Idiopathic heterotopic ossification of bilateral subscapularis tendons: illustration of a rare entity and a concise literature review

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Abstract: Ossification of the subscapularis tendon is an extremely uncommon, poorly described lesion with little known about its etiopathogenesis and clinical significance. To the best of our knowledge, only three cases of this entity have been reported till now, which were all unilateral. The authors present first case of ossification of bilateral subscapularis tendons in a 57-year-old male and hope that with increase in the number of reported cases, proper guidelines for management of such cases can be formulated.

Keywords: Heterotopic ossification; subscapularis tendon; subcoracoid impingement

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Introduction

Ossification of the subscapularis tendon is a rarer lesion compared to calcific tendonitis. Although heterotopic ossification of shoulder has been noted in cases of traumatic brain injury, spinal cord injury, local trauma and surgery, it’s quite uncommon in subscapularis tendon with little known about its exact mechanism of formation. Some authors have mentioned of the subscapularis tendon ossification as a rare cause of anterior shoulder pain, subcoracoid impingement and subscapularis tear (1-3). Overall, three cases of ossification of unilateral subscapularis tendon have been documented (1-3). Calcific tendonitis of subscapularis tendon, although rare, is a relatively commoner entity (4-13). The authors present first case of ossification of bilateral subscapularis tendons in a 57-year-old male.

Case presentation

A 57-year-old male patient consulted a peripheral hospital with history of fall over the right shoulder. There were complaints of pain and restriction of movements (adduction, internal rotation and forward flexion) in the right shoulder. Patient apparently had no symptoms in both shoulders prior to onset of trauma. Patient was advised CT of the right shoulder (as that was the only modality available in that hospital) and the scan was sent to our institute for reporting via teleradiology. There was evidence of multiple foci of ossification in right subscapularis tendon along with the soft tissue swelling (Figure 1). Author advised MRI of right shoulder for evaluating extent of injury and to confirm ossifications in the subscapularis tendon. Patient was referred to our hospital for further investigations and management.

MRI of the right shoulder was done and showed complete tear of the right subscapularis tendon (with mild retraction) along with the evidence of multiple foci of ossification in it. Coracohumeral distance was decreased and measured 4.5 mm (Figures 2-4). Effusion was also noted in the right shoulder joint and subcoracoid bursa (Figures 3,4). Limited sequences of the contralateral shoulder (of both CT & MR) were also taken for comparison and showed similar but smaller focus of ossification in the left subscapularis tendon (Figures 5,6). However, the tendon was intact. Open repair of right subscapularis tendon to its insertion site at lesser tuberosity was done along with the excision of ossifications. Patient recovered function of the right subscapularis including adduction and internal rotation.
Discussion

Subscapularis is the largest and most powerful muscle of the rotator cuff and has an important role in shoulder motion (especially internal rotation, adduction) and glenohumeral stability. Tendon ossification is a metaplastic process, as it consists of cells like osteoblasts, osteoclasts, osteocytes and the structural features of mature bone (including Haversian canals) along with the mineral components of the bone. The etiology of the heterotopic ossification of the shoulder is poorly understood. The proposed mechanisms are: traumatic brain injury; spinal cord injury; local trauma.

Figure 1 Axial CT images of the right shoulder (bone window) showing multiple foci of ossification in the right subscapularis tendon.

Figure 2 Axial T1 weighted MRI images of the right shoulder showing multiple hyperintense foci with low signal rim in the right subscapularis tendon confirming ossifications. The coracohumeral distance is decreased.
causing malunion or microfracture of the lesser tuberosity or even the coracoid; shoulder surgery; long-term evolution of osteochondritis; evolution of a previous calcification that had developed a blood supply; tissue hypoxia leading to metaplasia of tendon to fibrocartilage and ossification representing just an accessory bone. Absence of history of old trauma, bilaterality of lesions and subcoracoid impingement in our case rules out many theories. We postulated tissue hypoxia leading to metaplasia of tendon to fibrocartilage and subsequently evolution of previous calcification to ossification as the most likely cause of ossification in the case presented (1,2).

Clinical manifestations ranging from anterior shoulder pain with tenderness due to subcoracoid stenosis or impingement and tear of subscapularis tendon have been

Figure 3 Axial gradient recalled echo images of the MRI of the right shoulder showing ossifications and complete tear of the right subscapularis tendon along with minimal effusion in the right shoulder joint.

Figure 4 Sagittal fat saturated T2 weighted image (A) and axial fat saturated proton density image (B) of the right shoulder showing tear of the right subscapularis tendon with effusion in the right shoulder joint and subscapularis recess.

Figure 5 Axial CT image of the left shoulder showing ossification in the left subscapularis tendon.
reported by various authors (1-3).

To our knowledge, till now only three studies on ossification of subscapularis tendon have been reported. Peidro \textit{et al.} in 1999 reported first case of multiple foci of ossification with in the subscapularis tendon accompanied by subcoracoid impingement. Busilacchi \textit{et al.} [2012] described another case of heterotopic ossification of subscapularis tendon (causing subcoracoid impingement) in a patient with tuberculosis, who was managed with arthroscopic en bloc removal of ossification. Thereafter in 2014, Sohn \textit{et al.} presented a case of isolated complete tear of the subscapularis tendon associated with huge ossification within its substance (1-3).

Conservative treatment (physiotherapy and oral non-steroidal anti-inflammatory drugs) is reserved for asymptomatic patients or cases with mild anterior shoulder pain. Patients not responding to supportive management or with subcoracoid impingement or with tears of subscapularis tendon are best treated with arthroscopic excision of ossifications with or without tendon repair. Coracoplasty is also done in patients with decreased coracohumeral space and subcoracoid stenosis. Open surgery is indicated in patients with huge ossifications or ossifications at bone tendon interface not amenable to arthroscopy (1-4,6). More prospective studies would be required for defining appropriate treatment protocols.

**Conclusions**

Ossification of the subscapularis tendon is a rare entity with poorly understood etiopathogenesis. It should be considered as an uncommon cause of subcoracoid impingement and isolated subscapularis tear, which can be successfully treated with complete excision of the ossification and tendon repair.

Learning points:

(I) Subscapularis is the largest and most powerful muscle of the rotator cuff and has an important role in shoulder motion (especially internal rotation, adduction) and glenohumeral stability;

(II) Ossification of the subscapularis tendon is a rarer lesion compared to calcific tendonitis and is a rare cause of anterior shoulder pain, subcoracoid impingement and subscapularis tear;

(III) Tendon ossification is a metaplastic process, as it consists of cells like osteoblasts, osteoclasts, osteocytes and the structural features of mature bone (including Haversian canals) along with the mineral components of the bone;

(IV) The proposed mechanisms are traumatic brain injury; spinal cord injury; local trauma causing malunion or microfracture of the lesser tuberosity or even the coracoid; shoulder surgery; long-term evolution of osteochondritis; evolution of a previous calcification that had developed a blood supply; tissue hypoxia leading to metaplasia of tendon to fibrocartilage and ossification representing just an accessory bone.

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None.
Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Informed Consent: Informed consent was obtained from all individual participants included in the study.

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