

CLINICAL CHARACTERISTICS AND TREATMENT OUTCOMES IN SYNOVIAL CHONDROMATOSIS DISEASE. A CASE REPORT

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ABSTRACT

Background: Synovial chondromatosis is a rare condition that affects synovial joints, bursa or tendon sheaths. Synovial metaplasia can lead to the formation of nodules and loose bodies in the joint. The exact aetiology is not yet well known. This study aims to present two cases of synovial chondromatosis of the shoulder treated by arthroscopic surgery followed by specific physical therapy.

Case summary: We describe the clinical, radiographic, histological characteristics, furthermore, arthroscopic and rehabilitative treatment of two patients (A and B) with synovial chondromatosis. This rare disease was diagnosed at "Centro Clinico Diagnostico G.B. Morgagni" in Catania, between January 2015 and February 2018. In patient A the arthroscopy allowed complete recovery of the loose bodies and the specific rehabilitative program allowed the recovery of the Range of Motion and the decrease of the pain; in patient B the complete removal was not possible.

Conclusions: Our experience suggests that these rare conditions can be suspected in patients with characteristic symptoms and radiographic disorder.

Keywords: synovial chondromatosis, loose bodies, arthroscopy, shoulder.

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Introduction

Synovial chondromatosis (SC) is a rare condition that affects synovial joints, bursa or tendon sheaths. It is usually mono-articular occurs bilaterally 10% of the time⁽¹⁾. Is most commonly seen in patients between the third and the fifth decades of life and the prevalence in men is up to three times greater than in women⁽²⁾. SC has been reported in more than thirty locations, the most commonly involved joint is the knee followed by, in descending order of frequency, hip, shoulder, elbow, ankle and wrist⁽³⁾. Other locations where the lesions may be seen include bursae and tendon sheaths in the hands and feet in particular⁽⁴⁾. The exact aetiology is not yet well known, and SC may be considered metaplastic⁽¹⁾.

Histologically, synovial cells undergo metaplasia to chondrocytes, which produce multiple nodules of cartilage⁽⁵⁾. These nodules eventually break off and present as loose bodies in the joint. The majority of loose bodies are produced at the transitional zone between the synovium and articular cartilage; this is hypothesised to be due to the high density of pluripotent stem cells localised to this area⁽⁶⁾. Two-thirds of these foci undergo endochondral ossification, the rest of these bodies remain non-ossified⁽¹⁾. Although the loose bodies typically occur within the synovium-lined spaces, extra-articular involvement can happen if loose bodies escape through the joint capsule into the tendon sheath or bursa^(1,7,8). Clinical presentation may be asymptomatic or may include joint pain, decreased range of motion, swelling, recurrent effusions, crepitus, locking and loose bodies

symptoms such as palpable mass and joint line tenderness. Patients less commonly will report instability or evidence of compromise of surrounding neurovascular structures⁽¹⁾.

SC tends to be chronic and progressive, early diagnosis and rapid treatment can prevent irreversible cartilaginous erosions about the joint^(2,7,9). Synovial chondromatosis can usually be diagnosed on radiographs, but magnetic resonance imaging (MRI) and computed tomography (CT) can be useful in confirming the diagnosis in the early stages of the disease^(6,7). Because SC can be self-limiting conservative management with nonsteroidal anti-inflammatory drugs, activity modification, and cryotherapy⁽¹⁰⁾ can be considered. Conservative management would not be sufficient for a decreased range of motion or locking symptoms, the only effective treatment is surgical and consists of open or arthroscopic removal of loose bodies and with or without synovectomy. This study aims to present two cases of synovial chondromatosis of the shoulder treated by arthroscopic surgery followed by specific physical therapy.

Case presentation

The present case series study was carried out at the Centro Clinico Diagnostico G.B. Morgagni" in Catania, from January 2015 to February 2018 and presents the results of two patients with synovial chondromatosis treated by arthroscopic surgery and rehabilitation. All subjects gave their informed consent for inclusion before they participated in the study. The study was conducted in accordance with the Declaration of Helsinki, and the Ethics Committee of Polyclinic approved the protocol "G.B. Morgagni" Mediterranean Foundation, Orthopedics Traumatology and Rehabilitation Unit, Catania, Italy.

Clinical presentation

Patient A was a 44-year-old male while Patient B was a 31-years-old male.

Patients were both right-hand dominant and presented with pain in the left shoulder from one year. There was not a history of trauma or other pathological conditions. They reported pain at rest and with activity, with much pain during the night.

Physical examination

On physical examination, in both patients, shoulder movements were not limited, but painful in all directions. Only Neer test elicited pain, and the

other tests were negative. Plain radiography showed multiple radiopaque lesions inside the glenohumeral joint in-Patient A (Fig. 1), plain radiography and computerised tomography (TC) showed radiopaque lesions in the space above and below coracoid in-Patient B (Fig. 1). These physical and radiologic findings led to the diagnosis of synovial chondromatosis. Arthroscopic surgery was therefore planned for both patients.

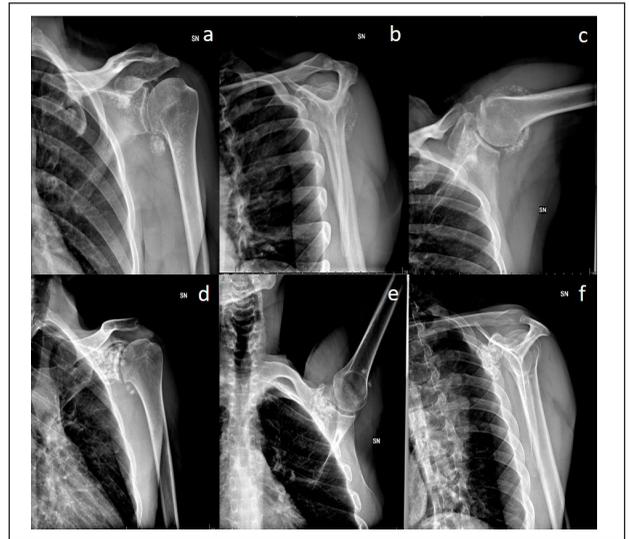


Fig. 1: (a, b, c) Patient A: Preoperative X-ray showing periarticular loose bodies around glenohumeral joint, and proximal epiphysis of the humerus. (d, e, f) Patient B: Preoperative X-ray showing periarticular loose bodies around the scapula.

Surgical technique

Arthroscopy was applied to the left shoulder with the patient in the beach-chair position under general anaesthesia. Standard posterior, lateral and anterolateral portals were used. The diagnostic glenohumeral arthroscopy showed severe synovial proliferation with numerous loose bodies (in patient A and B) and modest signs of degenerative arthritis (patient A). A large number of loose bodies were removed, the size ranged from a few millimetres to over a centimetre in diameter (Fig. 2). A complete synovectomy was also performed. In both patient, the histological examination of the synovium reported a chronic non-specific sclerosing villonodular synovitis (Fig. 2).

Physical therapy

We have divided the rehabilitation into 4 phases:

1° Phase: 0-1 weak

The arm was immobilised for one week with Gilchrist brace, and then the patient started physiotherapy. During this phase, the patient performed

exercises on all the movement planes for the cervical spine, the hand, the wrist and the elbow of the affected limb and the contralateral shoulder.

2° Phase: 1-4 week

Passive, active assisted, and active range of motion exercises was initiated. Physiotherapist began passive mobilisation techniques of the acromion / clavicular joints, sternum/cost/ clavicle joints and scapular / thoracic joints. The passive mobilisations of the glenohumeral joint in anterior flexion, in abduction on the scapular plane, in the intra and extra rotation, also began. Hydrokinesitherapy was also recommended.

3° Phase: 1°-2° month

We proceeded with the exercises of the previous phase, increasing the mobilisations for duration, intensity and amplitude of the joint excursion. In this phase, the isometric exercises for the intrinsic and extrinsic muscles of the shoulder are increased.

4° Phase: 2°-4° month

Progression of the third phase exercises by increasing the mobilisations on all the movement planes and loads of the muscular strengthening exercises. Exercises to allow the patient to resume his work. Each phase consists of 3 weekly meetings, and each meeting started with hot packs for 20 minutes and ended with ice for 20 minutes.

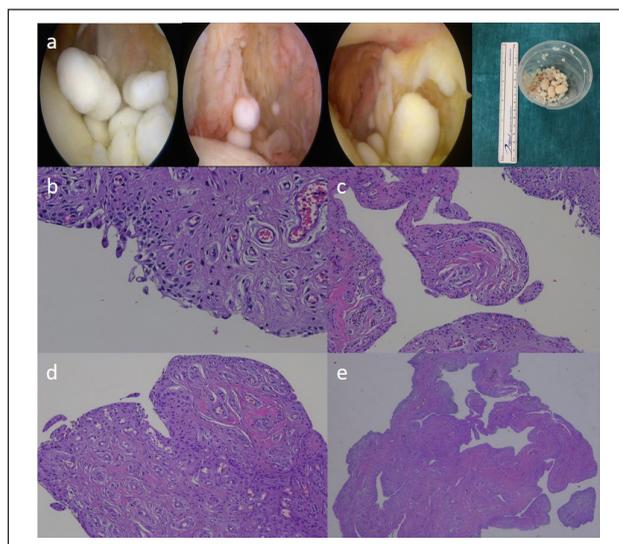


Fig. 2: (a) Arthroscopic removal of loose bodies. Note the different size of the loose bodies. (b, c) Patient A: Chronic non-specific sclerosing villonodular synovitis. (b) 20x magnification. (c) 10x magnification. (d, e) Patient B: Chronic non-specific sclerosing villonodular synovitis. (d) 20x magnification. (e) 10x magnification.

Outcomes follow up

Postoperative radiography showed the complete removal of all free fragments in Patient A (Fig.

3). Patient A was followed up at two months: he had a pain-free range of motion of the left shoulder. Flexion, abduction, external rotation and internal rotation were 170°, 160°, 40°, T10 respectively.



Fig. 3: (a, b) Patient A: Postoperative X-ray. Anterior-Posterior and Outlet view with complete removal of all free fragments. (c, d) Patient B: Postoperative X-ray. Anterior-Posterior and Outlet view with incomplete removal of all loose bodies with free fragments on the anterior side of the joint.

In patient B, postoperative radiography (Fig. 3) showed the incomplete removal of all loose bodies with free fragments on the anterior side of the joint. On physical examination, two months after arthroscopy and after intensive physiotherapy program, shoulder movements were disharmonious with hypotrophy and flexion, abduction, external rotation and internal rotation were 150°, 140°, -10°, at L4 respectively. Patient B continued physical therapy. At four months follow up a range of motion was complete except for the external rotation (15°) and was disharmonious with hypotrophy. No subsequent improvements in the clinical examination were noted at the subsequent follow-up, despite the physiotherapy and the indication to practice swimming. Two years after the first surgery and physiotherapy, new arthroscopy was necessary for anterior capsular realise and removal of the residual free bodies.

Discussion

Synovial chondromatosis is a rare disorder characterised by benign metaplastic proliferation of synovium in diarthrodial joints with the production

of multiple nodules of cartilage and consequently loose bodies. Once free, the loose bodies receive nutrition from synovial fluid and continue to grow. The loose bodies can also bind to the synovium, or they can be reabsorbed. Although the exact aetiology is not known, the disease may be classified as primary or secondary. Primary SC, also known as idiopathic SC, occurs in an otherwise normal joint. It does not seem related to trauma, synovial irritation, genetics, or infection. Generally, it occurs at an earlier age and is fairly rare^(1, 8). More commonly, secondary SC occurs in older patients in a joint with a disease or an intra-articular process that leads to joint destruction and synovitis, such as osteoarthritis, rheumatoid arthritis, and tuberculous arthritis traumatic injury, osteochondritis dissecans, Charcot joint arthropathic or advanced osteonecrosis⁽⁸⁾.

Moreover, secondary SC may result from a single substantial traumatic event or the mechanical stimulation caused by repetitive trauma⁽¹⁾. The size of the loose bodies can vary from being as tiny as a few millimetres to be as large as a coin⁽¹¹⁾. Primary SC often has small, round and uniform in size loose bodies⁽¹²⁾, whereas secondary SC is characterised by loose bodies of different size^(1, 7, 8). Milgram et al. described SC as a self-limiting disease with three phases from onset to resolution. Stage I disease represents an active, inflammatory intrasynovial process without loose bodies; patients present with localised joint pain and swelling or may be asymptomatic.

Stage II disease involves active synovial proliferation with loose bodies, which are in transition from a pedunculated process to a free loose body. Because of the loose bodies, patients often present with mechanical symptoms and a reduction of the range of motion. Stage III disease is characterised by the presence of multiple loose bodies with minimal synovial disease and minimal inflammation, patients are usually asymptomatic^(2, 6, 13). This classification system has limited practical value to the treating clinician but is useful in order to recapitulate the pathologic progression of the disease.

Accurate differential diagnosis from any condition that results in intra-articular loose bodies or synovial proliferation is essential. Joint effusion, crepitus, and palpable loose bodies are common physical exam findings⁽¹⁴⁾. Due to the vague symptomatology and rarity of the disease, diagnosis is often delayed or missed⁽¹⁵⁾. Multiple cartilaginous nodules and loose bodies are usually visible on plain X-ray, but MRI enables better diagnosis of intra- and extra-articular localisations. The recommended treatment of

synovial chondromatosis of the shoulder is the removal of loose bodies and synovectomy by arthroscopic surgery due to the lower recurrent rates by the use of this method (16). Arthroscopy through multiple ports allows complete visualisation of the glenohumeral joint without releasing the subscapularis tendon⁽²⁾. Complete synovectomy may reduce the incidence of recurrence and prevents joint degeneration. Despite synovial chondromatosis is considered a relatively benign process, the intra-articular loose bodies formed can ultimately lead to joint destruction and degeneration⁽¹⁴⁾. The rate of recurrence is variable.

Urbach et al. reported five cases of patients with synovial chondromatosis of the shoulder, treated with arthroscopic removal of loose bodies and partial synovectomy. In two out of five subjects' radiographs demonstrated the evidence of persisting or recurrent synovial chondroma at follow-up⁽¹⁷⁾. Lunn et al. retrospectively identified 18 consecutive patients with synovial chondromatosis of the shoulder who had arthroscopic treatment between 1989 and 2004. Of these, 15 were available for review at a mean follow-up of 5.3 years. Osteoarthritis was present in 8 patients at presentation and in 11 at the final review. Recurrence of the disease with new loose bodies occurred in 2 patients at an interval of 3 and 12 years postoperatively⁽¹⁸⁾.

Coolican and Dandy state that eighteen patients who underwent arthroscopic treatment for SC of the knee had no loss of range of motion postoperatively. Nevertheless, four of these eighteen patients experienced a recurrence of SC^(1, 19). Aramberri et al. described a technique of loose body removal and synovectomy through an endoscopic subcoracoid approach using a central transpectoral portal, reporting decreases of morbidity. With this technique, it is possible to avoid the potential damage to the brachial plexus due to optimal visualisation of the musculocutaneous nerve and axillary nerve⁽²⁰⁾. A further point of contention is the effectiveness of complete compared with subtotal synovectomy. Lim et al. reported a retrospective review of twenty-one patients with SC of the hip, thirteen of the patients were treated by incomplete synovectomy, the remaining eight were treated by complete synovectomy. They concluded that incomplete synovectomy resulted in a higher rate of recurrence⁽⁹⁾.

Our patients presented with pain and restriction on movements. They were treated with arthroscopic technique, loose bodies removal, complete synovectomy and specific physiotherapy. The clinical result

after two months was good and postoperative X-ray showed the complete removal of all the free fragments in Patient A. In Patient B, despite the same treatment, recovery was slower and less optimal. Recovery was slowed by shoulder stiffness, and a new intervention was needed. Our experience suggests that this rare condition can be suspected mostly in young patients when shoulder movements are not limited, but painful in all directions, and plain radiography shows multiple radiopaque lesions inside the glenohumeral joint. Arthroscopic surgery with the removal of most loose bodies, complete synovectomy and specific physical therapy are needed to improve range of motion and reduce pain. Long term follows up is necessary to verify the effectiveness of the treatment and the recurrence rate. Arthroscopic treatment of shoulder synovial chondromatosis is a recommended option due to its advantage of less surgical trauma, better visualisation during surgery, early recovery and rehabilitation. The central point is the rehabilitation period due to prevent muscular compensations.

Informed consent statement

All subjects gave their informed consent for inclusion before they participated in the study. The study was conducted, as previously described in the current literature⁽²¹⁻²³⁾ and in accordance with the Declaration of Helsinki, and the Ethics Committee of Polyclinic approved the protocol "G.B. Morgagni" Mediterranean Foundation, Orthopedics Traumatology and Rehabilitation Unit, Catania, Italy.

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