Case Report

Pseudoaneurysm Complicating Proximal Humeral Exostosis: A Case Report of a Missed Diagnosis on Computed Tomography Angiographic Examination

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Abstract
Vascular complications of osteochondromas of the humerus are extremely rare. Only 9 cases with pseudoaneurysms of the brachial artery complicating osteochondromas of the humerus were reported in 7 English articles. The computed tomography (CT) angiographic examination of all of them were positive for the diagnosis of the pseudoaneurysm. Up to our knowledge, this is the first case report in which the CT angiography misdiagnosed such lesion. In this article, we described a case with a pseudoaneurysm complicating a proximal humeral exostosis in a 27-year-old male patient with a known history of hereditary multiple osteochondromas since childhood that was unanticipatedly misdiagnosed by the CT angiographic imaging modality. The participating authors tried to infer reasoned possibilities behind this unreported phenomenon before.
Background

Osteochondromas – both solitary and multiple – represent the most common bone tumor in general. It is considered as a developmental lesion rather than a true neoplasm [1]. It constitutes 20–50% of all benign and 10–15% of all bone tumors. Topographically, the humerus was affected in 10–20% of cases [2]. Hereditary multiple osteochondromas on the other hand is a variant with an autosomal dominant inheritance and is amenable for a higher incidence of complications as cosmetic issue, bursa formation, secondary deformities, fracture, articular dysfunction, and peripheral nerves compression. Malignant transformation and vascular injuries are 2 additional most serious reported complications [3]. Unfortunately, malignant transformation has been reported in 3–5% of these cases compared to only 1% in cases with solitary osteochondromas [3]. Vascular complications are considered the second most serious ones [4, 5]. On reviewing the literature, only 9 cases with pseudoaneurysm of the brachial artery complicating osteochondromas of the humerus were reported in 7 English articles. The computed tomography (CT) angiographic examination of all of them were positive for the diagnosis of the pseudoaneurysm [6–12]. Up to our knowledge, this is the first case report in which the CT angiography misdiagnosed such lesion at the proximal humerus.

Case Description

A male patient with a known history of hereditary multiple osteochondromas, presented lately at the age of 27 years old with a recent painful and increasing in size swelling on top of a long preexisted large upper medial left arm exostosis abutting his lateral chest wall on December 2020. He declared that this recent swelling was noticed only in the last 4 weeks. On clinical examination, by inspection, it was a large swelling on the upper medial aspect of the left arm reaching the axilla and measuring about 14 × 12 × 8 cm in size with a visible overlying skin bruising. It was neither expansile nor pulsatile (shown in Fig. 1a). On superficial palpation, it was moderately tender and cystic (positive fluctuation test), and it has the normal body temperature, and no thrill could be felt. On deep palpation, a hard bony preexisting exostosis was felt at the button of this cystic swelling measuring roughly 5 × 3 × 2.5 cm, corresponding with

Fig. 1. Surgical steps: (a) showing the swelling, position, and incision. b Skeletonization of the pseudoaneurysm. c, d Opening of the sac with removal of the thrombus. e Exploration of the brachial artery. f Hemostats, ligation, and cut of the medial circumflex humeral artery. g, h Multiple drill holing and marginal excision of the main large medial osteochondroma. i Shaving of the posteromedial smaller osteochondroma.
that seen on the plain X-ray (shown in Fig. 2). The limb distal neurovascular state was normal. Contrast-enhanced MRI study of the left arm was carried out to evaluate the nature of the swelling as well as its effect on the nearby structures (shown in Fig. 2a–g), followed by CT angiography (shown in Fig. 2h, i) which, except for the incidental discovery of a high bifurcation of the left brachial artery, did not detect any vascular affection. As high index of suspicion for a pseudoaneurysm was raised, the orthopedic surgeon reached out to the radiologist who recommended for a complementary Doppler ultrasound (U/S) examination modality that was overlooked before. Amazingly, it successfully picked up active minor vascular leakage (flow) while moving the limb in different positioning during this (U/S) examination. It also demonstrated a hemorrhagic fluid density. So, it confirmed the diagnosis of a pseudoaneurysm. Accordingly, the patient was urgently scheduled for a vascular-orthopedic surgery.

**Operative Technique**

Under general anesthesia, the affected arm was abducted on a side table. The vascular surgeon started his job first. A longitudinal incision was made on the left medial upper arm centered over the pseudoaneurysm. The subcutaneous tissues and deep fascia were then reflected off the pseudoaneurysm. The pseudoaneurysm sac was opened, and the thrombus removed (shown in Fig. 1). The brachial artery was exposed above and below the pseudoaneurysm and looped loosely by a nylon tape. It was found to be intact. The medial circumflex humeral artery was found to be the feeding artery to this pseudoaneurysm where it was ligated and cut near its ostium at the wall of the aneurysmal sac. The aneurysmal sac was
found to be a cavitation measuring about $14 \times 12 \times 8$ cm in size, and its wall was formed by fibrous tissues with a traceable outer dissecting plane, separating it from the surrounding soft tissues, and filled with thrombus. It was skeletonized and excised. Then, the orthopedic surgeon excised the indexed osteochondroma. The operation was completed by closure of the surgical wound in layers after accomplishing good hemostasis. Histopathologic examination was done and proved that the excised specimens were for a pseudoaneurysmal sac and a benign osteochondroma.

**Discussion**

Pseudoaneurysm is much less common in the upper limb [10, 13–15]. As the tumor cartilaginous cap ossifies at the end of growth, it becomes a sharp hard bone spike that on limb motion, abrades the adjacent vessel wall, producing a traumatic tear [8, 15, 16]. Raherinantenaina et al. [5] (2015) in their great review article analyzing 101 cases of arterial pseudoaneurysm complicating osteochondromas showed that only 5% were in the upper arm. Clinically, the most common presenting symptom in all was painful swelling and hematoma [8, 10]. Currently, CT angiography is the preferred diagnostic imaging modality for this complication [13, 15]; however, its failure in detecting this complication in lower limb lesions was reported in many articles [15, 17, 18]. Soto et al. [19] (2001) reported that its sensitivity was 95.1%, and the specificity was 98.7% in detecting pseudoaneurysm [19, 20]. Authors owed this to either thrombosis of the pseudoaneurysm’s lumen [15, 18]. Others related this to either technical mishap as a result of a limited possible anatomic coverage with a single acquisition when a single-detector row CT scanner was used [19, 20]. In this work, the contributed authors (orthopedist, vascular surgeon, and radiologist) inferred this to a possible occurrence of what is called “intermittent bleeding phenomenon” from the injured feeding vessel (medial circumflex humeral artery), where in certain limb positioning, its ostium gets opened and bleeds, while at another position, it became shut off. So, if CT angiography was done while the limb is fixed in a position shutting off the bleeding ostium, it will give false-negative images for a present pseudoaneurysm.

Doppler ultrasound on the other hand, has more advantages on CT angiography in enabling the U/S examination of the limb while moving it in a different direction [13–15, 18]. This gives a high chance to disclose a possible “intermittent bleeding phenomenon” that detects and confirms the diagnosis of a pseudoaneurysm.

Regarding contrast-enhanced MRI, it is the modality of choice in determining osteochondroma-related complications as malignant transformation and fractures or its effects on the nearby surrounding anatomic structures as reactive bursitis [9, 14]. Up to our knowledge, this is the first case reporting an atypical pseudoaneurysm complicating humeral osteochondroma that was misdiagnosed by the CT angiographic diagnostic modality.

**Conclusions**

In cases with suspected pseudoaneurysm complicating an existing osteochondromas, it is advised to follow a pragmatic ascending cascade of imaging workup that is doing the routine plane X-ray, followed by U/S examination and then CT angiography in order to avoid misdiagnosis. A teamwork approach, including an orthopedist, vascular surgeon, and radiologist, is needed for proper diagnosing, preoperative planning, and surgical treatment of these cases.
Statement of Ethics

The study was approved by the (General Organization of Teaching Hospitals and Institutes Research Ethical Committee on April 12, 2021) and was conducted in accordance with the Declaration of Helsinki and Ethical Guidelines for Epidemiological Research. Informed written consent was obtained from the patient and his guardians before participation in the study. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conflict of Interest Statement

The authors declare that they have no competing interests.

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Author Contributions

A.A.K. did orthopedic operative steps and is the surgeon who supervised writing and finalizing the article. A.S.G. did vascular operative steps. T.Y.G. contributed in radiographic diagnosis and image reporting. All the authors contributed to writing and editing the article.

Data Availability Statement

The data that support the findings of this study are not publicly available due to ethical restrictions. Queries regarding the data in this article should be addressed to the corresponding author (A.A.K.).

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