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Intra-articular exostosis on the glenoid neck in a patient with hereditary multiple exostoses: a case report

Laurent Nové-Josserand, MD[a,*], Gaia Maccario, MD[b], Francesca G.M. Forni, MD[b]

[a]Shoulder Unit, Ramsay Générale de Santé, Jean Mermoz Private Hospital, Centre Orthopédique Santy, Lyon, France
[b]Università degli Studi di Pavia, Ospedale Policlinico San Matteo, Pavia, Italy

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Hereditary multiple exostoses (HME) is an autosomal dominant hereditary condition, caused in almost all cases (94%) by mutation in 1 of 2 genes: exostosin-1 and exostosin-2.[8] The prevalence of HME does not vary between men and women, but men with exostosin-1 mutations tend to have more severe disease.[12]

HME is characterized by the presence of multiple benign cartilage-capped bony outgrowths, termed exostoses or osteochondromas,[7] which can be broad-based (sessile) or pedunculated and form most commonly in the growth plate of long bones.[1]

The severity of the disease varies widely and depends on the number, size, shape and location of the exostoses.[8] The average number of exostoses per patient is 15-18 but some have more than 250.[1]

Isolated exostoses are usually asymptomatic and found incidentally on radiological examination.[7] The knee is the most common site for osteochondromas (40%), and the most frequently affected bones are the femur (30%), humerus (10%-20%), the bones of the hands and feet (10%), the pelvis (5%), the scapula (4%) and the spine (2%).[7]

The most common complaints in adults with HME are pain and functional impairment. Pain is often caused by compression of tendons and muscles (sometimes leading to chronic irritation and/or tears), nerve compression, bursitis above the cap of outgrowth, and joint misalignment. Traumatic fracture of pedunculated exostoses is also possible, but rarer.[8] The fact that exostoses are usually located in the vicinity of joints means that they often limit function and range of motion.[4]

Individuals with HME may have reduced skeletal growth, bone deformities, bone bowing, scoliosis, early osteoarthritis, and subluxation or dislocation of adjacent bones and joints.[1,5] Malignant transformation into chondrosarcoma occurs in about 4% of HME patients.[1]

Shoulder exostoses are associated with more severe disease and surgical removal is more common, particularly for exostoses of the scapula. We present the case of a 50-year-old male HME patient with progressive left shoulder pain due to an isolated exostosis on the anterior edge of the glenoid cavity, a very rare exostosis site.

Case report

A 42-year-old male patient with HME diagnosed since adolescence presented in 2015 to the orthopedic clinic for left shoulder pain which had been progressing for 4 years. He had already received 2 ultrasound-guided intra-articular corticosteroid injections in the shoulder, 1 in the acromioclavicular joint and 1 in the glenohumeral joint and had undergone surgery 15 years previously to remove a cervical intracanal exostosis and several times between the ages of 20 and 30 to remove painful exostoses around both knees. The shoulder was painful during daily activities but not at night. Clinical examination revealed full symmetric range of motion and normal rotator cuff function. X-ray imaging showed mid-sized exostoses in the metaphysis of the humerus (Fig. 1), but the patient did not feel any discomfort at this level.

Magnetic resonance imaging (MRI) findings were unremarkable apart from moderate bursal effusion. Scintigraphy findings were unremarkable. A working diagnosis of subacromial impingement was reached and the pain was successfully treated with additional corticosteroid infiltrations.

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*Corresponding author: Laurent Nové-Josserand, MD, Centre Orthopédique Santy, 24 Avenue Paul Santy, Lyon, 69008, France.
E-mail address: lnovejosserand.md@orthosanty.fr (L. Nové-Josserand).

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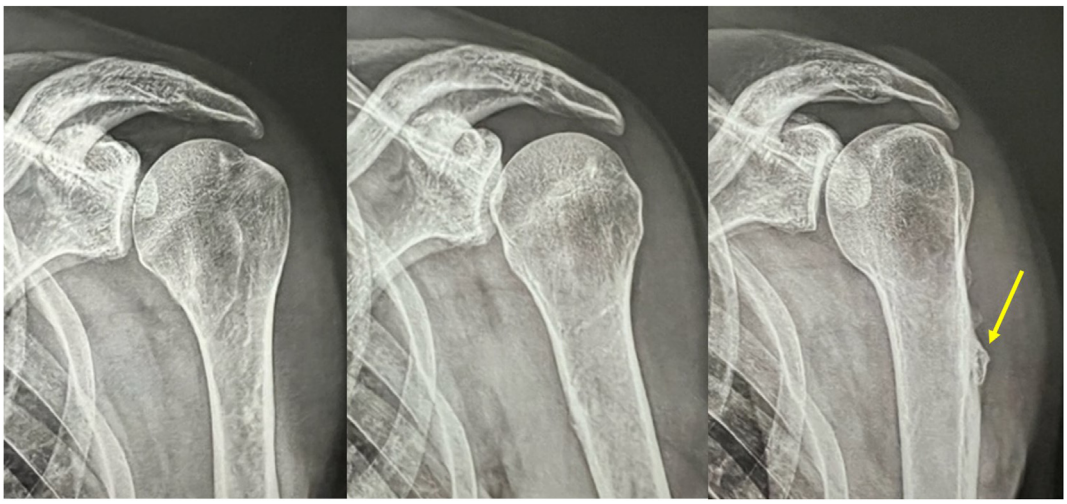

Figure 1 Antero-posterior radiographs of the left shoulder in neutral, internal, and external rotation. There are no visible abnormalities in the glenohumeral joint or the scapula. An exostosis (⟵) is visible at the level of the deltoid tuberosity in external rotation.

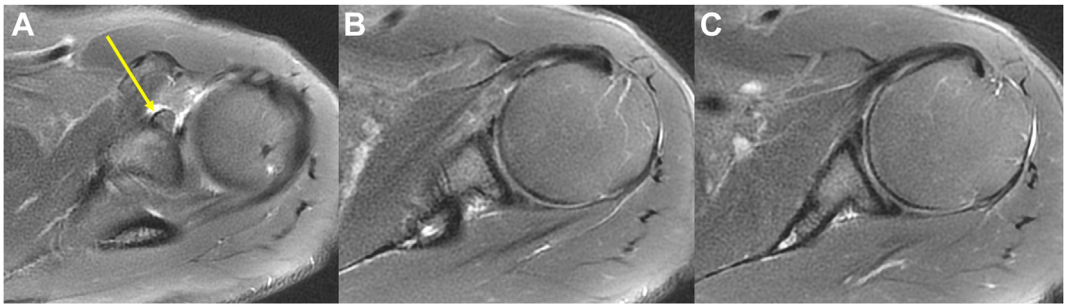

Figure 2 Magnetic resonance images of the left shoulder. (**A**) Transverse plane at the foot of the coracoid process. The exostosis was not observed at the time but is visible (⟵) in the anterior part of the glenoid neck, oriented toward the subscapularis tendon. (The subscapularis tendon itself is not visible on this plane.) Mild effusion can be seen in the anterior part of the joint. (**B**, **C**) Transverse planes in the middle and lower parts of the joint showing an apparently normal subscapularis tendon with an intact enthesis on the lesser tuberosity. The exostosis is no longer visible. There is no joint effusion. The appearance of the subscapularis muscle is also normal, with no fatty infiltration.

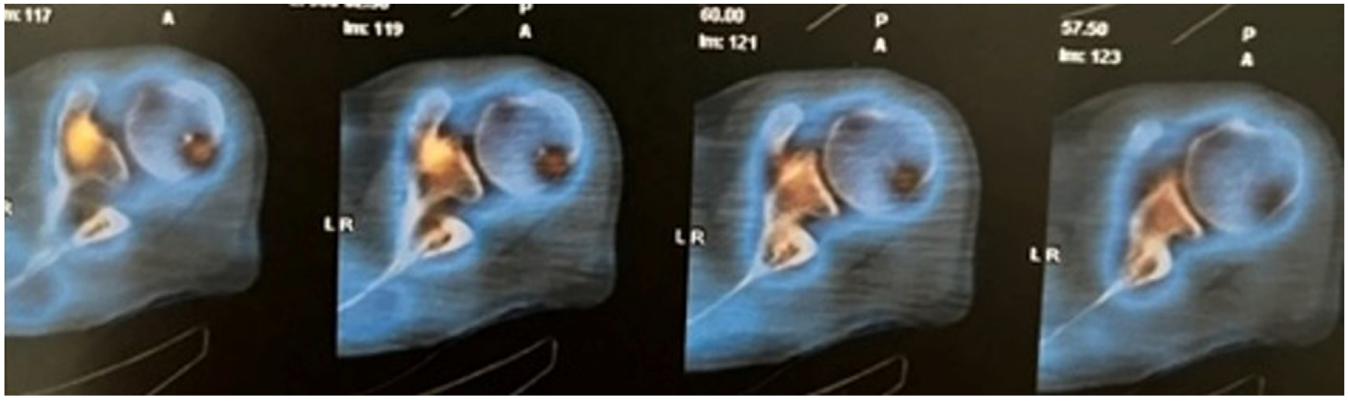

Figure 3 Transverse planes of a bone scan of the left shoulder showing hyperfixation at the base of the coracoid process rather than in the anterior part of the glenoid neck. The exostosis is not visible in these images.

The patient returned in January 2023 because the left shoulder pain had become permanent, varying with activity levels but overall more intense and preventing him from sleeping on his left side. The patient located the pain in the anterior central part of the shoulder at the level of the deltoid muscle. There was no pain at the level of the humeral exostosis either at rest, in movement or on palpation. Clinical examination showed full symmetric range of motion. Rotator cuff test results were normal but the belly-press test triggered pain. A small exostosis in the anterosuperior part of glenoid neck, facing the superior part of the subscapularis tendon, was observed on computed tomography scanner arthrography and in 2 sets of magnetic resonance images recorded in 2021 and 2022 (Fig. 2). Moderate bursal effusion was observed. There was no evidence of a subscapularis

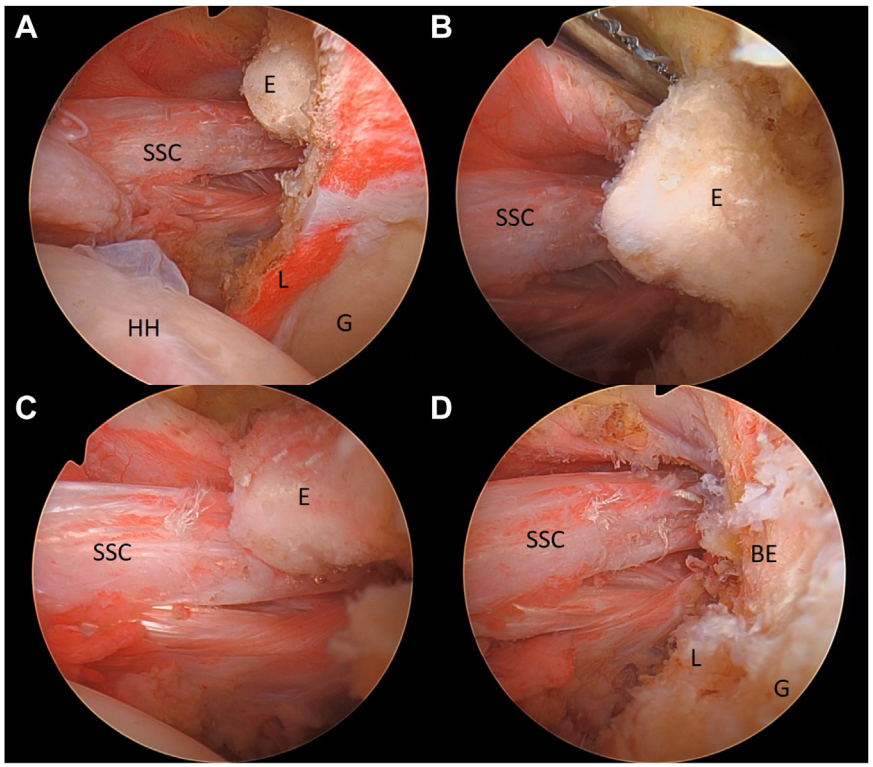

Figure 4 Arthroscopic views of the anterior part of the left shoulder joint through the posterior portal. (**A**) The E is visible in the anterosuperior part of the otherwise normal-looking L, and stands out because of its unusual location and shape. The SSC can be seen in front of the E, in its anatomical position but with a tear in its articular layer. (**B**) After dissection, the E with a broad base and no cartilage cap can be seen protruding from the anterior glenoid neck toward the SSC. (**C**) The E can be seen to impinge upon the superior part of the SSC. The delamination tear of the tendon at the musculotendinous junction can be explained by repeated friction during shoulder rotation. (**D**) Resection of the E with a burr (BE) has resolved the impingement of the SSC, which is not split in the transverse direction but delaminated longitudinally. The middle and inferior parts of the anterior L are also visible. *E*, exostosis; *G*, glenoid; *L*, labrum; *SSC*, subscapularis tendon.

tendon lesion and the insertion of the tendon on the lesser tuberosity was intact. The other rotator cuff tendons were intact and the corresponding muscles were evaluated as normal, including the subscapularis. Scintigraphy showed hyperfixation at the base of the coracoid process unrelated to the exostosis itself, as well as in the inferior part of the glenohumeral joint (Fig. 3).

Diagnostic shoulder arthroscopy was proposed to further investigate these atypical clinical findings. This was performed through the posterior portal with the patient in the beach chair position. The exostosis was rapidly located in the anterosuperior part of the glenoid neck, in front of a normal-looking anterior labrum. The exostosis was broad-based, without a cartilage cap, and was oriented perpendicular to the glenoid cavity in the direction of the superior part of the subscapularis tendon (Fig. 4, *A-C*), which had an atypical delamination tear of its articular layer leaving most of the tendon thickness and the humeral enthesis intact. No further glenohumeral or bursal lesions were observed on exploration of the joint.

The surgical procedure consisted in removing the entire exostosis with a burr down to its base on the anterior glenoid neck via an anterolateral portal in the bicipital groove (Fig. 4, *D*). The anterior labrum was left intact and in place. The subscapularis tendon was débrided with a shaver to stimulate healing of the ruptured fibers without tendon reinsertion or direct repair. The bursal effusion was treated by routine acromioplasty.

The patient was instructed to wear a 20°-abduction shoulder brace for 4 weeks and passive rehabilitation with hydrotherapy was started on day 6 after surgery. The patient was able to resume activities of daily living without pain 3 months after surgery with full and pain-free shoulder function.

Discussion

To our knowledge, this is the first reported case of intra-articular exostosis on the glenoid side of shoulder joint. Exostoses of the proximal humerus are common but are almost always extra-articular. Subacromial impingement due to an exostosis projecting into the subacromial space has been reported previously,[3] but the exostosis in this case was not strictly intra-articular. Intra-articular exostosis in the shoulder has also been described[9,11] but in these 2 cases, the exostosis was on the posteroinferior side of the anatomical neck of the humerus leading to pain and reduced range of motion, successfully treated in both cases by surgical excision.

Exostoses are also known to form less frequently, but classically, on the scapula, where they can lead to particularly painful snapping scapula syndrome if the exostosis is located on the ventral side, protruding up to the rib cage.[2]

The other notable feature of this case lies in the fact that the exostosis was directly responsible for an unusual subscapularis tendon lesion. The presence of the exostosis protruding perpendicular to the anterior neck of the glenoid led to direct mechanical wear of the subscapularis tendon and eventually to the described lesion. Usually, chronic and traumatic subscapularis tendon lesions involve detachment of the enthesis from the lesser tuberosity.[10] In this case, the impingement lead to a partial lesion of the articular surface of the subscapularis tendon at the level of the musculotendinous junction rather than at the enthesis.

The unusual nature and cause of the injury explains the diagnostic delay in this case. The fact that subscapularis tendon tests showed no weakness but were painful pointed toward a possible anterior rotator cuff tear. However, imaging findings were inconclusive because the enthesis of the subscapularis tendon was intact and because the tear only affected the articular layer of the tendon. Although the exostosis on the anterior glenoid neck was clearly visible on arthrography and MRI, possible impingement into the subscapularis tendon was not considered because of the apparent absence of tendon injury. The absence of close structures to impinge onto in the vicinity of the exostosis explain why it remained "asymptomatic" and undiagnosed for such a long time. While considered together, the clinical context (patient age, and level of physical activity), the fact that the pain increased during physical activity, the temporary effectiveness of a corticosteroid infiltration, the pain triggered by belly-press tests, and the scintigraphy findings pointed toward anterior subacromial impingement, diagnostic arthroscopy was required to reach a firm diagnosis.[6] The impingement was resolved by simply resecting the exostosis. The exostosis had no cartilage cap, probably because of chronic impingement with the subscapularis tendon, but the diagnosis was confirmed by the pathognomonic continuity of marrow and cortical bone between the exostosis and the parent bone.[9,11]

Conclusion

The noteworthy features of this case are the previously unreported location of the exostosis on the glenoid side of the shoulder joint and the unusual subscapularis tendon lesion it led to. This case highlights the value of diagnostic arthroscopy in patients with intractable joint pain and uninformative MRI and scintigraphy findings.

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