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Case Report

Complex regional pain syndrome type I (shoulder-hand syndrome) in an elderly patient after open cardiac surgical intervention; a case report

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Abstract. We described the first case report in the literature who developed Complex Regional Pain Syndrome (CRPS type I) symptoms in his right shoulder and right hand within 15 days after open cardiac surgery and discussed shoulder-hand syndrome (CRPS type I) and frozen shoulder diagnosis along with the reasons of no report of CRPS type I in these patients. We also speculated whether frozen shoulder seen in postthoracotomy and postcardiac surgery patients might be CRPS type I in fact.

Key words: Complex regional pain syndrome, cardiac surgery, frozen shoulder

1. Introduction

Complex Regional Pain Syndrome (CRPS) is complication of injuries which is seen at the distal end of the affected area characterized by pain, allodyni, hyperalgesia, edema, abnormal vasomotor and sudomotor activity, movement disorders, joint stiffness, regional osteopenia, and dystrophic changes in soft tissue (1,2). There are two types of CRPS according to absence (type 1) and presence (type 2) of nerve injury (3).

Adhesive capsulitis or frozen shoulder characterized by shoulder stiffnes as in CRPS type 1 is regarded as a distinct clinical entity showing a benign and regular course. Three stages, each lasting 4-6 months, mark the clinical course (4). Thoracic procedures are considered to be among the most painful surgical incisions and are associated with considerable postoperative pain and shoulder dysfunction, severely affecting mobility and activities of daily living.

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Improper patient positioning, muscle division, perioperative nerve injury, rib spreading, and consequent postoperative pain influence the patient's postoperative shoulder function and quality of life (5). In a study Tuten HR et al retrospectively evaluated for the incidence of adhesive capsulitis of the shoulder of two hundred fourteen consecutive male cardiac surgery patients. Thirty-five patients who had shoulder complaints were identified and evaluated. 3.3% incidence (seven patients) of adhesive capsulitis of the shoulder with no shoulder-hand syndrome in a male post-cardiac surgery population was established (6).

Apart from frozen shoulder reports after cardiac surgery, CRPS type 1 has not yet been reported as a complication of cardiac surgery. In this report we describe a cardiac surgery patient who developed CRPS type 1 (Shoulder-hand syndrome) in his right shoulder and hand.

2. Case report

A 67 year-old man admitted to rehabilitation clinic with swelling over the dorsum of the right hand, point tenderness over right hand, wrist, and shoulder restricted range of motion (ROM). Symptoms started 15 days after being operated for open cardiac by-pass operation and were of insidious onset. During the subsequent month the patient developed severe right shoulder pain,

increased restricted ROM of the right shoulder and progressive weakness of the right hand and wrist and was having difficulty performing daily activities such as shaving, brushing his hair and teeth, and writing. He also reported occasional paresthesia in the right hand and arm. The swelling persisted and the patient noted that the affected hand had decreased range of motion and increased temperature in comparison with the asymptomatic hand. His medical history was unremarkable aside from being open cardiac surgical intervention (coronary bypass operation) three months ago. Plain radiographs revealed patchy osteoporosis in the right wrist carpal bones, hand, fingers, distal ulna and radius, right proximal humerus and right shoulder. (Figures 1A-1B, 2A-2B).

Figure 1A. Figure 1B.

Fig. 1A-1B. Plain radiographs of the hands show patchy osteoporosis in right hand and fingers.

In three-phase bone scintigraphy there was increased 99mTc methylene diphosphonate (Tc-MDP) uptake in right coracoid, acromion and humeral head (Figure 3).

On physical examination active range of motion of the right shoulder were; abduction 40 degree, flexion in the sagittal plane 45 degree, lateral rotation 5 degree. There were also restrictions in passive range of motion; abduction 50 degree, flexion in the sagittal plane 60 degree, lateral rotation 10 degree. There were paresthesia, swelling, and increased temperature in the right hand and arm in comparison with the asymptomatic left hand.

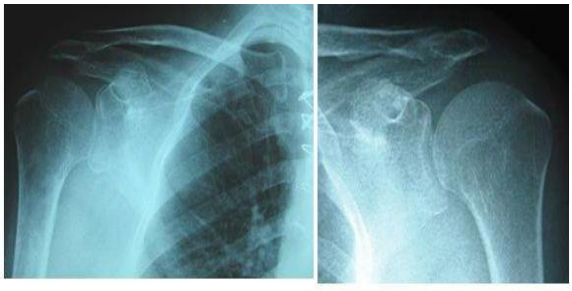

Figure 2A. Figure 2B.

Fig. 2A-2B. Plain radiographs of the shoulders show patchy osteoporosis in right shoulder.

Fig. 3. Increased 99mTc methylene diphosphonate (Tc-MDP) uptake in right coracoid, acromion and humeral head

Based on these clinical and radiological findings, diagnosis of frozen shoulder and shoulder-hand syndrome (CRPS type 1) was made.

3. Discussion

CRPS type 1 is a clinical diagnosis. Diagnostic criteria of CRPS type 1 are the presence of pain and hyperesthesia on the shoulder and the hand, edema of the hand–wrist and fingers, change in the color and temperature, and presence of sweating, limitation in the ROM of the shoulder and the hand (1-3).

Various medical treatments (pacemaker implantation, drawing blood, local anesthesia, surgery, cast, etc.) as well as some diseases including stroke and myocardial infarction may induce CRPS type 1 (7).

Mueller XM et al studied the location, distribution, and intensity of pain in a sample of adult cardiac surgery patients during their postoperative hospital stay and found shoulder pain in %0.5-6.5 of the patients (8).

The majority of CRPS cases occur after orthopedic surgical procedures. But to our knowledge, consistent with the literature although frozen shoulder reported a 3.3% incidence (seven patients among two hundred fourteen consecutive male cardiac surgery patients), shoulder-hand syndrome (CRPS type 1) after cardiac surgery has not been reported so far, which is an interesting situation (6,9).

There may be two reasons of this surprising situation. 1. Frozen shoulder may be a CRPS type 1 process. 2. CRPS type 1 co-exist with frozen shoulder may be overlooked.

Our observations in our case (frozen shoulder and CRPS type 1 diagnostic symptoms co-existincy) and following explanations in the literature support these hypotheses, possibly leading to earlier diagnosis and extended therapeutic, rehabilitative management of CRPS (Shoulder-hand syndrome) after cardiac surgery.

The term ‘‘frozen shoulder’’ is defined as a clinical condition with restricted active and passive range of motion (ROM) in all directions, including flexion, abduction, and rotation. Frozen shoulder has been reported subsequent to non-shoulder surgical procedures, such as cardiac surgery, cardiac catheterisation through the brachial artery (6).

As well as CRPS type 1 diagnosis, frozen shoulder is a clinical diagnosis. The three hallmarks of frozen shoulder are insidious shoulder stiffness; severe pain, even at night; and near complete loss of passive and active external rotation of the shoulder (1,9).

There are also references in the literature that assume frozen shoulder to be an CRPS type 1 process. Diagnostic and clinical features of the frozen shoulder and CRPS type 1 are similar in many aspects. Three-phase 99mTc-MDP bone scintigraphy shows an increased uptake in affected areas in both diseases, while plain radiographs show a progressive demineralisation (1,9,10).

The symptoms of CRPS type 1 often progress in three stages—acute, dystrophic, and atrophic. The acute stage occurs during the first 1–3 months and may include burning pain, swelling, increased sensitivity to touch, increased hair and nail growth in the affected region, joint pain, and color and temperature changes (9).

As seen in CRPS type 1, there is a triphasic natural history of the frozen shoulder: the painful phase, which, as the pain eases, leads to the adhesive/frozen phase, when limitation of movement reaches its extreme.

This is followed by the resolution phase, which leaves significant numbers of patients with residual limitation of shoulder movements (10).

As a result recognising of shoulder-hand syndrome (CRPS type 1) after cardiac surgery leads to earlier diagnosis and extended, more effective therapeutic and rehabilitative management.

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