available from: https://www.rcpch.ac.uk/sites/default/ files/2019-02/child_protection_evidence_-_fractures.pdf.
- Cardiff University. 1.1 Rib fractures. Child protection evidence systematic review on fractures: Royal College of Paediatrics and Child Health; 2018 [updated April 2018; 11 March 2020]. Available from: https://www.rcpch.ac.uk/sites/default/files/2019-02/ child_protection_evidence_-_fractures.pdf.
- Ayoub DM, Hyman C, Cohen M, Miller M. A critical review of the classic metaphyseal lesion: traumatic or metabolic? AJR Am J Roentgenol 2014;202:185–96.
- Miller M, Mirkin LD. Classical metaphyseal lesions thought to be pathognomonic of child abuse are often artifacts or indicative of metabolic bone disease. Med Hypotheses 2018; 115:65–71.
- Chalumeau M, Foix-L'Helias L, Scheinmann P, Zuani P, Gendrel D, Ducou-le-Pointe H. Rib fractures after chest physiotherapy for bronchiolitis or pneumonia in infants. Pediatr Radiol 2002;32: 644–7.
- van Rijn RR, Bilo RA, Robben SG. Birth-related mid-posterior rib fractures in neonates: a report of three cases (and a possible fourth case) and a review of the literature. Pediatr Radiol 2009;39: 30-4.
- Kwon DS, Spevak MR, Fletcher K, Kleinman PK. Physiologic subperiosteal new bone formation: prevalence, distribution, and thickness in neonates and infants. AJR Am J Roentgenol 2002;179: 985–8.
- Glaser K. Double contour, cupping and spurring in roentgenograms of long bones in infants. Am J Roentgenol Radium Ther 1949;61:482–92.
- 10. Oestreich AE. Distinguishing child abuse fractures from rickets. Pediatr Radiol 2018;48:305-7.
- Quigley AJ, Stafrace S. Skeletal survey normal variants, artefacts and commonly misinterpreted findings not to be confused with non-accidental injury. Pediatr Radiol 2014;44:82–93; quiz 79–81.

- 12. Eide P, Djuve A, Myklebust R, Forseth KF, Nottveit A, Brudvik C, et al. Prevalence of metaphyseal injury and its mimickers in otherwise healthy children under two years of age. Pediatr Radiol 2019;49:1051–5.
- 13. Williams HJ, Davies AM, Chapman S. Bone within a bone. Clin Radiol 2004;59:132–44.
- 14. Fishman EK, Ney DR, Heath DG, Corl FM, Horton KM, Johnson PT. Volume rendering versus maximum intensity projection in CT

angiography: what works best, when, and why. Radiographics 2006;26:905–22.

- 15. Shur N. A case of broken bones and systems: the threat of irresponsible testimony. Am J Med Genet A 2019;179:429–34.
- Brown SD, Moreno JA, Shur N, Servaes S. Society for Pediatric Radiology (SPR) Child Abuse Committee response regarding Miller, Stolfi and Ayoub's flawed theories about child abuse and metabolic bone disease. J Pediatr Endocrinol Metab 2020;33:547–8.