

# A brachial artery pseudoaneurysm in the form of a “malignant tumour” as a complication of a proximal humerus exostosis

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## Abstract

**Background:** Osteochondromas are the most common form of benign bone tumour. They are mostly asymptomatic, but sometimes they can irritate surrounding structures, like vessels or nerves, and cause complications. More often they apply to the lower extremities, especially around the knee joint. Osteochondromas which cause complications in the upper extremities are much less common. This paper presents a rare case of a brachial artery pseudoaneurysm as a complication of a proximal humerus exostosis, which was initially described and treated as malignant tumour of the arm. A comprehensive review of the literature has also been carried out.

**Methods:** A 19-year-old male patient was admitted with acute pain of the left arm. Eight years earlier he had been diagnosed with hereditary multiple osteochondromas. Examination revealed a palpable, non-painful tumour of the axilla's area and the posterior part of the left arm. An magnetic resonance imaging (MRI) with contrast demonstrated three osteochondromas in the area of the proximal humerus. From the free end of one of these, a big, nodular structure was spreading. From the MRI, a chondrosarcoma was suspected. Samples were taken for histopathological examination. After the surgery, increased pulsations were observed around the operative area. A ultrasonography (USG) revealed a fibrotic pseudoaneurysm of the left brachial artery. Histopathological examination showed deposits of hemosiderin without any cancer cells. With the change in diagnosis, the next stage of treatment was planned – artery reconstruction using a saphenous vein graft and an osteochondroma excision.

**Results:** A few weeks after surgery, the patient recovered full function of the upper extremity and did not report any discomfort. A control USG showed proper flow through the brachial artery and venous graft.

**Conclusion:** In the case of a tumour in the area of an osteochondroma, caused by trauma, a pseudoaneurysm should be suspected. Diagnostic and therapeutic treatment must be properly planned, as unrecognised it could cause a severe, life-threatening haemorrhage during the operation.

**Keywords:** pseudoaneurysm, osteochondroma, malignant transformation, brachial artery, chondrosarcoma.

## Introduction

Osteochondromas, or osteocartilaginous exostoses, represent the most common form of benign bone tumour. They occur in 3% of the general population and represent 35% of all benign tumours and 8% of all bone tumours (1). In most cases (85%), they are single and take form of a solitary osteochondroma (SO) while in 15% of cases there are multiple lesions which are caused by hereditary multiple osteochondromas (HMO) (2–4). The median age of diagnosis is three years and the vast majority of affected individuals are diagnosed by the age of 12 (3)

with a significant male predominance (86%) (5). Osteochondromas appear in the growth plate area of long bones (e.g., femur, tibia, ulna, radius, humerus) or on the surface of flat bones (e.g., scapula, pelvis) (3, 6), but the most common place of occurrence is the distal part of the femur (around the knee) which concern approximately 70 – 86% of cases (3, 5). Generally, osteochondromas are symptomless and are found incidentally. The only clinical symptom is a painless, slow-growing mass on the affected bone. However, there may be various, significant complications, which occur in 4% of cases (7)

and these apply to both the skeletal system and the soft tissue surrounding the exostoses. They can mainly cause bone deformations, fractures or problems with the joint mechanism (2, 3). Sometimes they can irritate surrounding structures like blood vessels, nerves, skin or tendons and cause specific symptoms like numbness, weakness and other symptoms of nerve compression (2, 3, 8–13). Less often they can cause vascular compression, arterial/venous thrombosis, distal embolism, aneurysm, and pseudoaneurysm (2, 3, 5, 7, 13–27). They mainly affect the popliteal nerve and artery.

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**Figure 1:** Pre-operative MRI scan with contrast of the left arm, coronal slice. The tumour is marked.

When the tumour is located under the tendon it can irritate it and cause pain, difficulties in moving or, in extreme cases, to rupture it (2, 27–30). The most serious complication is a sarcomatous transformation to the chondrosarcoma or, less common, other sarcomas. This applies to 0.5–2% patients and can increase to 25% among patients with HMO (3, 31, 32). Factors that significantly increase the risk of malignant transformation are: increasing age, lesions located in the axial skeleton such as the pelvis, scapula, ribs, and spine or in the proximal aspect of the extremities as well as recurrent tumours and HMO (3, 4). Pain and/or an increase in the size of lesions are the most common clinical signs of transformation especially in a physically mature person. The treatment of choice for osteochondroma is surgical removal of the lesion. Furthermore, the risk of recurrence is observed in 5.8% of cases (2). We would like



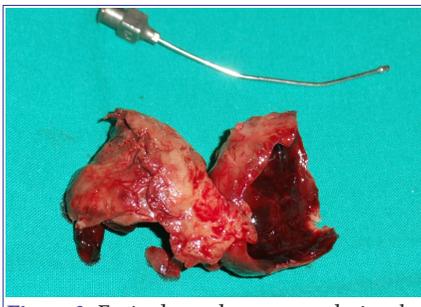
**Figure 2:** Pre-operative MRI scan with contrast of the left arm, horizontal slice. The tumour is marked.

to report a much rarer complication of proximal humerus osteochondroma, which is a brachial artery pseudoaneurysm. Only several cases reported this kind of location for this complication in the literature (13, 19, 22, 25–27, 33). Moreover, we would like to stress the fact that this lesion was not initially described as an artery pseudoaneurysm, as in the publications found, but as a malignant transformation to a chondrosarcoma.

**Material& methods**

A 19-year-old male patient was admitted with acute pain of the left arm. The pain appeared a week earlier during an arm swing and it was accompanied by bruising of the limb. Eight years earlier he had been diagnosed with HMO with three osteochondromas of the left humerus, which were gradually increasing but did not show any symptoms. Physical examination revealed a change in the contour and palpable tumour of the axilla area and the posterior part of the left arm, which coincided with localisation of exostoses on radiography. The patient reported only slightly local ailments. There was a full range of motion. Warmth, vascularity and sensations of the upper extremity were

normal too. An magnetic resonance imaging (MRI) with contrast was performed, and it demonstrated three osteochondromas, two in the posterior and one in the anteromedial surface of the proximal humerus. From the free end of one these, through continuity, a big nodosum structure was spreading with dimensions of 55x55x50mm, showed heterogeneous signal and irregular areas of contrast enhancement (Fig 1 - Pre-operative MRI with contrast, coronal slice; Fig 2 - Pre-operative MRI with contrast, horizontal slice). The radiologist suspected a chondrosarcoma. When the patient came back with the MRI result, he no longer reported any local symptoms and still performed a full range of motion. Examination revealed a movable, non-painful mass above one of the exostoses on the medial side of the arm. He was urgently admitted to the hospital in order to take samples for histopathological analysis. During the surgery, to avoid damage to the brachial artery, the operator used a stethoscope to determine the probable location of this blood vessel. When samples were taken it was found that the lesion didn't look typical for a chondrosarcoma. At the end of the biopsy no significant bleeding was observed. After the surgery, a feeling pulsation in the operative area was confirmed with palpation, which led to ultrasonography (USG) imaging and revealed a pseudoaneurysm of the left brachial artery. Meanwhile, the histopathological test result showed that the biopsied tumour didn't have any cancer cells, but only deposits of heamosiderin. It confirmed the USG diagnosis that, it was a fibrotic brachial artery pseudoaneurysm. The patient qualified for vascular-orthopaedic surgery. An computed tomography angiography (angio-CT) showed the pseudoaneurysm on the medial side of the proximal brachial artery. An operation was performed, where the



**Figure 3:** Excised pseudoaneurysm during the operation.



**Figure 4:** Saphenous vein before graft.



**Figure 5:** Grafted saphenous vein.



**Figure 6:** Post-operative X-ray of the left arm with small rest of unexcised osteochondroma; anterior-posterior projection.



**Figure 7:** Post-operative X-ray of the left arm with small rest of unexcised osteochondroma; lateral projection.

damaged part of the brachial artery was replaced by a graft from the saphenous vein, and the osteochondroma was excised (Fig 3 - Excised pseudoaneurysm; Fig 4 - Saphenous vein before graft; Fig 5 - Grafted saphenous vein). A few days after surgery, the patient didn't report any major complaints, the wound was healing properly and a control X-ray was performed (Fig 6 - Post-operative X-ray of the left arm, anterior-posterior projection; Fig 7 - Post-operative X-ray of the left arm, lateral projection).

### Results

After the operation, the patient recovered full function of the upper extremity and did not report any discomfort. The control USG showed proper flow through the brachial artery and venous graft.

### Discussion

Arterial pseudoaneurysms are a rare complication of osteochondromas and most publications on this topic are case reports (5, 34). The most common clinical presentation of this complication (51%) is painful swelling. The distal femur is the most frequent site and in 77% of all cases the popliteal artery is involved (5). The arm is an extremely rare site for the occurrence of this kind of complication and only a few cases of a brachial artery pseudoaneurysm are reported in the literature (13, 19, 22, 25–27, 33). The most frequent treatment is open surgery with osteochondroma excision and artery

reconstruction using a saphenous vein graft, because often an end-to-end anastomosis cannot be performed (5, 34). Our case is extraordinary, because the clinical presentation, risk factors and image explorations clearly indicated a malignant transformation. However, it turned out that the real problem was an unrecognised pseudoaneurysm of the brachial artery. There was a typical medical history of multiple osteochondromas with characteristic symptoms like swelling and pain in the arm in the area of the exostoses. Additionally, the patient presented several main risk factors for sarcoma transformation, like: a lesion located in the proximal end of the extremity, a history of HMO and mature age (2–4). According to our knowledge there was a bigger probability of a malignant transformation (3, 31, 32), then a much less common complication like an artery pseudoaneurysm, especially in such an extremely rare location of an upper limb, and more specifically, the proximal end of the humerus. Furthermore, according to the description by radiologists, the MRI scans showed a big nodosum structure with a heterogeneous signal and irregular areas of contrast enhancement, which continued from the free end of one of the exostoses. This further increased the probability of a chondrosarcoma. It's hard to say if there was a diagnostic mistake, or if the fibrotic brachial artery pseudoaneurysm took the form of a malignant tumour which in MRI scans with

contrast was similar to a chondrosarcoma. Another interesting aspect is the fact that the operator (an orthopaedist) showed extraordinary caution before the first incision and by listening to the operative area with a stethoscope he avoided the route of the brachial artery and did not cause life-threatening complications in the form of a massive arterial haemorrhage. In fact, the correct diagnosis was made only after an ultrasound examination, which confirmed a fibrotic brachial artery pseudoaneurysm. Probably the sharp end of one of the osteochondroma ruptured the brachial artery after trauma, caused by the arm swing. This case shows, that an artery pseudoaneurysm can imitate a malignant tumour in an MRI with contrast. Therefore, the standard differential diagnosis should include an artery pseudoaneurysm and, besides the necessary MRI with contrast examination, a USG should be considered, especially where there is a history of osteochondroma, recent history of trauma or pain, and also when exostoses are close to the region of the artery. After the surgical exostectomy it is also important to follow-up radiologically because the osteochondroma may spontaneously recur, even after adequate resection and to remember that this is one of the risk factors of a malignant transformation or other typical osteochondroma complications (3, 4).

### Clinical relevance

Artery pseudoaneurysms are a much less common complication of osteochondromas, especially if they are located in an upper extremity. In the case of a tumour in the area of an osteochondroma, particularly after a trauma, a pseudoaneurysm should be suspected and there must be properly planned diagnostic and therapeutic medical treatment. Therefore, besides the necessary MRI with contrast, where scans can give the false positive result of a sarcoma transformation, we recommend considering a cheap and easily available ultrasound examination, to complement the diagnostic process because an unrecognised artery pseudoaneurysm could be very dangerous during an operation, and could cause a severe, life-threatening haemorrhage.

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