CASE REPORT
Massive Heterotopic Ossification in the Subdeltoid Space after Shoulder Surgery and Symptomatic Improvement from Conservative Treatment: A Case Report

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Background: We herein report a case of heterotopic ossification (HO) in the subdeltoid space after open resection and rotator cuff repair in a patient with refractory calcific tendinitis. Case: A 56-year-old man was admitted to our hospital because of right shoulder pain and difficulty in raising his arm. The patient was diagnosed with calcific tendinitis with contracture of the right shoulder joint and was scheduled for surgery because of refractory shoulder pain. Calcium removal was performed using a mini-open approach. Postoperative radiographs showed no calcium deposits. There was mild residual pain at 3 months postoperatively, and the range of motion (ROM) had deteriorated when compared to preoperative levels. A massive ossified shadow was observed in the subdeltoid space on radiographs. Etidronate disodium was orally administered, and the patient continued to undergo careful rehabilitation. HO occurred at 3 months postoperatively, matured at 1 year postoperatively, and showed no progression between 1 year and 2 years postoperatively. The clinical symptoms corresponded with the image findings, and restricted ROM and decreased shoulder function scores were observed at 3 months postoperatively. Although the ROM remained restricted and the function score remained low until 1 year postoperatively, a gradual recovery was achieved at 2 years postoperatively, and the patient did not require reoperation. Discussion: Although early diagnosis, evaluation, and resection are recommended for treating HO, the maturation of bone may lead to symptomatic improvement and the prevention of reoperation, provided that careful rehabilitative measures are performed to avoid ankyloses.

Key Words: conservative treatment; massive heterotopic ossification; subdeltoid space

INTRODUCTION

Studies on heterotopic ossification (HO) are often reported in association with spinal cord injuries, brain disorders, burns, and joint arthroplasty.1–4) The ossification mostly occurs in proximal joints, and there are numerous reports concerning the hip and elbow joints.5–7) In terms of the shoulder, excepting reports on joint arthroplasty, there have been reports on HO after superior capsular reconstruction (SCR),8 acromioplasty,9 acromioclavicular joint injury,3 and rotator cuff repair.9,10) In these reports, despite severe restrictions on the range of motion (ROM) and ankylosis, good results have been obtained by surgical interventions to remove abnormal bone when HO progresses and matures.

We herein report a case of HO in the subdeltoid space after open resection and rotator cuff repair on a patient with refractory calcific tendinitis. Although mild ROM restrictions remained, activities of daily living (ADL) disability disappeared, and the patient achieved high patient satisfaction after careful rehabilitation without surgical interventions.

CASE

A 56-year-old man was admitted to our hospital because of right shoulder pain and difficulty in raising his arm. Under careful examination, the patient was diagnosed with calcific
tendinitis with contracture of the right shoulder joint. There was no history of disorders that could induce abnormal blood calcium levels, including parathyroid gland dysfunction, chronic renal failure, or hypervitaminosis.

At the first visit, the patient exhibited no swelling around the right shoulder, no sensation of heat, and no muscle atrophy. There was tenderness in the bicipital groove and insertion of the infraspinatus muscle, and muscle weakness in flexion and external rotation was confirmed. There was no improvement after 4 months of conservative therapy (anti-inflammatory analgesics and physical therapy), and the patient was scheduled for surgery because of refractory shoulder pain.

The preoperative active ROMs were as follows: flexion, 135°; abduction, 75°; extension, 55°; and external rotation, 30°. In addition, internal rotation was limited to the Th7 level. The University of California Los Angeles (UCLA) shoulder score was 22 points (Table 1), and the blood test results were normal. The patient’s shoulder joint exhibited an observable calcified shadow directly above the greater tubercle in preoperative radiological findings (Fig. 1).

An area of low T1 signal intensity was found in front of the supraspinous tendon on magnetic resonance imaging (MRI), but there was no cuff tear. Intraoperative findings showed no cuff tear under arthroscopy and no apparent calcium from the joint side/subdeltoid space side. A 3-cm skin incision was made from the midpoint of the anterior edge of the acromion towards the axilla. The anterior deltoid muscle was divided longitudinally along the myofibers. Redness and mild injury were observed at the insertion of the supraspinous muscle, and a discharge of white calcium deposits was observed upon incision.

Table 1. Preoperative and postoperative values for functional outcomes

<table>
<thead>
<tr>
<th></th>
<th>Preoperative</th>
<th>3 months</th>
<th>6 months</th>
<th>1 year</th>
<th>2 years</th>
</tr>
</thead>
<tbody>
<tr>
<td>Flexion</td>
<td>135</td>
<td>105</td>
<td>120</td>
<td>110</td>
<td>120</td>
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<td>Abduction</td>
<td>75</td>
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<td>Extension</td>
<td>55</td>
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<td>External rotation</td>
<td>30</td>
<td>5</td>
<td>10</td>
<td>15</td>
<td>25</td>
</tr>
<tr>
<td>Internal rotation</td>
<td>Th7</td>
<td>L4</td>
<td>L3</td>
<td>Th12</td>
<td>Th8</td>
</tr>
<tr>
<td>UCLA score</td>
<td>22</td>
<td>25</td>
<td>28</td>
<td>23</td>
<td>32.5</td>
</tr>
<tr>
<td>Patient satisfaction</td>
<td>-</td>
<td>55</td>
<td>70</td>
<td>70</td>
<td>70</td>
</tr>
</tbody>
</table>
When the calcium deposit was curetted and refreshed, a large 1.5 × 2 cm defect was formed in the rotator cuff, and we sutured the defect with a transosseous suture while maintaining a position at 0° shoulder abduction. We confirmed that there were no calcium deposits on the postoperative radiograph (Fig. 2). When the calcium deposit crystals were examined by infrared absorption spectrometry, calcium phosphate was detected. We were unable to quantify the individual components because of protein contamination. After surgery, the patient underwent rehabilitation for the rotator cuff tear. The arm that underwent surgery was fixed with an abduction pillow for 2 weeks at 70° abduction and 30° horizontal flexion. Elbow joint active flexion/extension exercises, shoulder girdle (trapezius, rhomboids major and minor, levator scapulae, and serratus anterior) relaxation, and passive shoulder ROM training were started on the day after surgery. At 2 weeks after surgery, the pillow was replaced with a small abduction pillow. Three weeks after surgery, active ROM training in the complete hang-down position was initiated. Three months after surgery, muscular strength reinforcement training was started on the extrinsic muscles, and light work was permitted.

At 3 months postoperatively, there was mild residual pain, and the ROM had deteriorated compared to the preoperative levels. The postoperative active ROMs were as follows: flexion, 105°; abduction, 55°; extension, 35°; and external rotation, 5°. The internal rotation was limited to the L4 level. The UCLA score was 25 points, and the visual analog scale (VAS) for patient satisfaction was 55 out of 100 points. A massive ossified shadow was observed in the subdeltoid space on radiographs (Fig. 2). Etidronate disodium was orally administered (1000 mg/day for 3 months, followed by withdraw for 3 months, repeated for 3 courses), and the patient continued to undergo careful rehabilitation.

The 1-year postoperative active ROMs were as follows: flexion, 110°; abduction, 60°; extension, 40°; and external rotation, 15°. The internal rotation was limited to the Th12 level. The UCLA score was 23 points, and the VAS for patient satisfaction was 70 points.

Pain disappeared at 2 years postoperatively. The 2-year postoperative active ROMs were as follows: flexion, 120°; abduction was 60°; extension, 45°; and external rotation, 25°. The internal rotation was limited to the Th8 level. The UCLA score was 32.5 points, and the VAS for patient satisfaction was 70 points. Although radiographs and computed tomography (CT) imaging showed that the ossification remained at 2 year postoperatively, good mobility of the glenohumeral joint was confirmed in radiographs showing anteroposterior exposure with the shoulder in elevated positions. There was no progression of ossification between 1 year and 2 years postoperatively (Fig. 2, 3). MRI also demonstrated extensive ossification at the inferior surface of the deltoid, and the cuff integrity after rotator cuff repair was good (Fig. 4). The pain disappeared, and the patient decided not to undergo surgery and opted for observation because of the mild nature of ADL-related disorders. The patient was informed that data from his case would be submitted for publication and provided consent.
HO refers to abnormal periarticular bone formation in soft tissues and appears as lamellar bone with irregular cement lines on plain radiography. The exact mechanism and pathophysiology that leads to HO formation remains unknown. In HO, the bone grows at triple the normal rate and has more than twice the number of osteoclasts of normal age-matched bone.\(^1\)\(^-\)\(^4\) HO is a common phenomenon after spinal cord injury, head injury, neurologic disorders, burns, trauma, and joint arthroplasty.\(^1\)\(^-\)\(^4\)

Soft tissue trauma, bleeding, and disruption of the local vasculature contribute to decreased oxygen tension and a likely bone morphogenetic protein-stimulated upregulation of osteoprogenitor differentiation alongside systemic hormonal sensitization.\(^5\) In the present case, there was no systemic upregulation that increased the patient’s propensity for HO, such as burns or neurologic injury. During surgery to alleviate the pain of refractory calcific tendinitis, calcium removal was performed under a mini-open approach, and there may have been damage to soft tissues and decreased blood flow.

Although two cases of minor HO in the insertion of the rotator cuff after calcific tendinitis have been reported in the
literature, to the best of our knowledge, there have been no reports of a massive HO as described in the present case. Periarticular HO of the shoulder has been previously reported, and there have been numerous reports on HO after hemi-, total-, or reverse-shoulder arthroplasty. The progression of HO after arthroplasty rarely leads to complete bony ankylosis. In addition, even in large HO, only mild restrictions of ROM are reported, and most cases are asymptomatic. On the other hand, excepting reports on HO that required surgical intervention in the bone, such as arthroplasty around the shoulder and fracture repair, HO after SCR, acromioplasty, acromioclavicular joint injury, and rotator cuff repair has been described in the literature. Because ROM was severely restricted in these cases, leading to ankylosis with bone bridging the acromion to the humerus, resection of the HO or glenohumeral fusion was required at 4–44 months after the initial surgery.

To prevent postoperative recurrence, the conventional advice on HO management has been to wait for the HO to mature and become less radiologically active before surgical intervention. However, recent experience with HO around the shoulder and elbow has suggested that earlier interventions can provide successful results. Because delayed intervention can cause contractures and muscle atrophy, early excision and exercise may be effective for the treatment of HO to achieve a low incidence of recurrence and satisfactory function. Open resection was the only way to obtain an effective release of ankylosis.

Because our case involved calcific tendinitis, surgery was performed under a mini-open approach, and a rotator cuff repair was performed for the resected defect. Subsequently, massive ossification occurred in the subdeltoid space; however, the ossification did not lead to bony ankylosis and was relieved with conservative treatment. HO can often be detected by radiographs at approximately 6 months from onset, and reports suggest that it becomes less radiologically active at 12–24 months. In our case, HO occurred at 3 months postoperatively and had matured at 1 year postoperatively. There was no progression between 1 year and 2 years postoperatively. In addition, the clinical symptoms also corresponded with the image findings, and restriction of ROM and a decreased shoulder function score were observed at 3 months postoperatively. Although the ROM remained restricted and the function score remained low until 1 year postoperatively, a gradual recovery was achieved with a UCLA score of 32.5 out of 35 at 2 years postoperatively, and the patient did not require reoperation. The ROM did not improve sufficiently because of obstruction from the massive HO in the subdeltoid space at 2 years after surgery. We believe that the lack of improvement can be attributed to the limited ROM of 60° abduction at 2 years postoperatively and the position of the HO, which was located laterally to the shoulder and obstructed motion in the abduction direction.

Our conservative treatment involved a careful rehabilitation program and the oral administration of etidronate di-sodium. At high doses, etidronate disodium can suppress calcification by binding with hydroxyapatite at the site of ossification, thereby suppressing HO. To suppress HO, patients with a high risk of HO are orally administered with the drug.

The morphology of the bone is formed over time by a physiological remodeling process following Wolff’s law of adaption to mechanical stress. The morphology of the bone around the shoulder may be associated with its clinical function. Because of continued passive and active shoulder joint ROM training, there was no ankylosing bone that bridged the acromion to the humerus. Careful rehabilitation may have contributed to the maintenance of physical function and the prevention of further progression of HO that could have led to ankyloses. Although early diagnosis, evaluation, and resection are recommended for treating HO, the maturation of bone may lead to symptomatic improvement and prevention of the need for reoperation, provided that careful rehabilitative measures are performed to avoid ankyloses, as was done in the present case.

There are some limitations to this study. The patient was diagnosed with HO based on its features, including being located at a frequent site of occurrence, the clinical course, and imaging findings. However, the trabecular bone structure could not be pathologically confirmed because tissue samples were not collected. Therefore, the differential diagnosis of calcification could not be definitively ruled out. We were unable to collect surgical specimens because of the improvements after conservative treatment. Moreover, biopsy was not performed to avoid the risk of HO recurrence.

**CONFLICTS OF INTEREST**

The authors received no specific funding for this project.

**REFERENCES**


