

Available online at www.sciencedirect.com

ScienceDirect

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



Case Report

Vascular complications of tibial exostosis: A case of popliteal vein thrombosis $^{\mbox{\tiny $^{$\!\!\!\! $}$}}$

Zied Mansi^a, Mohamed Taghouti^a, Mohsen Chamakh^a, Islem Chneti^a, Hedi Rbai^a, Wajdi Chermiti^b, Ali Haggui^c, Bacem Zaidi^d, Wael Gazzah^e

^a Department of Orthopedic Surgery, IBN EL JAZZAR University Hospital, Kairouan, Tunisia

^b Department of Orthopedic Surgery, SAHLOUL University Hospital, Sousse, Tunisia

^c Department of Orthopedic Surgery, Hospital of Kasserine, Kasserine, Tunisia

^d Department of General Surgery, IBN EL JAZZAR University Hospital, Kairouan, Tunisia

^e Department of Urology, IBN EL JAZZAR University Hospital, Kairouan, Tunisia

ARTICLE INFO

Article history: Received 5 March 2024 Revised 29 March 2024 Accepted 1 April 2024

Keywords: Osteochondroma Popliteal artery occlusion Popliteal vein thrombosis Treatment, Tibial exostosis

ABSTRACT

Tibial exostosis, also known as osteochondroma, is a common benign bone tumor found predominantly in adolescents and young adults. Vascular complications associated with this tumor, such as arterial occlusion and pseudoaneurysm formation, are rare but can lead to significant morbidity if not promptly diagnosed and managed. We present a case of a 25-year-old patient who presented with thrombosis of the left popliteal vein and a painless swelling in the popliteal fossa. Radiographic and CT angiography revealed an exostosis on the proximal tibia causing arterial occlusion and venous compression. Surgical resection of the exostosis via a posterior knee approach resulted in successful resolution of symptoms and a favorable outcome at a 12-month follow-up. Histopathological examination confirmed the benign nature of the tumor with no evidence of malignant transformation. This case highlights the importance of prompt recognition and surgical intervention in managing vascular complications associated with tibial exostosis. A multidisciplinary approach involving orthopedic and vascular specialists is crucial for achieving optimal outcomes in such cases. © 2024 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

Osteochondromas represent common benign bone tumors, predominantly encountered in adolescents or young adults. They may exist in sessile or pedunculated forms, with approximately 85% presenting as solitary lesions, while 15% are associated with hereditary multiple exostoses (HME), an autosomal dominant genetic disorder. Despite often being asymptomatic and discovered incidentally, symptoms may arise from compression or dislocation of neighboring structures, such as vessels, nerves, fractures, osseous deformities,

https://doi.org/10.1016/j.radcr.2024.04.005

^{*} Competing Interests: The authors confirm that they have no conflicts of interest associated with this publication. The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

E-mail address: doc.zm@hotmail.fr (Z. Mansi).

^{1930-0433/© 2024} 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/)



Fig. 1 - Radiographic image showing exostosis on the posterior aspect of the proximal tibia.

bursa formation, or malignant transformation [1]. Indicators of potential malignant transformation include a cartilage cap thickness exceeding 2 cm in adults or 3 cm in children, the onset of new pain or growth, or rapid lesion expansion, particularly after the closure of the growth plate [2]. The lower extremities, specifically the metaphyseal structures of the femur and tibia around the knee joint, are commonly affected [3,4]. Surgical resection is recommended for symptomatic lesions, complications, cosmetic concerns, or malignant transformation, with local recurrence occurring in less than 2% of cases when complete resection is achieved.

Vascular complications associated with osteochondromas are rare and involve vessel perforation, thrombosis, arterial thromboembolic events, and pseudoaneurysm formation [3]. Pseudoaneurysms are more frequently observed than thromboembolic complications from an epidemiological standpoint [5]. The pathogenesis and development of these vascular complications remain unclear, as exostoses are typically not detected until secondary symptoms manifest. Only a limited number of cases involving osteochondromas of the proximal tibia causing popliteal artery occlusion and acute lower limb ischemia have been reported.

Case report

A 25-year-old patient urgently presented to the emergency department with a swollen and red leg, leading to the initial diagnosis of thrombosis of the left popliteal vein. In response, a treatment plan involving low molecular weight heparin was promptly initiated. Concurrently, the patient exhibited a painless, firm swelling in the popliteal fossa. Radiographic investigations further revealed the presence of an exostosis on the posterior aspect of the proximal tibia (Fig. 1). Subsequent CT angiography demonstrated close relationships with the popliteal vessels, resulting in arterial occlusion and venous compression (Fig. 2).

The patient underwent a surgical procedure known as exostosectomy, utilizing a posterior knee approach based on the Triquet method (Figs. 3 and 4). Postoperative recovery was uneventful, and a comprehensive histopathological examination of the excised exostosis definitively ruled out any malignant transformation. Impressively, at a 12-month follow-up, the patient demonstrated a favorable functional outcome, emphasizing the success of the intervention in restoring normal lower limb function.

This case underscores the significance of a multidisciplinary approach in addressing complex vascular and orthopedic issues. The timely initiation of anticoagulation therapy and the subsequent surgical intervention not only resolved the acute occlusion but also effectively managed the underlying vascular occlusion due to the tibial exostosis. The uneventful recovery and sustained positive functional outcome highlight the success of the comprehensive treatment strategy, providing valuable insights into the management of similar cases.

Discussion

Ninety percent of vascular complications associated with exostosis manifest as pseudoaneurysms [6], as illustrated in the postoperative radiograph confirming the total removal of the exostosis. The predilection of the popliteal artery for involvement stems from the frequent localization of exostoses near the knee and the anatomical constraints within the Hunter's canal, making it susceptible to injuries from repetitive knee movements [7,8].

An intriguing finding is that half of the patients experiencing vascular complications report a history of trauma, with ex-



Fig. 2 – CT 3D reconstruction and Angiography CT imaging illustrating close relationships with popliteal vessels and resulting arterial and venous compression.



Fig. 3 – Image depicting the posterior knee approach during exostosectomy based on the Triquet method.

ostosis aligning with a diagnosis of multiple exostoses in half of these cases [8,9]. In the current case, the manifestation of atypical symptoms at 25 years occurred without antecedents of trauma or prior exostosis-related conditions, despite the typical location affecting the popliteal artery.

Studies by Tornquist et al. [10] and Lowel et al. [11] highlight cases of acute popliteal artery occlusion associated with proximal tibial exostosis in individuals aged 45 and 21, respectively. The discussion emphasizes the need for prompt diagnostic measures when signs indicate a vascular concern in a young patient, initiating with a radiograph followed by Doppler ultrasound and/or angiography to diagnose complications and precisely delineate their relationships with the tumor [12].

Surgical intervention is underscored as urgent, particularly in cases involving arterial occlusion, where immediate measures such as thrombectomy or bypass with a venous graft may be necessary [10].

The discussion introduces controversy surrounding preventive measures for vascular complications, with varying viewpoints. Some advocate for a wait-and-see approach until signs manifest, while others argue for the systematic resection of any exostosis. The decision for preventive treatment is influenced by factors such as the presence of arterial Doppler signs or contact with vessels [13,14]. Aligning with the perspectives of Tornquist et al. [10] and Asselineau et al. [15], the authors assert that if concerns about vascular conflict arise based on the appearance and location of the exostosis, imaging should be employed to assess the relationships between vessels and the tumor. Subsequently, a recommendation for excision of the exostosis alone may be proposed based on this assessment.



Fig. 4 - The exostosis after excision.

Conclusion

Vascular complications associated with bone exostoses are rare but require prompt surgical treatment. The systematic removal of every exostosis located near a vascular pathway should not be routine. However, it should prompt the performance of a CT angiography in the presence of any abnormalities during clinical examination or an appearance raising concerns about potential vascular conflict.

Patient consent

Written informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

CRediT authorship contribution statement

Zied Mansi: Conceptualization, Formal analysis, Supervision, Validation. Mohamed Taghouti: Formal analysis, Investigation, Methodology, Resources, Writing – original draft. Mohsen Chamakh: Data curation, Formal analysis, Investigation. Islem Chneti: Investigation, Methodology, Resources. Hedi Rbai: Supervision, Validation. Wajdi Chermiti: Conceptualization, Investigation, Resources. Ali Haggui: Supervision, Validation. Bacem Zaidi: Investigation, Methodology. Wael Gazzah: Investigation, Methodology.

REFERENCES

- Alonso-Gómez N, Diego MC-D, Martínez-Izquierdo A, Sáinz-González F. Unusual clinical complication: acute lower limb ischemia caused by a tibial osteochondroma. Reumatol Clin 2019;15:182–4.
- [2] Tepelenis K, Papathanakos G, Kitsouli A, Troupis T, Barbouti A, Vlachos K, et al. Osteochondromas: an updated review of epidemiology, pathogenesis, clinical presentation, radiological features and treatment options. In Vivo 2021;35:681–91.
- [3] Argyriou C, Drosos G, Tottas S, Tasopoulou K-M, Kougioumtzis I, Georgiadis GS. A rare case of tibioperoneal arterial trunk entrapment caused by a fibular osteochondroma. Ann Vasc Surg 2019;55:308.e11–308.e16.
- [4] Tanigawa N, Kariya S, Kojima H, Komemushi A, Fujii H, Sawada S. Lower limb ischaemia caused by fractured osteochondroma of the femur. Br J Radiol 2007;80:e78–80.
- [5] Rossi G, Mavrogenis AF, Angelini A, Rimondi E, Battaglia M, Ruggieri P. Vascular complications in orthopaedic surgery. J Long Term Eff Med Implants 2011;21:127–37.
- [6] Negulescu M, Arend P. Thrombose d'un pseudo-anévrisme poplitée secondaire à un ostéochondrome du fémur : un cas atypique. Rev Med Brux 2014;35:436–9.
- [7] Kiefer E, Maraval M, Trico JF, Natali J. Anévrysme artériel compliquant une exostose ostéogénique de l'extrémité supérieure du tibia. Rev Chir Orthop 1978;64:155–62.
- [8] Vasseur MA, Fabre O. Vascular complications of osteochondroma. J Vasc Surg 2000;31:532–8.
- [9] Parratte S, Launay F, Jouve JL, Malikov S, Petit P, Bollini G. Rupture de l'artère brachiale etparalysie radiale secondaire àune exostoseproximale de l'humérus chez un enfant de 14 ans. Rev Chir Orthop 2007;97:186–9.
- [10] Tornquist FA, Leopardo CC, Segala TC, Hamd A. Vascular complication of osteochondroma. J Vasc Bras 2007;6:92–6.
- [11] Lowel B, Carl Magnus R, Pavel L, Erik GL. Popliteal artery thrombosis in a young woman, secondary to bony exostosis. J Cardiovasc Surg 1979;20:193–6.
- [12] Vallance R, Hamblen DL, Kelly IG. Vascular complications of osteochondroma. Clin Radiol 1985;36:639–42.
- [13] Asselineau A, Courbet PH, Lahoud JC. Faux anévrisme de l'artère fémorale compliquant une exostose. Rev Chir Orthop 1993;79:411–14.
- [14] Solhaugh JH, Olerud SE. Pseudoaneurysm of the femoral artery caused by ostéochochondroma of the femur. A case report. J Bone Joint Surg [Am] 1975;57:867–8.
- [15] Boscher Y, Lescalie F, Moreau P, Enon P, Enon B, Chevalier JM, et al. Thrombose artérielle poplitée sur ostéochondrome de l'extrémité supérieure du tibia. À propos d'un cas. J Chir 1984;121:327–30.