Diagnostic and Interventional Imaging (2016) xxx, xxx—xxx

PICTORIAL REVIEW / *Musculoskeletal imaging*

Imaging of benign complications of exostoses of the shoulder, pelvic girdles and appendicular skeleton

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KEYWORDS
Exostoses;
Complications;
Impingement

Abstract Exostoses are the most common benign bone tumors, accounting for 10 to 15% of all bone tumors. They develop at the bone surface by enchondral ossification and stop growing when skeletal maturity has been reached. At first, exostoses are covered by a smooth cartilage cap that progressively ossifies with skeleton maturity. Then they may regress, partly or even completely. Osteochondromas may be solitary or multiple, with the latter associated with hereditary multiple exostoses (HME). Exostoses develop during childhood and become symptomatic during the third decade of life in the case of solitary exostoses, or earlier, in case of HME. They stop growing after puberty, when the epiphyseal plates close. Most exostoses remain asymptomatic. Local complications, usually benign, may occur, such as fractures or mechanical impingements upon nearby structures. In rare cases, sarcomatous degeneration occurs. Most of these complications have been described in case reports. This article describes the imaging features of benign complications of exostoses of the shoulder, pelvic girdles and appendicular.

Exostoses are the most common benign bone tumors accounting for 10 to 15% of all bone tumors. They develop at the bone surface by enchondral ossification and stop growing after skeletal maturity has been reached. At first, exostoses are covered by a smooth cartilage cap that progressively ossifies with skeleton maturity. Then they may partly regress, even completely [1]. Osteochondromas may be solitary or multiple, with the latter associated with hereditary multiple exostoses (HME) [2], a rare hereditary autosomal dominant syndrome. Exostoses develop during childhood and become symptomatic during the third decade of life in the case of solitary exostoses, or earlier, in case of HME. They stop growing after puberty, when the epiphyseal plates close [1]. Most exostoses remain asymptomatic.

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http://dx.doi.org/10.1016/j.diii.2015.11.021
2211-5684/

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The goal of this review is to illustrate benign complications of exostoses of the shoulder, pelvic girdles and appendicular skeleton as observed on computed tomography (CT) and magnetic resonance imaging (MRI).

Imaging features of osteochondromas

Typical features of osteochondroma include cortical and medullary continuity, metaphyso-diaphyseal location, pedunculated or sessile base. Osteochondromas raise parallel to the long bones. MRI shows a hyaline cartilage cap [3]. After intravenous administration of gadolinium chelate, a thin rim of enhancement corresponding to the fibrovascular tissue that covers the cartilage can be observed.

Complications of exostoses

Fractures

Fractures are unusual complications of exostoses that result from trauma. They occur through the neck of a pedunculated exostosis, because the pedicle is the weakest part [1].

Radiographs often are adequate to show fractures. CT and MRI are useful in case of doubtful diagnosis (Fig. 1) [4]. Cases of spontaneous osteochondroma regression and even resolution have been reported [5].

Figure 1. Volume rendered reformations show a large pedunculated exostosis at the medial border of the inferior metaphysis of the femur, fractured at the base (arrow).

Osseous deformities

Osseous deformities are the most frequent complications that affect growth and cause local side effects such as misalignment and bone inclination. These abnormalities are almost exclusively encountered in patients with HME. In very young patients, undertubulation of bone in the metaphyseal region of the hip and knees may be the first sign seen at radiography. Radiographs reveal metaphyseal widening with an Erlenmeyer flask deformity in the distal tibia.

Osseous deformities are often located in the forearm or wrist (30 to 60% of HME). They include: disproportionate ulnar shortening, ulnar deviation of the wrist, deformed radial articular surface with distal radio-ulnar joint disruption or dislocation of the radial head. Other parts may be affected: coxa valga, genu valgum, ankle valgus by talocrural disjunction, leg length discrepancy and short stature [2]. These deformities result in asymmetric growth, functional impairment and unsightly deformities (Fig. 2).

When exostosis develops in contact with another bone surface, such as in the interosseous space between the tibia and the fibula, deformities of the adjacent bone may be observed, resulting in erosion or scalloping of the bone surface (Fig. 3). Surgery aims to prevent (or limit) and correct deformities [6].

Impingement

On joints, exostosis may cause impingement and repeated friction during movement. Mechanical effects of impingement are limited range of motion, friction and trigger tendons or ligaments and early osteoarthritis. The scapula is involved in 3.0 to 4.6% of complications [7,8]. Usually located at the anterior surface of the scapula, they are often symptomatic. Two consequences may follow. One is the impingement upon the posterior thoracic wall, with or without bursitis, resulting in a snapping scapula with typical crackling when the scapulothoracic joint moves. The other is a posteriorly displaced scapula with limited range of motion. These positional disorders generally imply a winged scapula. Positional disorders mainly concern young patients and are usually painless [7,9].

Radiography, unlike CT or MRI, cannot always identify osteochondromas. MRI can also detect bursae. Dynamic sequences help detect positional disorders [10] (Fig. 4). Several cases of ischiofemoral impingement caused by exostosis have been reported. The syndrome results from impingement upon the quadratus femoris muscle in a small space located between ischial tuberosity and lesser trochanter. The most common radiographic features are edema and hemorrhage in the quadratus femoris muscle or at its myotendinous junction (Fig. 5). Bilateral ischiofemoral impingement is frequent in case of HME [11].

Bursa formation

Bursal formation overlying exostosis has a prevalence of approximately 1.5% [12]. The main areas affected are scapulothoracic joint, hip and shoulder. Clinically, a palpable lump is observed near an osteochondroma that may occasionally grow rapidly. Often painful, bursae may simulate malignant

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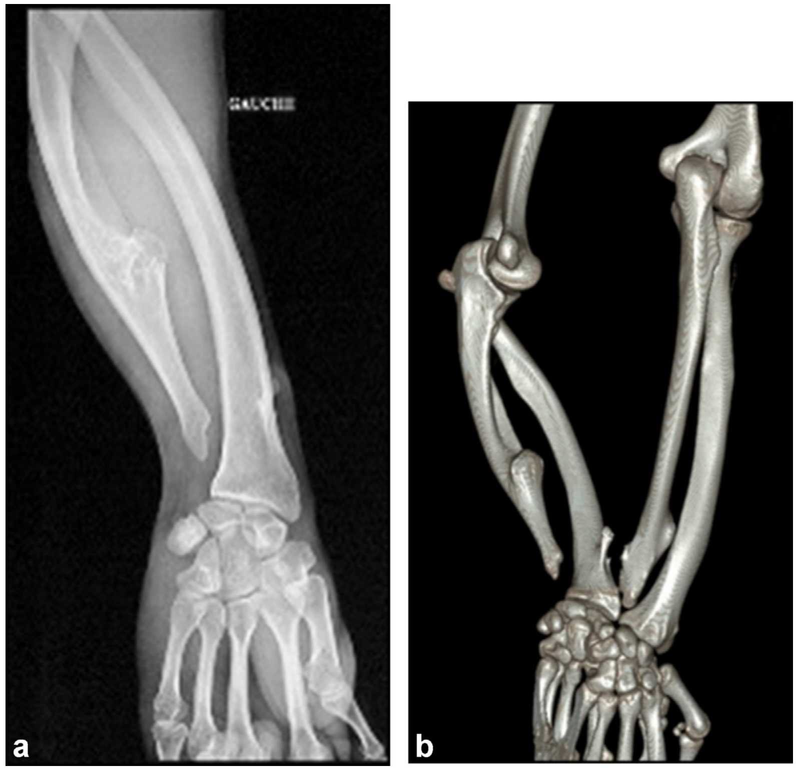

Figure 2. 21-year-old patient with hereditary multiple exostoses. a: standard radiograph shows deviation of the two bones of the forearm with right ulnar shortening and dysplasia; b: 3D reformatted CT image confirms the diagnosis.

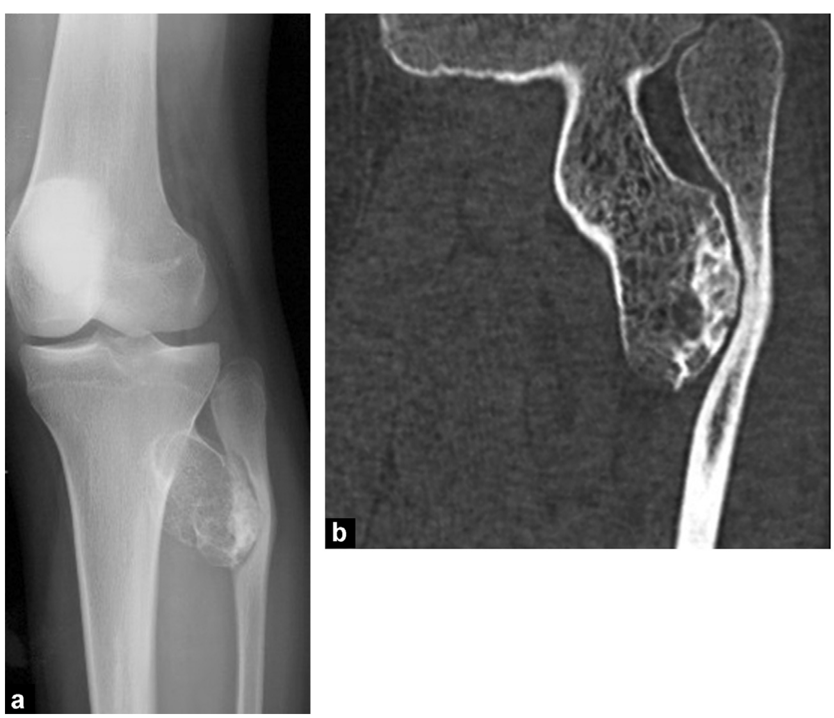

Figure 3. 20-year old woman presenting with pain in the left leg. a: radiograph shows a pedunculated tibial exostosis causing fibular deformity; b: CT images in the coronal plane using bone window show fibula deformity.

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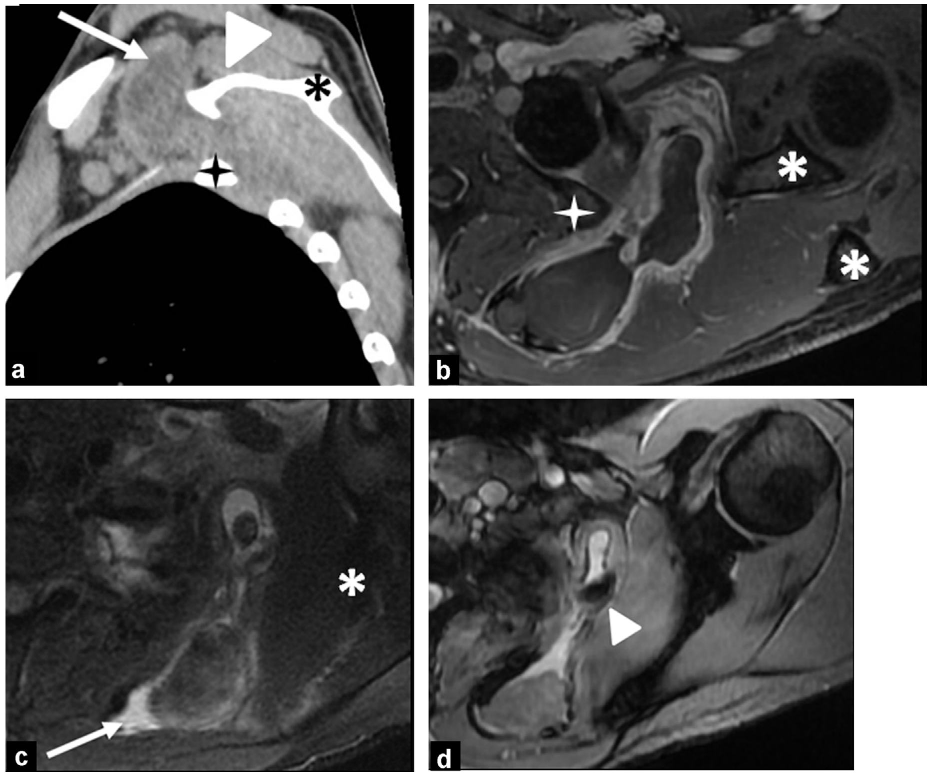

Figure 4. 23-year-old man presenting with pain in the left shoulder, reduced motion and cracklings. a: CT image in the sagittal plane shows a large exostosis (arrowhead) on the anterior and supero-medial aspect of the scapula (star) causing impingement upon the posterior arch of the 2nd rib (cross) and large bursitis (arrow) in the interscapulo-thoracic area; b: T2-weighted MR image in the transverse plane reveals a liquid-filled mass (arrow) in front of the sub scapular muscle and scapula (star), near the secondrib (cross); c: MR image obtained after intravenous administration of a gadolinium chelate shows hyperintense synovial (arrow); d: T2*-weighted MR image in the transverse plane shows hypointense clots (arrowhead).

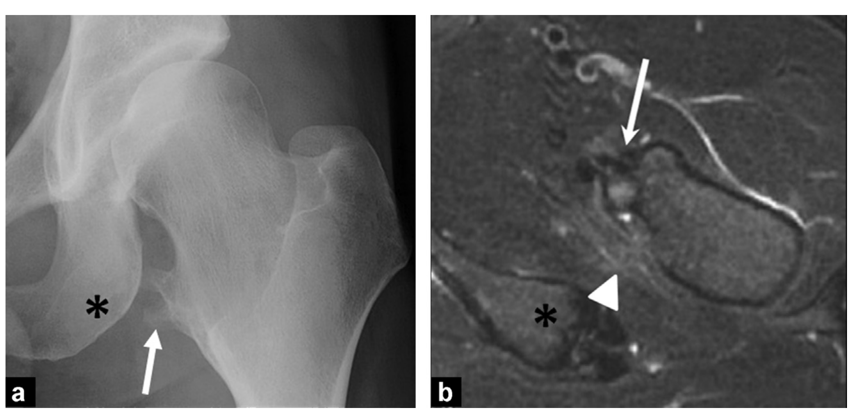

Figure 5. 30-year-old man, with atypical left inguinal pain. a: radiograph shows exostosis (arrow) of the medial aspect of the left femoral neck associated with widening (coxa valga); b: T2-weighted MR image in the transverse plane shows bursitis surrounding the exostosis (arrow) and impingement upon the external obturator and quadratus femoris muscles (arrowhead) between the exostosis and the ischium (star).

chondrosarcoma transformation. The bursae may become inflamed, infected or even hemorrhagic [5], and contain small cartilage fragments. Histopathological analysis shows fibrous connective tissue, partially bordered by a vascularized synovial membrane.

Newly formed bursae overlying an osteochondroma are not common, and occur on large osteochondromas. Most cases of large bursae involve the scapulothoracic joint, because of the almost constant friction between the scapula and the posterior arch of the ribs [13,14].

Imaging is useful for the diagnosis as well as for the differential diagnosis with sarcomatous transformation. Radiographs show a soft-tissue mass overlying the exostosis. CT and MRI show a fluid-filled mass with synovial lining, in contact with the cartilaginous cap of the exostosis. There may be hemorrhage (Figs. 4 and 6) [3].

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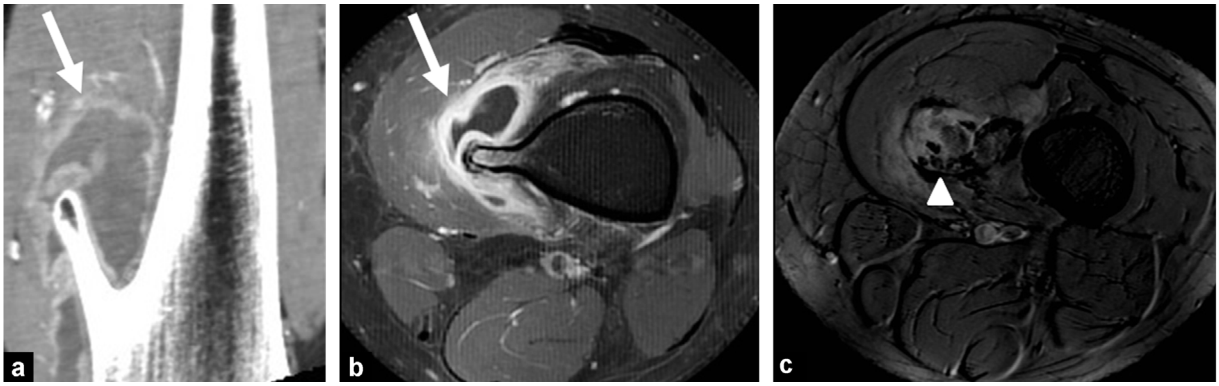

Figure 6. 22-year-old man complaining of swelling at the internal aspect of the left lower third thigh. a: CT image in the coronal plane obtained after intravenous administration of iodinated contrast material shows pedunculated exostosis at the medial border of the femoral distal diaphysis, ascending in the vastus medialis with large collection inside the vastus medialis covering the exostosis, many foreign bodies; b: T1-weighted fat-suppressed MR image in the transverse plane obtained after intravenous administration of gadolinium chelate shows enhancing bursal wall; c: T2-weighted MR image in the transverse plane shows hemorrhage in the collection, with many foreign bodies (arrowhead).

Bursae may contain fibrin or cartilage bodies, which may cause synovial chondrometaplasia leading to secondary synovial chondromatosis. This is extremely rare and is suspicious for malignant degeneration; calcifications appear in soft tissue in contact with the exostosis, accompanied by pain [15].

Vascular complications

Vascular complications are unusual. Arterial complications represent 90% of vascular complications. Four types can be observed including, vessel displacement, stenosis or thrombosis, occlusion and pseudoaneurysm formation. Venous complications include thrombosis and compression.

Some trauma or intense physical exercise during the previous weeks has been identified in about 35% of the patients [16]. Therefore, it is believed that some physical exercises may cause an adventitial defect and promote pseudoaneurysms.

Displacement — compression — stenosis and occlusion

Displacement is frequent, but asymptomatic. Its frequency correlates with size (Fig. 7). In patients with HME, stenosis is observed if osteochondromas are large and more particularly in the arteries in the legs. There are no hemodynamic consequences in young patients, due to the high arterial blood supply. Occlusion is exceptional.

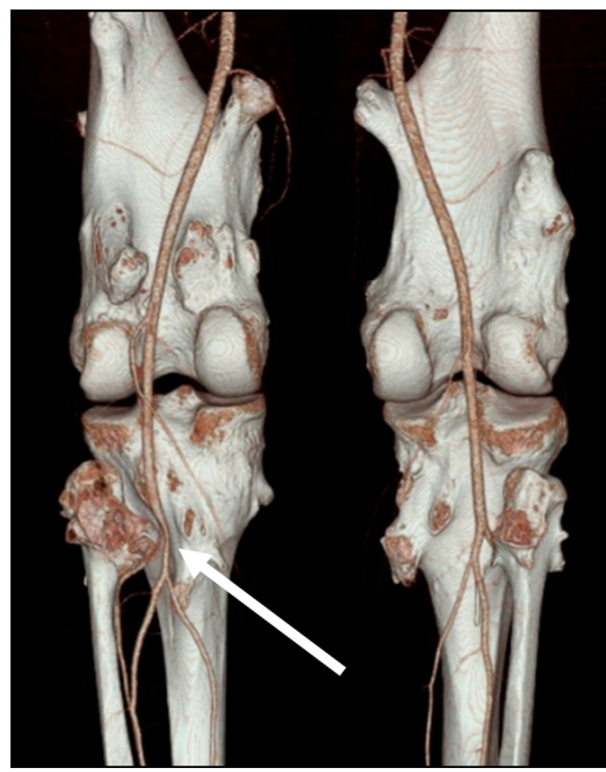

Figure 7. 21-year-old patient with hereditary multiple exostose syndrome. 3D volume rendered CT image shows sessile exostoses that project into the anterior tibiofibular interval and compress the left tibial-fibular trunk (arrow).

Arterial pseudoaneurysm

The most frequent vascular complication is pseudoaneurysm (64%) [17]. Popliteal artery is frequently involved, because it is fixed in the adductor canal, posteriorly at the aponeurotic hiatus in the adductor magnus, anteroexternally by the vastus medialis, internally by a fibrous layer and above by the femoral branch of the internal saphenous nerve, thus making it difficult for the artery to move. The involved exostosis is located on the distal metaphysis of the femur. Other arteries, such as the superficial femoral artery and the brachial artery, may be affected because of their location near the bones and the reduced mobility [18]. Most pseudoaneurysms are associated with sessile exostoses that are more likely to engage in repeated friction compared to pedunculated exostoses [2].

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Because of the pressure, vessel walls start to get eroded. Two hypotheses have been proposed to explain the mechanism. The cartilaginous cap being very thin or nonexistent could form a bony spike that causes repetitive strain injuries resulting in erosion of the vessel wall [19]. The loss of cartilage cap may also result from pressure necrosis of exostosis induced by enlarging pseudoaneurysms [20]. This complication occurs in young patients (average age is 23 years) when the cartilage cap [16] ossifies.

A pulsating (39%) or nonpulsating (24%) soft-tissue mass is observed, often associated with pain. There may be paresthesia, claudication, and even acute ischemia. Three cases of swollen soft popliteal tissues and distal acute ischemia that result from peripheral embolism secondary to pseudoaneurysm have been described [16].

Currently, angiography is essentially used for treatment. Pseudoaneurysm is confirmed by Doppler ultrasound, CT angiography or dynamic MRI. The images show soft-tissue swelling. Doppler ultrasound shows the narrow collar and the turbulent blood flow [21]. On CT, pseudoaneurysm is analyzed after intravenous administration of a gadolinium chelate, to detect vessel abnormalities. MRI shows an ''onion-like lamellar structure'' due to peripheral hemosiderin deposits and thrombi. In case of pseudoaneurysm, MRI shows typical pulsation artifacts [2].

Development of pseudoaneurysm paradoxically can lead to osteochondroma regression, even complete resorption. The mechanism is believed to be the growth arrest by erosion secondary to the pressure of the pseudoaneurysm. Active resorption and metaphyseal remodeling follow [22]. Ruptures are extremely rare, occurring in young male patients, following trauma [23—25].

Several cases of hemothorax secondary to rib exostosis have been reported: by erosion of a particularly cardiophrenic artery, by direct lung or pericardium (haemopericardium) injury. CT displays the exostosis, hemothorax, its cause and its impact [26,27].

Venous complications

Venous complications include thrombosis and extrinsic compression. Several cases of venous thrombosis secondary to compression by pseudoaneurysm have been described [21,28,29]. All the cases observed involved the popliteal vein following trauma (Fig. 8). O'Brien et al. reported an exceptional case of subclavian vein thrombosis at the level of the thoracic outlet secondary to first rib osteochondroma [30].

Peripheral nerves compression

Peripheral nerves are rarely compressed which represents less than 1% of symptomatic lesions. Nerve entrapment occurs in areas close to the bony structures [31]. The common peroneal nerve is the most frequently injured nerve. The site of compression is adjacent to the fibular head. This compression is evidenced by pain or strength loss. Longstanding compression results in muscular atrophy in the area innervated by the affected branch [32] and is accompanied by fatty involution.

Cases of sciatic nerve compression due to exostosis on the posterior aspect of the femoral neck have been reported [33,34]. This sciatic nerve entrapment must be differentiated from disco-radicular impingement. The nerve shows vasogenic edema due to increased capillary permeability and increased endoneural pressure [35]. Demyelinating neuropathy follows [36]. Electromyography confirms peripheral nerve involvement.

MRI shows the relationship of the exostosis to the nerves. Transverse sections are required to visualize the nerve and compression site [37]. Hyperattenuating areas are observed [35,36]. MRI shows post-denervation muscle lesions: first, muscle edema then atrophy and fatty involution (Fig. 9). Although MRI is the ideal examination modality, ultrasound is useful to explore dynamically long segments of nerve trunks. In case of inflammation, the examination shows a diffuse or segmental thickening, decreased echogenicity and loss of parallelism [38].

Follow-up

Patients with HME require regular clinical and radiological examinations to monitor the progression of skeletal deformities and detect complications, especially because of the risk of chondrosarcoma. Radiographs are adequate to evidence deformities but MRI is required to confirm sarcomatous transformation. Solitary lesions do not require routine monitoring [5].

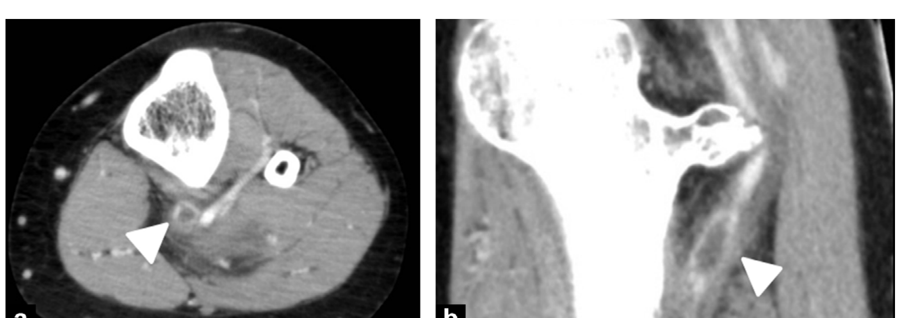

Figure 8. CT image obtained in the transverse (a) and the sagittal (b) planes during the venous phase after intravenous administration of iodinated contrast material shows a thrombosed popliteal vein compressed by a large exostosis at the lateral face of the tibia (arrowhead).

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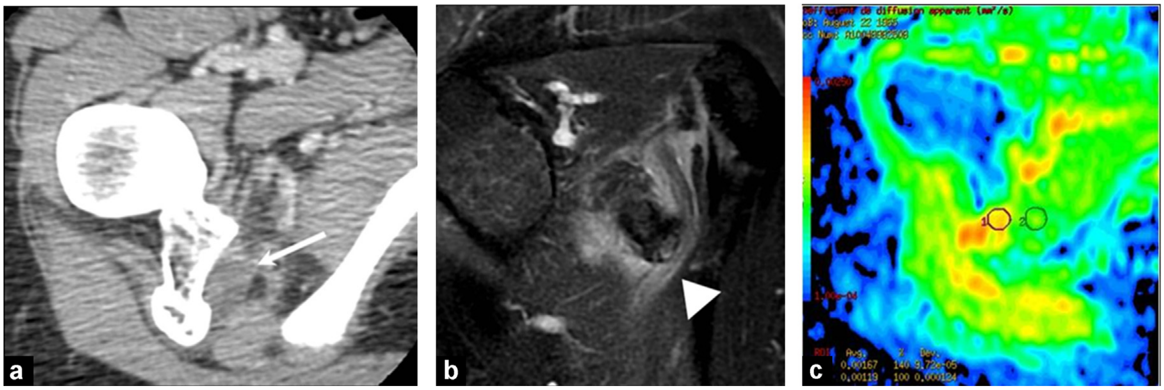

Figure 9. Exostosis at the femoral neck indicated by a mass on the sciatic nerve displaced on the medial aspect of the exostosis. a: CT image in the transverse plane shows injured sciatic nerve after intravenous administration of iodinated contrast material; b: MR image in sagittal plane (arrowhead); c: increased value of apparent diffusion coefficient on diffusion weighted MR image indicates vasogenic edema.

Conclusion

Many complications are associated with osteochondroma, such as skeletal deformities, fractures, dynamic mechanical complications related to friction (bursitis) or joint disorders, vascular compromise, peripheral nerve entrapments and central nerve compression. These complications are more frequent in patients with HME than in patients with solitary exostosis. Imaging helps characterize and identify underlying causes and confirms the diagnosis.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at http://dx.doi/org/10.1016/j.diii.2015.11.021.

Disclosure of interest

The authors declare that they have no competing interest.

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