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Case report

# A solitary giant osteochondroma of the femur in the shape of a devil's head pushing back the superficial femoral artery: Case report and literature review

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ABSTRACT

*Introduction:* The most common benign bone tumors are osteogenic exostoses or osteochondromas. They occur during growth and are rarely the cause of vascular or nervous complications.
*Case presentation:* We present the case of a young 34-year-old patient who consulted for a swelling in his right thigh. The X-ray revealed a giant, exuberant bony tumor in the lower third of the femur. The CT angiography allowed us to see the repression of the superficial femoral artery without interfering with blood flow. By resecting a giant tumor resembling a devil's head, the obstacle on the vascular structures was removed. Histology confirmed the diagnosis of solitary osteogenic exostosis. The patient made a complete recovery and there has been no recurrence after one year of follow-up.
*Discussion and conclusion:* A solitary femoral diaphysis exostosis causing arterial compression is a rare complication. Resection and relief of artery compression should be considered early, before serious vascular sequelae develop, which can be irreversible and result in amputation. Better recognition and more comprehensive evaluation of these rare cases should be emphasized.

## 1. Introduction

The most common benign bone tumor is osteochondroma, also known as exostosis. It is often asymptomatic and accounts for 35 % of benign bone tumors and 8 % of all bone tumors, with a slight male predominance [1]. It is a bony outgrowth of an ectopic growth cartilage that develops and matures through the normal endochondral ossification process. It is a small benign lesion that usually affects the long bone metaphysis [2]. We present the case of a 34-year-old man with a historical form of a giant osteochondroma of the right femur, and we will describe the clinical, radiological, and therapeutic aspects with a review of the literature. This manuscript has been reported in line with SCARE's 2020 Criteria [3].

## 2. Case presentation

This is the observation of a 34-year-old with no pathological history. The patient is neither a tobacco nor an alcohol user. He consulted regarding an important swelling on the inner side of the right thigh that appeared 5 years ago and gradually grown in size. Clinical examination revealed a hard, firm, deep, and superficially adherent mass on the inner thigh. Furthermore, there are no inflammatory signs or skin ulcerations nearby. The limb's vascular and nerve examinations were both normal. The patient reported a moderated pain at the tumor site.

A standard X-ray of the thigh revealed a giant bony growth perpendicular to the axis of the right femur to which it is attached by its base, then fleeing in an axial direction from the knee joint (*stalagmite*), raising the possibility of an osteochondroma giant femur in its pedunculated form (Fig. 1).

The MRI revealed a 76 × 41 mm cortico-medullary bone mass at the expense of the lower third of the right femur, 5.5 cm from the knee and 24 cm from the coxo-femoral joint, with irregular contours, exophytic development, heterogeneous T1 and T2 signal, without diffusion restriction, and heterogeneously enhanced after gadolinium injection. The vastus intermedius, rectus femoris, and vastus medialis muscles were infiltrated with a 25 × 10 mm cap (Fig. 2).

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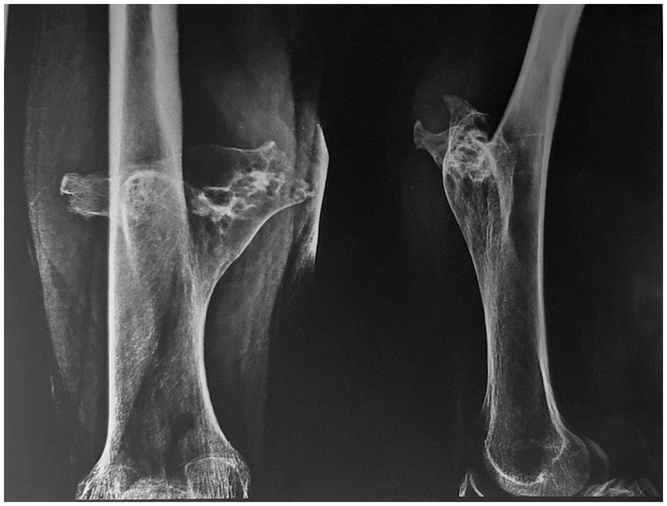

**Fig. 1.** A standard AP and lateral radiograph of the right femur reveals a giant bone formation developing perpendicular to the diaphyseal axis.

An angio-scan with 3D reconstruction showed the mass coming behind and pushing back the superficial femoral pedicle at the level of its passage at the level of the Hunter's canal, which remained permeable (Fig. 3).

A surgical biopsy (Fig. 4) confirmed the absence of malignancy, revealing spongy bone tissue and cartilaginous tissue locally covered with fibrous tissue, punctuated by a few inflammatory cells and some hemorrhagic alters.

Overall, this is a surface tumor in strict radiological continuity with the metaphyseal cortex from which it arose, with a cartilaginous cuff and no sign of malignancy, implying an osteogenic exostosis (osteochondroma) of the right distal femur pushing back the superficial femoral artery at the Hunter's canal.

With a direct antero-internal surgical approach (Fig. 5A), the osteochondroma was completely resected measuring 10 cm long axis resembling a devil's head (Fig. 5B). The surgery was performed by the same surgeons who performed the biopsy.

Standard AP and postoperative lateral radiography of the femur revealed complete exostosis resection with no bone defect and a relatively good aspect of the cortical bone, allowing the patient to bear weight without risk of fracture (Fig. 6). In addition, the postoperative vascular and nervous examinations were normal. There was no recurrence of the osteochondroma during the patient's 18-month follow-up period and the patient was satisfied.

## 3. Discussion

The most common benign bone tumors are osteogenic exostoses or osteochondromas. They happen during the growth phase. They start developing in childhood and continue to develop until puberty. They can affect any bone, but are most commonly found near the epiphyseal growth plate of the long bones. They can be solitary or multiple. Solitary exostosis is an osteochondral formation caused by the subperiosteal development of an aberrant growth plate island [4]. Multiple exostosis disease is an autosomal dominant disorder known as Hereditary Multiple Exostoses (HME) and characterized by the development of multiple bony outgrowths from the epiphyses [5].

Solitary exostosis is generally asymptomatic and they are discovered by chance during a radiological examination and rarely the cause of vascular or neurological complications. Vascular complications are an unusual and rare complication of solitary osteochondroma. Complications associated with HME include deformity, fractures, vascular impairment, pouch formation, malignant transformation, and neurological sequelae. Osteochondromas can have a direct mass effect on the artery, obstructing circulation and causing thrombosis. Exostosis involvement in the deep venous system accounts for 5 % of vascular complications. These various vascular lesions are clinically suspected

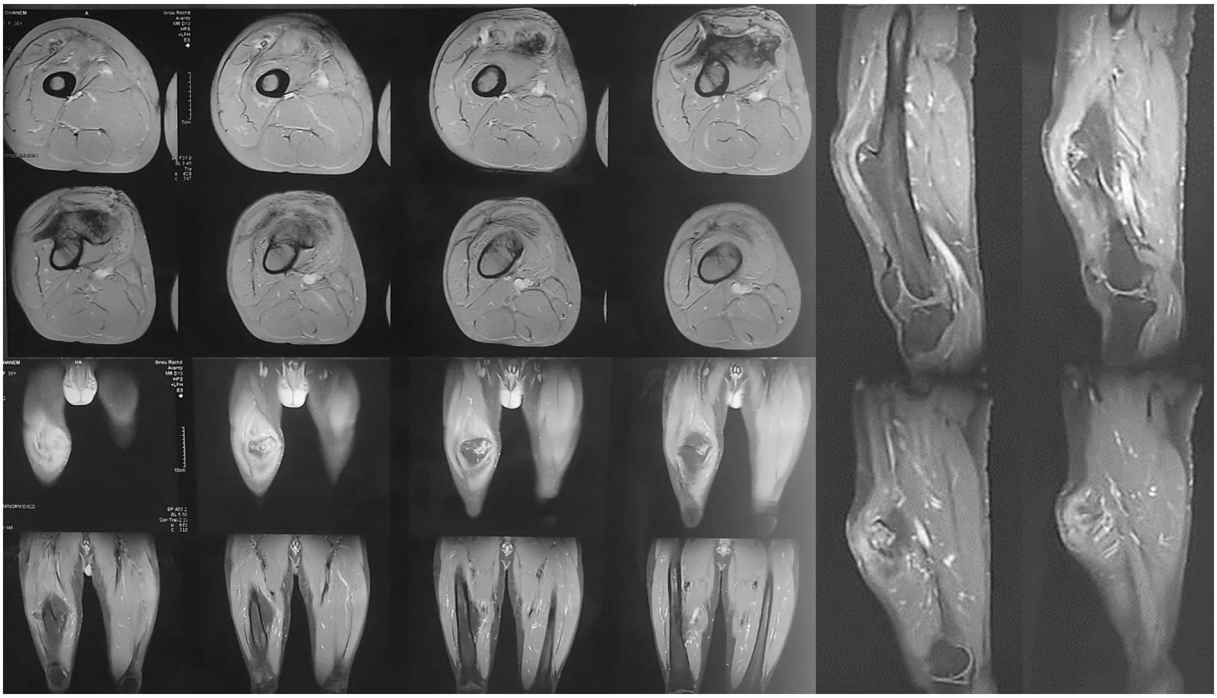

**Fig. 2.** MRI sections demonstrating osteogenic exostosis topped by a cartilaginous cap (identical density and bone structure and in continuity with the cortex and the spongy metaphysis).

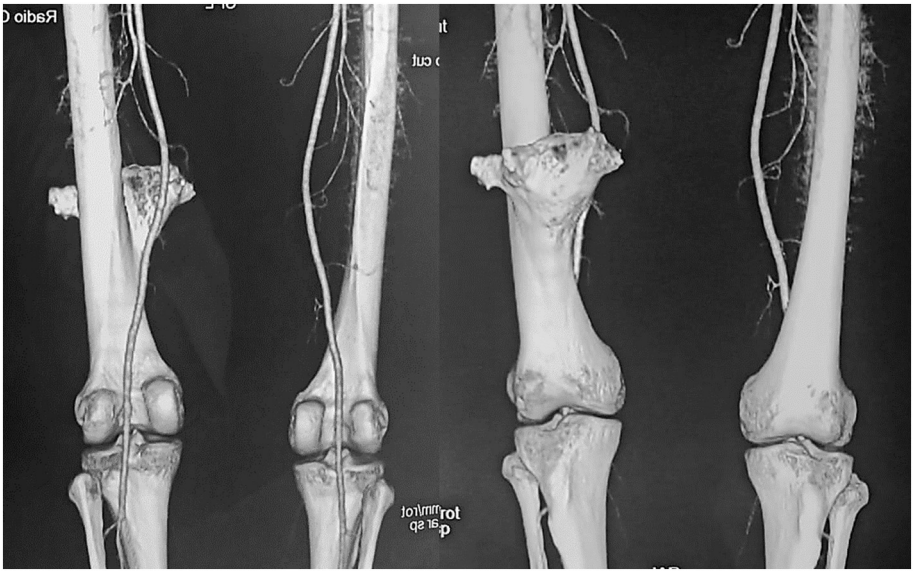

**Fig. 3.** Tridimensional reconstruction of CT angiography showing repression of the superficial femoral artery by the giant exostosis.

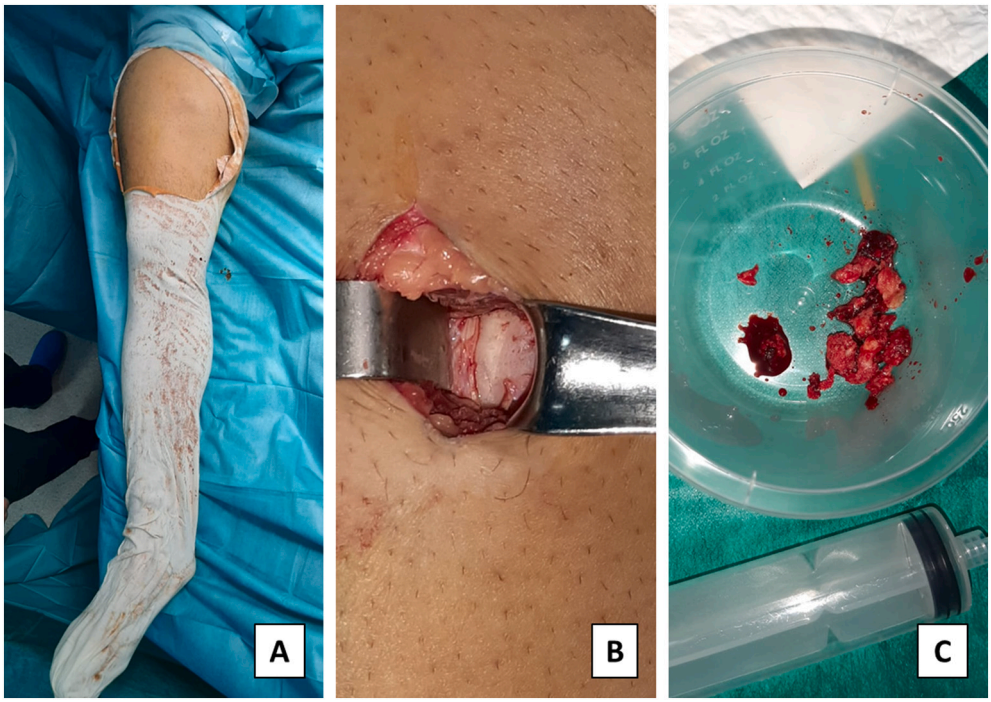


**Fig. 4.** A. Before the biopsy, the appearance of the right lower limb with a manifest swelling around the thigh. B. The biopsy was performed directly over the swelling on the right thigh's antero-medial part. C. The intraoperative characteristics of the biopsy specimens.

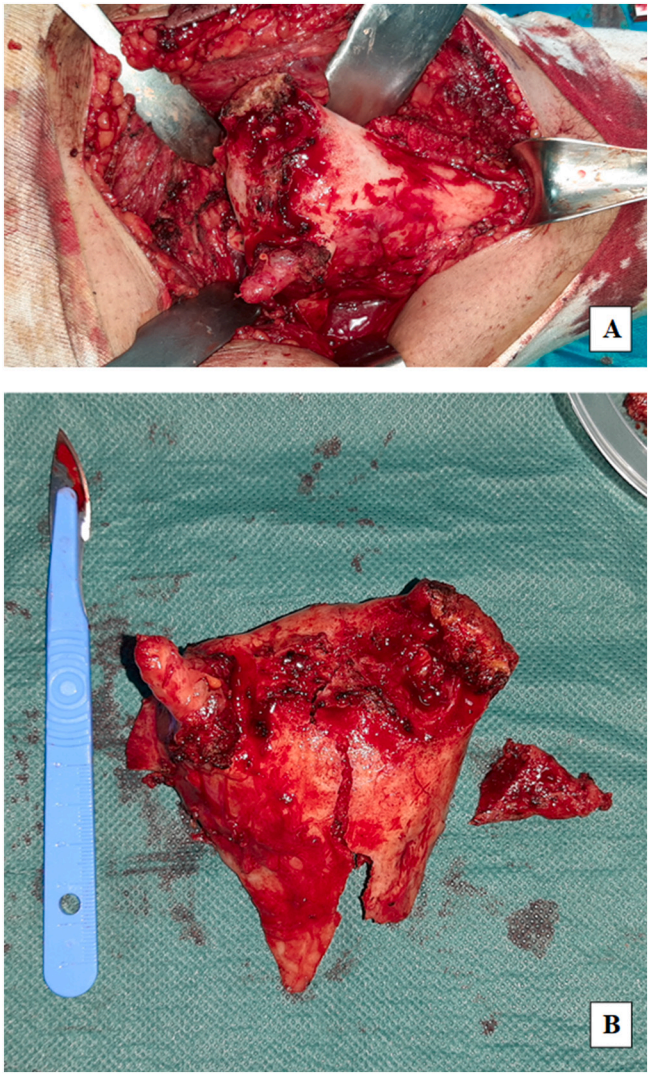


**Fig. 5.** A. The intraoperative aspect of the giant exostosis at the level of the femur before complete resection. B. The appearance of a devil's head-like giant exostosis following resection.

and confirmed by CT angiography, which is still the gold standard [6–8].

In most cases, X-rays and CT scans are diagnostic and allow for anatomic delineation of the lesion [9]. On imaging, osteochondroma typically appears as a pedicle or sessile bone-like protrusion. The cancellous and cortical bones are linked to the normal bone. The shadow of cartilage can be seen at the top of the tumor, with irregular calcification and/or ossification in the middle. In our case, CT and 3-D reconstruction clearly showed the size and boundary of the osteochondroma, as well as the degree of tumor compression to the superficial femoral artery, allowing us to design an operative plan.

The risk of malignant transformation of osteochondroma is less than 1 % [10]. Differential diagnosis from other neoplasms, such as chondrosarcoma, is required. Chondrosarcoma can develop as a primary or secondary tumor to osteochondroma [11].

Except in deep locations or near a vascular pedicle, resection of a solitary exostosis is usually straightforward [12]. Excision of an exostosis with a broad implantation base leaves a significant cortical defect, increasing the risk of fracture, particularly in the lower limb.

Depending on the defect, partial or total limb offloading is a safety measure. Exostosis recurrence is extremely rare. It occurs when cartilaginous cap fragments are left in place. This is why the excision has to be extra-periosteal. Furthermore, recurrence should raise concerns about malignant transformation [1,11]. To avoid serious and irreversible sequelae, surgical treatment of neurovascular complications of exostoses is strongly advised [4].

## 4. Conclusion

A solitary exostosis of the femoral diaphysis causing arterial compression is a rare complication. The standard treatment, which yields the best results, is a biopsy, removal of the exostosis, and relief of artery compression. It should be considered early, before the onset of serious vascular sequelae, which can be irreversible and lead to amputation. To avoid complications, better recognition and more comprehensive evaluation of these rare cases should be emphasized.

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## Ethical approval

N/A.

## Consent

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

## Author contribution

Omar Fadili: study concept, data collection, analysis and interpretation, writing the paper.

Mohamed Laffani: managed the analyses, and the correction of the manuscript.

Oussama El Adaoui: managed the analyses, and the correction of the manuscript.

Yassir El Andaloussi: managed the analyses, and the correction of the manuscript.

Ahmed Reda Haddoun: managed the analyses, and the correction of the manuscript.

Driss Bennouna: managed the analyses, and the correction of the manuscript.

## Registration of research studies

Not applicable.

## Guarantor

Omar Fadili.

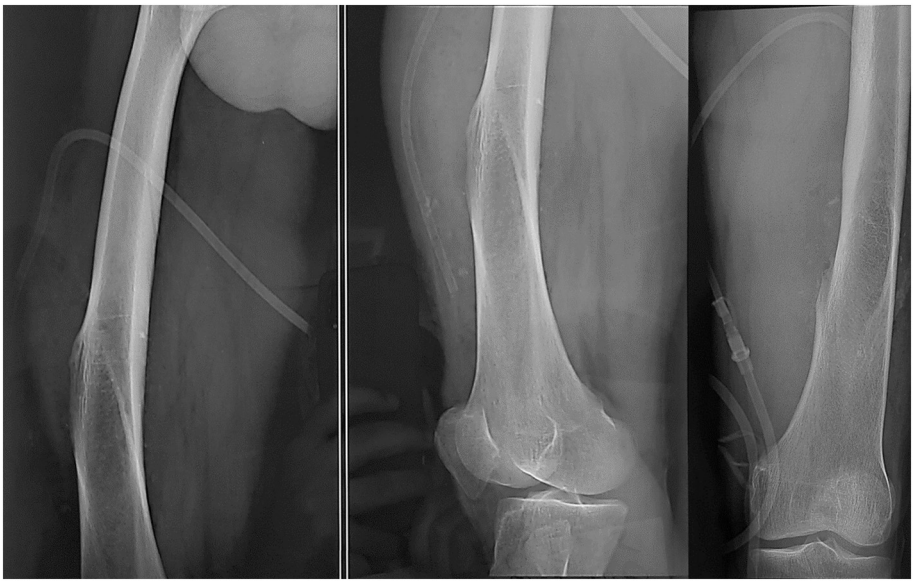

**Fig. 6.** Standard postoperative AP and lateral X-ray showing complete resection of the osteogenic exostosis.

## Declaration of competing interest

None.

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