Pediatric Rheumatology

CORRESPONDENCE

Open Access

Significance of differential diagnosis for the elucidation of rare and complicated diseases

Jiro Ichikawa[1*], Tomonori Kawasaki[2], Kojiro Onohara[3], Masanori Wako[1], Satoshi Ochiai[4] and Tetsuo Hagino[4]

Keywords Pigmented villonodular synovitis, Hemosiderotic synovitis, Magnetic resonance imaging, Histopathological findings, Differential diagnosis

We read with great interest the article by Karami et al. titled "Pigmented villonodular synovitis in pediatric population: review of literature and a case report" [1]. Pigmented villonodular synovitis (PVNS; now classified under tenosynovial giant cell tumors) is extremely rare in children [2]. Karami et al. conclude that PVNS can cause patellar dislocation; however, we suggest considering a different perspective on this issue. Hemosiderotic synovitis (HS) may be caused by trauma and is difficult to differentiate from PVNS [3]. Given that HS may result from patellar dislocation, we present the main points of differentiation between HS and PVNS. The influence of age and sex on HS remains unclear; however, HS in a child has previously been reported [4].

HS occurs after repeated joint hematomas and has various causes, including hemophilia, anticoagulant use, trauma, and osteoarthritis [2]. HS often affects the knee, with typical symptoms including swelling and pain. Similarly, PVNS frequently presents with knee pain and swelling; as such, it is difficult to differentiate from HS based on clinical presentation [2, 3].

Magnetic resonance imaging (MRI) is a powerful tool for examining intra-articular lesions, but faces challenges in distinguishing between HS and PVNS. Several characteristics on MRI in HS, such as lateral meniscus or cartilage damage, synovial thickening, and contrast enhancement level, might be of use in this regard [5]. However, these features were noted in only a few cases, none of which included pediatric patients; therefore, further studies are required [5].

Pathological examination remains crucial, with most cases being diagnosed based on histological findings. In our review of the histopathological findings in this case, mononuclear cells and osteoclast-type multinucleated giant cells, which are characteristic of PVNS, were less abundant; however, macrophages—especially hemosiderin-laden macrophages—were noticeably present [1]. HS is morphologically characterized by the presence of hemosiderin, mainly restricted to the inner cellular layer of the synovium, and a finer villous appearance compared to the plump papillary pattern seen in PVNS [3]. Therefore, to improve diagnostic accuracy, we recommend assessment of the histopathological presentation of PVNS while taking into account a potential diagnosis of HS. Furthermore, recent research has reported the usefulness of clusterin, colony-stimulating factor 1, and desmin immunohistochemistry for PVNS diagnosis [2].

The standard treatment for both HS and PVNS is surgery, and arthroscopic surgery may be considered more

*Correspondence:
Jiro Ichikawa
jichi@sb4.so-net.ne.jp
[1]Department of Orthopaedic Surgery, Interdisciplinary Graduate School of Medicine, University of Yamanashi, Chuo, Yamanashi, Japan
[2]Department of Pathology, Saitama Medical University International Medical Center, Hidaka, Saitama, Japan
[3]Department of Radiology, Interdisciplinary Graduate School of Medicine, University of Yamanashi, Chuo, Yamanashi, Japan
[4]Department of Orthopaedic Surgery, National Hospital Organization (NHO) Kofu National Hospital, Kofu, Yamanashi, Japan

beneficial; however, there are no reports on differences in clinical outcomes between arthroscopic surgery and open surgery for HS [3]. Additionally, in many cases of HS, osteoarthritis has already existed or will develop, and it is anticipated that joint replacement surgery will be performed in the future. However, the long-term prognosis is currently unclear.

We highlight this case from a different disease, although rare, HS. Atypical or uncommon conditions such as synchronous dislocation and rare tumors should be considered during clinical assessments. Careful assessment of clinicopathological information is warranted for their accurate diagnosis.

Abbreviations

PVNS	Pigmented villonodular synovitis
HS	Hemosiderotic synovitis
MRI	Magnetic resonance imaging

Acknowledgements
The authors thank Ms. Kahori Sano and Ms. Azusa Sakamoto for their secretarial assistance.

Author contributions
Conceptualization, all authors; acquisition, J.I., T.K., K.O., M.W., S.O formal analysis, J.I., T.K., K.O., M.W, S.O.interpretation, J.I., T.K., K.O., M.W, S.O. writing—original draft preparation, all authorswriting—review and editing, all authors. All authors have read and agreed to the published version of the manuscript.

Funding
None.

Data availability
No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate
Not applicable.

Consent for publication
Not applicable.

Competing interests
The authors declare no competing interests.

Received: 22 November 2024 / Accepted: 30 January 2025
Published online: 05 February 2025

References

1. Karami M, Soleimani M, Shiari R. Pigmented villonodular synovitis in pediatric population: review of literature and a case report. Pediatr Rheumatol Online J. 2018;16:6. https://doi.org/10.1186/s12969-018-0222-4
2. De Saint Aubain Somerhausen N, van de Rijn M. Tenosynovial giant cell tumor. WHO classification of Tumours. Soft tissue and bone tumours. Volume 3, 5th ed. Lyon IARC. 2020;133–6.
3. Jayalakshmi V, Chikhale NP, Mishra A, Cherian S. Nonhemophilic hemosiderotic synovitis of the knee: a case report and review of literature. Indian J Pathol Microbiol. 2014;57:473–5. https://doi.org/10.4103/0377-4929.138779
4. Khasawneh RA, Mohaidat ZM, Gharaibeh MM, Hdeeb A. Post-traumatic hemosiderotic synovitis of the knee mimicking pigmented Villonodular Synovitis on magnetic resonance imaging (MRI) in a child: a Case Report. Am J Case Rep. 2021;22:e931992. https://doi.org/10.12659/ajcr.931992
5. Ando T, Kato H, Kawaguchi M, Nagano A, Hyodo F, Matsuo M. MR imaging findings for differentiating nonhemophilic hemosiderotic synovitis from diffuse-type tenosynovial giant cell tumor of the knee. Jpn J Radiol. 2021;39:76–83. https://doi.org/10.1007/s11604-020-01034-z

Publisher's note