Reply to Letter to the Editor

Unexplained Fractures: Child Abuse or Bone Disease

A Systematic Review

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We thank Dr. Lang for further clarification regarding our article. We are happy to address her concerns.

First, regarding the issue of inclusion of the article by Paterson et al. [4], we acknowledged in a response to a previous Letter-to-the-Editor [3], that the existence of “temporary brittle bone disease” as a variant of osteogenesis imperfecta (OI) has been challenged and in some ways shown to be nonscientific. However, all authors of the original paper [2] were unaware of the court ruling from the United Kingdom related to the study by Paterson et al. [4], and we welcome the clarification provided by Dr. Lang. This point is even more relevant in light of the fact that the MOOSE group (Meta-analysis of Observational Studies in Epidemiology) state in their guidelines that because observational studies are vulnerable to bias, confounding, and chance, readers should consider explanations for the observations other than the conclusions of the authors [5]. However, even had we known a priori about the court ruling, the study by Paterson et al. [4] still would have met our search criteria and our inclusion and exclusion criteria. As such, strict observation of systematic review technique would dictate that it be included as the

article has not been retracted from its originally published journal. However, we would have made note of the controversy and indicated that his ‘disease’ has been challenged, and that Dr. Paterson has been banished from medical practice. We by no means support the existence of “temporary brittle bone disease” or the work of Dr. Paterson; we simply reported the results of our systematic review. Exclusion of the article would have made our study a selective, not a systematic review because we would have excluded an article that met inclusion and exclusion criteria simply because we did not agree with the authors or their findings. Furthermore, we do not believe that inclusion of the article discredits our entire study. In our article, reference to the work by Paterson et al. [4] regarding temporary brittle bone disease [4] is made only sparingly in the article. Furthermore, of the 914 cases we pooled in our study, the article by Paterson et al. contributed only 39 cases (less than 5%). The omission of their article (based on the facts provided) would not have changed our main conclusion: various bone disorders (primarily OI, rather than temporary brittle bone disease) might be mistaken for child abuse. The main result of our study [2] which would have been affected is the following: “Pooling the studies in which confirmed cases of metaphyseal dysplasia/OI were present, there was an 18.7% (162 of 866) rate of initial misdiagnosis of CAN [child abuse and neglect] or bone disease.” Through exclusion of the article by Paterson et al., that sentence should now read: “Pooling the studies in which confirmed cases of metaphyseal dysplasia/OI were present, there was a 15.7% (130 of 827) rate of initial misdiagnosis of CAN or bone disease.” The purpose our study was simply to note that bone disease (particularly OI) can be mistaken for child abuse and neglect, and should be in the mind of the clinician. The exclusion of the work by Paterson et al. does not change that conclusion.

Re: Pandya NK, Baldwin K, Kamath AF, Wenger DR, Hosalkar HS. Unexplained fractures: child abuse or bone disease: a systematic review. *Clin Orthop Relat Res.* 2011;469:805–812.

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Second, Dr. Lang proposed that we did not correctly cite the data in the 1980 study by Horan and Beighton [1]. The cited article was a review of notes and radiographs of patients with reported bone disease. Dr. Lang stated that we incorrectly presented the information by including 13 children (six in the bone disease category and the remaining seven in the child abuse and neglect category). Dr. Lang believes that the exact data included 15 patients. The actual text of the article reports on 13 cases (representing 15 patients, two of the cases represented two siblings which we treated as a ‘case’). We should have clarified this in our manuscript using the word “cases” as opposed to patients, but our analysis remains the same. On page 246 of the article in Table 1, 13 cases (representing 15 patients) are listed with complete data regarding their initial diagnosis and final diagnosis [1]. From this table, there are six cases in the bone disease category, and seven in the child abuse category. We should have clarified this in our manuscript, but believe that data should still be reported as 13 cases – with our subsequent proportions accurate. Regardless, this once again does not change our conclusion.

Finally, Dr. Lang takes issue that we state in our article [2] that: “In many instances, a physician’s own instincts make the diagnosis.” She isolates this statement out of the context of the paragraph. The entire paragraph reads, “The limitations of our study and the literature are not trivial. First, there is no gold standard test for diagnose [sic] CAN; it remains a synthesis of history, physical examination, and radiographic findings. In many instances, a physician’s instincts make the diagnosis. Therefore, any study of CAN is fraught with the danger of misclassification.” This statement is placed in the limitation section of our study as a limitation [2]. This is not our recommendation. We even state in the following sentence that since physicians

incorrectly rely on instincts (not a gold standard test), child abuse and neglect are fraught with misclassification.

We do not believe, as Dr. Lang states, that the results of our study are inherently incorrect. Our study still reports on 827 cases of metaphyseal dysplasia/OI of which 130 were misdiagnosed as CAN [2]. These data are simply from the systematic review we performed, and we are reporting the results of this review. Exclusion of the 39 cases of Paterson et al. [4] does not inherently change the conclusions of our study as they represent a minimal percentage of our total cases. Furthermore, our use of the patients from the Horan and Beighton [1] study as 13 cases as opposed to 15 patients does not change the results of our study, and represents how the authors reported their data. Our study simply summarizes the literature present regarding bone disease and child abuse and neglect.

References

1. Horan FT, Beighton PH. Infantile metaphysal dysplasia or “battered babies”? A reassessment of material in the Fairbank Collection. *J Bone Joint Surg Br.* 1980;62:243–247.
2. Pandya NK, Baldwin K, Kamath AF, Wenger DR, Hosalkar HS. Unexplained fractures: child abuse or bone disease? A systematic review. *Clin Orthop Relat Res.* 2011;469:805–812.
3. Pandya NK, Baldwin K, Kamath AF, Wenger DR, Hosalkar HS. Reply to letter to the editor: unexplained fractures: child abuse or bone disease? A systematic review. *Clin Orthop Relat Res.* 2011 Jun 22. [Epub ahead of print]
4. Paterson CR, Burns J, McAllion SJ. Osteogenesis imperfecta: the distinction from child abuse and the recognition of a variant form. *Am J Med Genet.* 1993;45:187–192.
5. Stroup DF, Berlin JA, Morton SC, Olkin I, Williamson GD, Rennie D, Moher D, Becker BJ, Sipe TA, Thacker SB. Meta-analysis of observational studies in epidemiology: a proposal for reporting. Meta-analysis Of Observational Studies in Epidemiology (MOOSE) group. *JAMA.* 2000;283:2008–2012.