BAISHIDENG PUBLISHING GROUP INC
8226 Regency Drive, Pleasanton, CA 94588, USA
Telephone: +1-925-223-8242 Fax: +1-925-223-8243
E-mail: bpgoffice@wjgnet.com http://www.wjgnet.com

ANSWERING REVIEWERS

Name of journal: World Journal of Orthopedics
ESPS manuscript NO: 24872
Title: A novel case of Trevor's disease: adult onset and later recurrence

Reviewer's code: 03069318

COMMENTS TO AUTHORS

To my knowledge, the literature reports more than 3 adult cases. The authors report only the recently reported cases. They should consult a recent systematic review for more details. The authors should provide the MRI images too. Also, the authors should provide CT images for the recurrence that correspond to the level of the primary CT images, so as to be able to compare size. Why the initial lesion was not excised at the first surgery, since the pre-surgical diagnosis was osteochondroma? Imaging of the tibia post the initial excision is critical to support late recurrence. Did a histology showed removal of the lesion with clear margins? Also, the authors should report the exact time that the symptoms of recurrence occurred. Since the symptoms started several months prior the presentation 2.5 years after the initial treatment, this means that the recurrence might occurred earlier. This is critical, since a potential explanation of the recurrence may be that of inadequate initial management (curettage versus excision). The authors should comment and discuss this. Imaging of the 2-year follow-up showing the non-recurrence of the lesion is also important. Discussion The discussion can be improved by in depth presentation of certain aspects of management, e.g. what is the current gold standard treatment, what are the authors' recommendations, etc. Also, there is some inconsistencies between the case report and discussion (2.5years VS. 3 years, curettage VS. excision). In addition, the discussion would benefit from a more comprehensive discussion of the literature regarding recurrence. (Does it occur in children? Frequency? Adults? Factors affecting recurrence)

Response

After our subsequent literature search, we were able to find 3 more adult cases in the literature after consulting a more recent systematic review by Gokkus et al (most of the reported cases are pediatric). The initial lesion was entirely intra-articular which prevented it from being excised completely; as a result, the histology did not report clean margins. Thus, we mention in the discussion that inadequate management is a possible explanation for recurrence, as curettage was the only initial option, as complete excision would have left the patient deficient of too much articular surface. We were unfortunately unable to obtain the CT of the recurrence from the patient, but the cuts provided were taken to be approximately at the same level of the primary CT images in order to compare size. We also added to the discussion what the most recent literature states about the current gold

standard treatment, as well as our own recommendations, and clarified that recurrence specifically was reported at 2.5 years from the patient

Reviewer's code: 02726026

COMMENTS TO AUTHORS

Well written case report of an interesting and rare pathology. This article is of high interest for publication, but can be strengthened. First of all, it would be better if authors numbered the lines in order to facilitate revision. INTRODUCTION Authors say that disease affects mostly foot and ankle. What about the knee? Please re-revise the literature. There are more than 3 adult cases. 3 cases on D.B. Kettelkamp, C.J. Campbell, M. Bonfiglio Dysplasia epiphysealis hemimelica, a report of fifteen cases and a review of the literature 3 cases already cited in the present work T.J. Fairbank reports one 27yo case in Dysplasia epiphysialis hemimelica (tarso-ephiphysial aclasis) Another on V.M. Rosero, S. Kiss, T. Terebessy, K. K?ll?, G. Sz?ke. Dysplasia epiphysealis hemimelica (Trevor's disease): 7 of our own cases and a review of the literature Another: Sadeghifar AR, Heshmati AA. Dysplasia epiphysealis hemimelica (trevor syndrome) of talus in a 21-year old woman; case report. Arch Bone Jt Surg. 2014 Mar;2(1):66-8. Epub 2014 Mar 15. PubMed PMID: 25207317; PubMed Central PMCID: PMC4151426. DISCUSSION Why authors performed curettage instead of resection at the first surgery? Please discuss on that. Do authors have CT or X-rays of the 1 or 2year follow-up to ensure this was really a late recurrence? Since it is the only recurrence in adults, please discuss recurrence in children and possibles explanations, so we can avoid it in our patients.

Response

Please see above highlighted response for answer to why curettage was performed. We incorporated the studies mentioned by this editor and added the additional ones we found in the literature.

Reviewer's code: 02705063

COMMENTS TO AUTHORS

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This is a well written case report about a new-onset proximal tibial dysplasia epiphysealis hemimelica (DEH) in an adult recurring approximately 3 years after currettage. There are only 3 cases of adult DEH described in the literature to date, with no case reporting on recurrence of this pathology after surgery. The recurrent lesion was resected and after a 2 years follow-up the patient remains symptom-free with no appreciable mass recurrence. The authors should address the following points and then re-submit: 1. Please provide MRI images from the primary lesion in the Figures section. 2. Please describe the first and second surgical procedure in detail. It is important to learn from possible faults during the first operation. Why does the patient underwent an open curettage of the right lateral proximal tibial lesion during the first operation? Why not a complete excision initially? Maybe resection was not sufficient during the initial operation which may explain why the patient had recurrence of the lesion. Please discuss this point. 3. Special molecular tests are used for the analysis of genetic expressions to differentiate between DEH and osteochondroma. These are within normal ranges in DEH, while they are lower in ostechondroma. Did you perform genetic analysis to confirm diagnosis of DEH? Nevertheless, this is a very interesting case report which should be of interest to the readership of World Journal of Orthopaedics.

Response

Please see above highlighted response for why patient underwent curettage. Genetic analysis was not performed to confirm diagnosis of DEH because it was unavailable at the institution.