

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Popliteal artery pseudoaneurysm caused by non-penetrating trauma in a patient with hereditary multiple osteochondromatosis [☆]

Oscar Andrés Parada Duarte, MD^a, Juan Guillermo Arámbula Neira, MD^b,
Valeria del Castillo Herazo, MD^b, María Fernanda Oviedo Lara, MD^c,
Adriana Lucía López Polanco, MD^d, Andrés Felipe Durán Omaña, MD^e,
Andrés Felipe Herrera Ortiz, MD^{f,*}

^aRadiology Department, Hospital Universitario Erasmo Meoz, Cúcuta, Colombia

^bFaculty of Medicine, Universidad El Bosque, Bogotá, Colombia

^cFaculty of Medicine, Universidad de Pamplona, Cúcuta, Colombia

^dFaculty of Medicine, Universidad Libre, Barranquilla, Colombia

^eFaculty of Medicine, Universidad de Santander, Cúcuta, Colombia

^fRadiology Department, Universidad El Bosque, Bogotá, Colombia

ARTICLE INFO

Article history:

Received 3 October 2021

Revised 10 October 2021

Accepted 11 October 2021

Available online 12 November 2021

Keywords:

Pseudoaneurysm

Popliteal artery

Vascular lesion, Hereditary multiple osteochondromatosis

Osteochondroma

massage

ABSTRACT

Hereditary multiple osteochondromatosis is a genetic condition characterized by the appearance of numerous osteochondromas, which can cause pseudoaneurysms in rare cases. The following article describes a 15-year-old patient with a history of current massages as part of his gym routine, who arrived at the emergency department with 4 days of pain, and ecchymosis in the right popliteal region. Therefore, duplex ultrasonography and arteriography were performed, confirming the diagnosis of popliteal pseudoaneurysm, which was subsequently treated by open surgery, providing a satisfactory outcome.

© 2021 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

[☆] Competing Interests: None.

* Corresponding author

E-mail address: Afherrera@unbosque.edu.co (A.F.H. Ortiz).

<https://doi.org/10.1016/j.radcr.2021.10.025>

1930-0433/© 2021 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Introduction

Hereditary Multiple Osteochondromatosis (HMO) is an autosomal dominant genetic condition caused by mutations in the genes *EXT1* and *EXT2*, which generates the appearance of numerous osteochondromas (benign bone tumors associated with low rate of malignant transformation) [1,2]. HMO has a prevalence of 1 for every 50,000 individuals in occidental countries, with a man: woman relation of 1,5: 1 [3]. Clinically HMO tend to be asymptomatic in their initial stages; nevertheless, when the osteochondromas start growing, they may compress adjacent structures and generate complications such as bursa formation, nerve injury, or arterial pseudoaneurysm [4]. The development of arterial pseudoaneurysms due to HMO represents a rare complication of the disease reported in approximately 100 cases in the literature [1]. In this paper, we aim to discuss a case of a patient with HMO who developed a popliteal pseudoaneurysm secondary to current lower limb massages.

Case report

A 15-year-old male patient with a history of HMO arrived at the emergency department with 4 days of pain in the right lower limb with difficulties walking after a leg massage as part of his gym routine. During the physical examination, a patient with prominent knees deformities was evident, associated with edema, and ecchymosis of approximately 5 cm x 2cm in the right popliteal region (Fig. 1). Therefore, intravenous and intramuscular analgesia was established with dexamethasone, dipyrrone, and diclofenac.

Two hours after the initial assessment, the patient was reevaluated, showing persistent pain despite analgesic therapy. Due to lack of pain relief and the medical history of HMO, a comparative knee radiography was requested, showing multiple huge osteochondromas located in the distal femur, and proximal tibial of both knees (Fig. 2). To better characterize the osteochondromatous lesions and assess the possibility of malignant conversion, a 3D computed tomography (CT) reconstruction of the right knee was performed, showing a better depiction of the osteochondromas with no signs of malignant transformation (Fig. 3).

The patient persisted with pain; therefore, a right knee ultrasonography (US) was requested, showing a small intra-articular leakage with fluid in the suprapatellar bursa associated with a right popliteal artery pseudoaneurysm measuring 80 × 50 × 35 mm with a thrombus inside (Fig. 4). Additionally, multiple osteochondromas were seen, one of them closely related to the popliteal artery.

Once the diagnosis of popliteal pseudoaneurysm was performed, the patient was referred to the vascular surgery service, where a right lower limb arteriography was carried out to have a better anatomic detail before surgery.

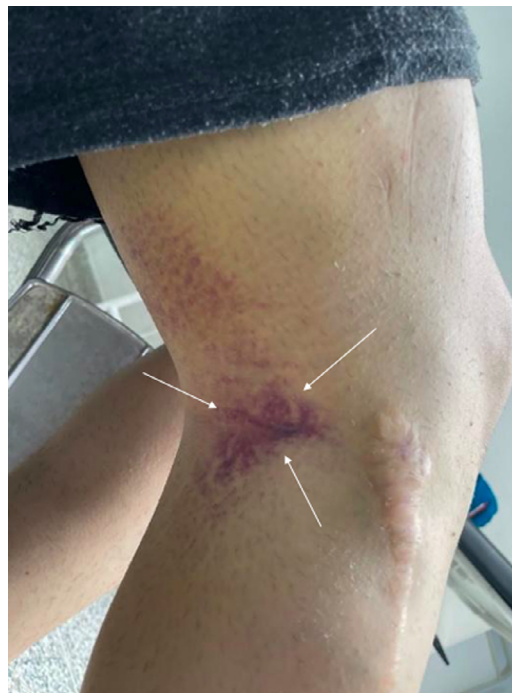


Fig. 1 – Ecchymosis located in the right popliteal region (white arrows).



Fig. 2 – Right knee x-ray in anteroposterior (A) and lateral views (B) showing multiple osteochondromas located in the distal femoral metaphysis (white arrows) and proximal tibial metaphysis (orange arrow) (Color version of the figure is available online.)

The right lower extremity arteriography showed multiple spiculated bone deformities localized in the distal metaphysis and epiphysis of the femur, closely related to the right popliteal artery in portions P1 and P2, leading to compression of the popliteal artery wall, and generating a partially

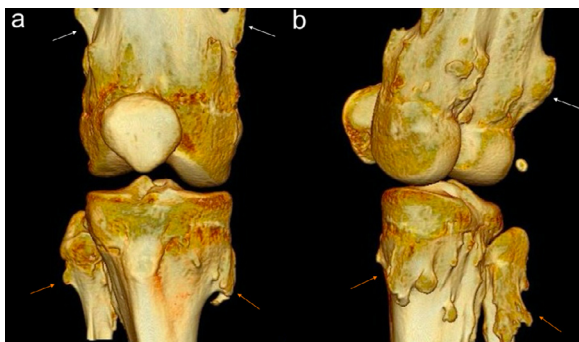


Fig. 3 – A 3D-CT of the right knee (A,B) showing multiple osteochondromas in the distal femur (white arrows), proximal tibia, and fibula (orange arrows) (Color version of the figure is available online.)

thrombosed traumatic pseudoaneurysm without compromising the distal blood flow of the extremity (Fig. 5).

Twenty-four hours later, the patient was referred for surgery. During the procedure, a big pseudoaneurysm of the popliteal artery with 350 cc of thrombosed blood content was observed. Therefore, the surgeon repaired the pseudoaneurysm, and resected the spiculated part of the osteochondroma related to the popliteal artery. A drainage was left to evacuate the remaining content.

After 48 hours of observation, the patient presented a good clinical evolution, for which discharge was given.

Discussion

Isolate osteochondromas have less than 1% rate of malignant transformation; nevertheless, when presented in the context of an HMO, the chances increase to 5% [5].

In rare cases, HMO can show vascular complications such as stenosis, arteriovenous fistulas, or pseudoaneurysms, which are usually presented in the context of a young male patient, as shown in this case [1].

Arterial pseudoaneurysms are the most common vascular complication caused by HMO. The lesion mechanism is due to a repeated abrasion of the arterial wall by a prominent ossified osteochondroma, which erodes the arterial surface, and generates the pseudoaneurysm [6]. Even though these pseudoaneurysms can be presented in any artery of the body, the popliteal artery represents the most frequent location of pseudoaneurysms in patients with HMO (77%) [1,5]. In this case, a constant trauma due to regular massages in the lower limb, as part of the patient's exercise routine, could have generated the chronic microtrauma between the distal femoral osteochondroma and the popliteal artery, causing a defect in the adventitia and subsequently producing the pseudoaneurysm (Fig. 6).

In patients with HMO, popliteal pseudoaneurysms manifest more frequently in the context of a non-traumatic event (65%) than in the context of a traumatic episode (35%). Clinically, popliteal pseudoaneurysms tend to manifest as pain and swelling on the popliteal fossa with preserved distal blood flow, as shown in this case. Nevertheless, the presence of ecchymosis or pulsatile mass in the popliteal region is not frequently observed but is highly suggestive of a popliteal pseudoaneurysm [6].

Duplex US is one of the most frequently used methods to confirm the diagnosis of pseudoaneurysms in patients with HMO because it can clearly depict the vascular lesion and its relationship with the osteochondroma [7]. Nevertheless, duplex US is less accurate in patients with obesity or huge hematomas due to more thickness between the vascular structures, and the transducer. Therefore, some authors have argued in favor of angiography as the imaging modality of choice to depict the anatomic details before surgery [8]. Nonetheless, it can lead to iatrogenic arterial lesions,

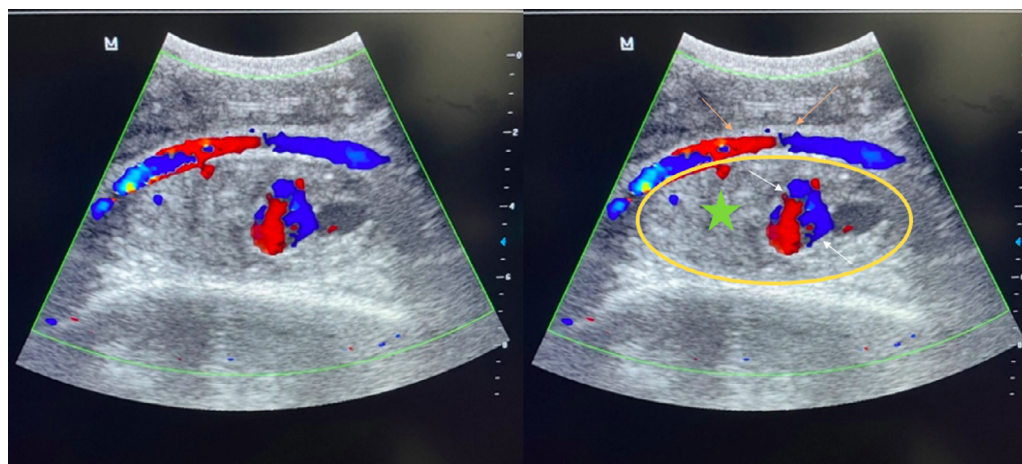


Fig. 4 – US of the popliteal fossa. A popliteal artery pseudoaneurysm measuring 80 x 50 x 35 mm (yellow circle) is observed with a thrombus inside (green star). Notice that the pseudoaneurysm is generating a turbulent flow within its lumen, producing the ying-yang sign (white arrows). Additionally, the popliteal artery lumen is being displaced by the pseudoaneurysm (orange arrows) (Color version of the figure is available online.)

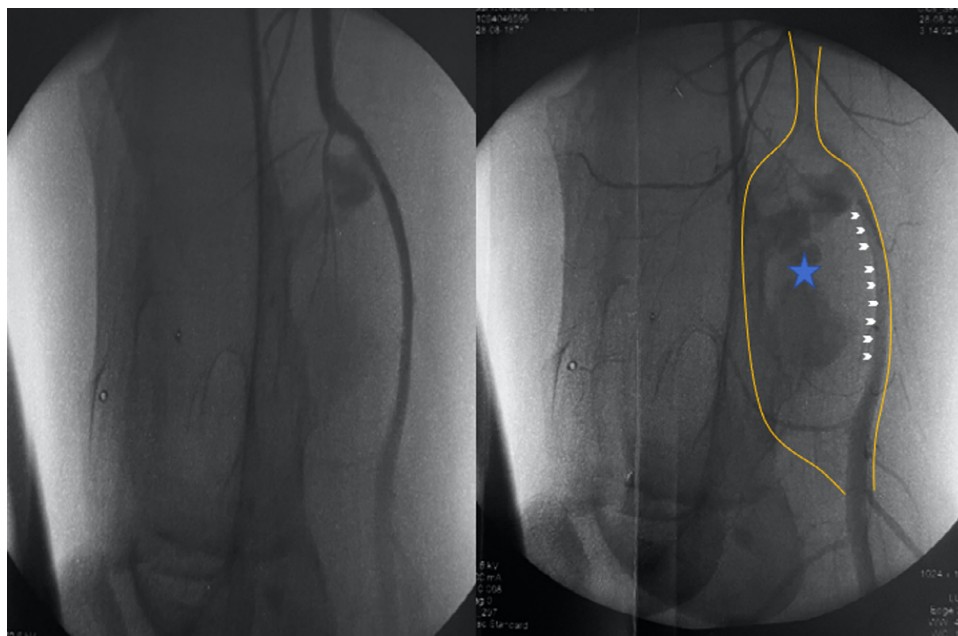


Fig. 5 – Right lower limb arteriography. A big pseudoaneurysm is seen (delimited by the yellow lines), with an abnormal contrast flow inside representing its lumen (blue star). Notice how the popliteal artery lumen is displaced by the partially thrombosed pseudoaneurysm (white arrowheads) (Color version of the figure is available online.)

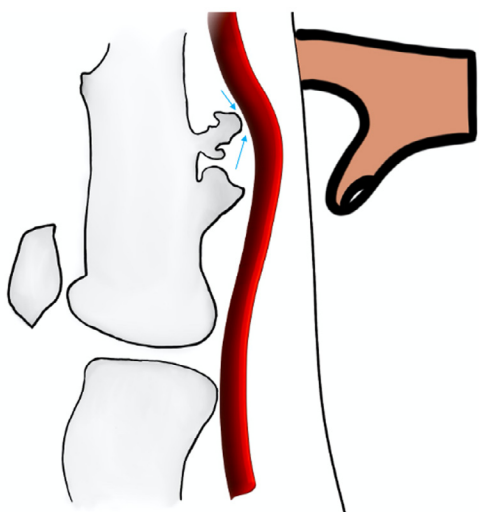


Fig. 6 – Illustration showing the physiopathological mechanism proposed in this case. Notice how a distal femoral osteochondroma is closely related to the popliteal artery (blue arrows), generating chronic friction by frequent massages in the lower limb, producing the pseudoaneurysm (Color version of the figure is available online.)

contrast medium allergic reactions, and misdiagnosis of the pseudoaneurysm when the artery has complete thrombosis because the contrast medium does not pass through the pseudoaneurysm [6]. For all above, the preferred imaging methods

nowadays to diagnose a pseudoaneurysm in a patient with HMO are CT angiogram, and magnetic resonance angiogram. These modalities are more expensive but can provide accurate and non-invasive anatomic details of the peripheral vasculature, in addition to an excellent depiction of the relationship between the pseudoaneurysm, and the osteochondroma, which represents vital information for surgical planning [7].

Looking retrospectively, we could have used a CT angiogram since the beginning to carry out the diagnosis of popliteal pseudoaneurysm, and avoid repeated radiological examinations to the patient; nevertheless, its clinical suspicion was not raised until the duplex US was performed. Therefore, it is mandatory to consider the diagnosis of popliteal pseudoaneurysm in every patient with HMO with an ecchymosis located in the popliteal fossa since this is the most frequent place.

In the duplex US, our patient presented with a pseudoaneurysm generating a turbulent flow within its lumen, producing the ying-yang sign. This bidirectional blood flow within the pseudoaneurysm acted as a risk factor to develop a thrombus inside, which ended up displacing the popliteal artery.

In the context of a patient with HMO with knee pain, it is mandatory to assess the appearance of the osteochondromatous lesions through radiological images to rule out the possibility of a malignant transformation of the bone lesions, as was done in this case by performing a CT.

Pseudoaneurysms in patients with HMO are treated with open surgery because it allows to simultaneously repair the vascular lesion and perform the excision of the osteochondroma that caused the pseudoaneurysm, as was done in this scenario [9].

Conclusion

In patients with HMO with popliteal ecchymosis, a popliteal pseudoaneurysm must be raised as the first differential diagnosis; therefore, the images modalities must be targeted to rule out this complication since the beginning by performing a CT angiogram or an MRI angiogram. Additionally, patients with HMO must receive proper education to avoid injuries related to chronic microtraumas, such as frequent massages.

Acknowledgments

None.

Patient consent

Verbal and signed consent was obtained from the patient concerned. The study was conducted anonymously.

REFERENCES

- [1] Bateman DK, Bar-Eli HY, Rahimi SA, Bowe JA. Post-traumatic pseudoaneurysm of brachial artery in multiple hereditary exostoses. *BMJ Case Rep* 2018;1(1):1–3. doi:[10.1136/bcr-2018-224353](https://doi.org/10.1136/bcr-2018-224353).
- [2] Pacifici M. Hereditary multiple exostoses: new insights into pathogenesis, clinical complications, and potential treatments. *Curr Osteoporos Rep* 2017;15(3):142–52. doi:[10.1007/s11914-017-0355-2](https://doi.org/10.1007/s11914-017-0355-2).
- [3] D'Arienzo A, Andreani L, Sacchetti F, Colangeli S, Capanna R. Hereditary multiple exostoses: current insights. *Orthop Res Rev* 2019;11(1):199–211. doi:[10.2147/ORR.S183979](https://doi.org/10.2147/ORR.S183979).
- [4] Yasuda N, Nakai S, Nakai T, Outani H, Takenaka S, Ichiro Hamada K, et al. A pseudoaneurysm of the popliteal artery probably pierced by a bone spike arising in the spontaneously regressed osteochondroma: a case report. *J Orthop Sci* 2021;26(3):514–20. doi:[10.1016/j.jos.2018.09.006](https://doi.org/10.1016/j.jos.2018.09.006).
- [5] Trivedi H, Link TM, O'Donnell RJ, Horvai AE, Motamedi D. Multiple hereditary exostoses: a pseudoaneurysm masquerading as tumor. *J Radiol Case Rep* 2016;10(8):50–9. doi:[10.3941/jrcr.v10i8.2849](https://doi.org/10.3941/jrcr.v10i8.2849).
- [6] Raheerintanainaina F, Rakoto-ratsimba HN, Rajaonnanahary TMA. Management of extremity arterial pseudoaneurysms associated with osteochondromas. *Vascular* 2016;24(6):628–37. doi:[10.1177/1708538116634532](https://doi.org/10.1177/1708538116634532).
- [7] Chamlou R, Stefanidis C, Lambert T, Munck D. Popliteal artery pseudo-aneurysm and hereditary multiple exostoses. *Acta Chir Belg* 2002;102(6):467–9. doi:[10.1080/00015458.2002.11679354](https://doi.org/10.1080/00015458.2002.11679354).
- [8] Perez-Burkhardt JL, Gómez Castilla JC. Posttraumatic popliteal pseudoaneurysm from femoral osteochondroma: case report and review of the literature. *J Vasc Surg* 2003;37(3):669–71. doi:[10.1067/mva.2003.48](https://doi.org/10.1067/mva.2003.48).
- [9] Boyacıoğlu K, Kayalar N, Sarioğlu S, Yıldızhan I, Mert B, Erentuğ V. Popliteal artery pseudoaneurysm associated with solitary osteochondromatosis. *Vascular* 2014;22(4):286–9. doi:[10.1177/1708538113486784](https://doi.org/10.1177/1708538113486784).