

## A Strange Evolution of Rheumatoid Arthritis – Case Report

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### Abstract

This paper brings in discussion a strange case evolution of rheumatoid arthritis in a female patient of 72 years old who was admitted in the Department of Plastic Surgery and Reconstructive Microsurgery for a tumoral structure of 4/8 cm in the right deltoid region, with incision marks on its surface, non-adherent with the surrounding tissues and no inflammatory sign locally. The patient has a history of more than 20 year of rheumatoid arthritis, for whom she had been treated with immunosuppressors (Etanercept) for 10 years. Taking in consideration this fact, the most significant side effect of these medications is the increasing risk of all types of infections, including tuberculosis, especially skin tuberculosis or even an abscess due to a possible local skin reactions after injection of the anti-TNF agent. The deltoid region presented skin incisions, drainage and unsuccessful multiple antibiotic therapy, which led us to question marks regarding the origin of the collection. Rheumatoid involvement of the shoulder seems to be relatively not so common as the involvement of peripheral joints, but painful symptoms in this region are present.

**Keywords:** Rheumatoid arthritis; Effusion shoulder joint; Deltoid region; Synovial fluid

right shoulder, associating impairment of the multiple big joints (shoulders, knee).

### Introduction

Rheumatoid arthritis is an autoimmune and a chronic disease that causes pain, stiffness, swelling and limited motion and function of many joints. While rheumatoid arthritis can affect any joint, the small joints in the hands and feet tend to be involved most often. Inflammation sometimes can affect organs as well, for instance, the eyes or lungs [1-3]. It is characterized by synovial proliferation and a symmetric erosive arthritis of peripheral joints [4]. For the Romanian geographic particularity, it is appreciated that rheumatoid arthritis affects around 1% from general population, so the total number of suffering people are estimated to be up to 180,000 [5]. One of the gold standard in treating rheumatoid arthritis are the biologicals one, they are new class of drugs that have been used since 1998. Currently available biologic agents act as inhibitors of the cytokines IL-1 or TNF. [1] The tumor necrosis factor-alpha (TNF-alpha) blockers and the recombinant interleukin 1 (IL-1) receptor antagonist are effective in patients with active rheumatoid arthritis, they have been associated with reactivation of tuberculosis and possibly development of other opportunistic infections (histoplasmosis, listeriosis, and pneumocystis) [6]. Rheumatoid arthritis affecting the shoulder is typically associated with destruction of the glenohumeral joint and rotator cuff impairment, which can result in severe glenoid erosion [7]. On magnetic resonance imaging the most frequent abnormalities of the shoulder joint are joint effusion, synovial sheath effusion, bone erosion, cartilage irregularity and thinning [8]. In time shoulders malfunction leads to surgical treatment in order to improve function of severely deformed joints that don't respond to medicine and physical therapy. In this paper we present a case of an advanced stage disease that came into our department for a collection situated inferior to the

### Materials and Methods

The patient was admitted in the Department of Plastic Surgery and Reconstructive Microsurgery because she presented a subcutaneous structure of about 8/4 cm, ovoid, non adherent to the surrounding tissues, no inflammatory signs and with multiple drainage incision. The patient had a history of long-standing deforming rheumatoid arthritis (rheumatoid factor positive), for more than 20 years, treated with an anti-TNF inhibitor for 10 years, but also with different disease-modifying antirheumatic drugs (DMARDs - mesalazina, methotrexate, leflunomid). Past history is significant for bilateral total knee replacement in 2010, bronchitis, microcytic anemia, total abdominal hysterectomy for menorrhagia, hypoalbuminemia and right humerus osteomyelitis in childhood. The patient has also chronic problems with her elbow, right shoulder and both hands. A clinical examination made 18 month before she presented to our department, showed that both wrist had synovial thickening with mild tenderness and some restriction in extension and flexion. Shoulder movements were fairly good. She also presents varicose vein at the ankle and few hammertoes changes at the toes. 6 month before she stops taking anti-TNF agent, but she begins one month later the therapy with another anti-TNF inhibitor (golimumab – 50 mg/month). However, even prior to that she noted swelling and discomfort at the right deltoid region. It has since become more swollen and erythematous, but she denies having fever. She has numerous incision – three, along with antibiotics – Cephalexin 500 mg/day, Ciprofloxacin 500 mg/day and she is recommended to interrupt golimumab. At 4 weeks from diagnosed of the lump some fluid is aspirated from it for microbiological analyses, the culture was negative. After completing the cure with Cephalexin, the anti-TNF agent is reintroduced, along with prednisone 5 mg/daily and

vitamins. The right deltoid region still presents erythema and a lump. The current medication at admission was furosemide 40 mg/day, opioid pain medication, acetaminophen – codeine and vitamins. The recurrence of the collection situated in the deltoid region, the unsuccessful multiple antibiotic therapy and the patient's history, guided us to a presumptive differential diagnosis of skin tuberculosis. Golimumab can lower the ability to fight infections. There are reports of serious infections caused by bacteria, fungi, or viruses that have spread throughout the body, including tuberculosis. Some of these infections have been fatal [9]. We performed an ultrasound examination of the deltoid region and the result was: fusiform collection of about 8/2, 2 cm, non-homogeneous content, with gas bubbles, well bounded, with the infiltration of the surrounding tissues. The blood test revealed anemia and no leukocytosis. Surgery is performed on the day of admission, by making an incision in the tumoral structure and removing a mucilaginous, green odorless secretion, the content was in a thick membrane ~ 0,5 cm. Both the content and the membrane were sent to microbiological and morphopathological examination.



**Figure 1:** 3rd days after incision, Mucilaginous secretion and a part of the capsule.

**The result was:**

**Morphopathological exam:** tissue fragments formed by skeletal muscle with areas of lymphoplasmocitary inflammation and micro-hemorrhages, dystrophic lesions of the skeletal muscle. Interstitially dense zone of fibrosis. The taken fragments present areas of necrosis with signs of nonspecific granulation tissue. Examination of the evacuated content: suspicion of anaerobes bacterias, no bacterial growth in aerobiosis. Microscopical examination showed common leukocytes, rare intracellular grampositive cocci, common gram-positive cocci. We perform therapy with: Cefoperazone 4 grams/day for 6 days, with stationary evolution, after which we switched to Ciprofloxacin orally 1000 mg/ day for 7 days. Because the antibiotherapy didn't had any positive results we decide to stop it. Locally the evolution is stationary: every day mucilaginous secretion is evacuated through the wide incision. The remaining cavity was washed with Betadine, Cloramine and we applied Rifampicine powder. Slowly the secretion is decreasing and at 2 week from admission we performed excision of the fibrous capsule.



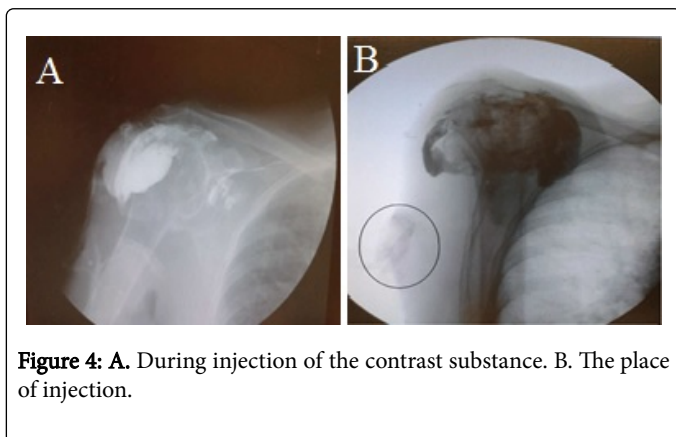
**Figure 2:** Before second surgery, when the quantity of secretion slowly reduced.

During admission the patient is seen by the rheumatologist who recommended a more detailed investigation of her general status, including of her rheumatoid arthritis. The patient received orally Plaquenil (Hydroxychloroquine) 200 mg – 2 pills/day. To exclude pulmonary tuberculosis an X-ray was performed and the result revealed no TB modifications. The abscess is still producing mucous secretion, although the capsule was evacuated, we sent a sample at the National Institute of Pneumology “Marius Nasta” in order to determine if there are any Koch bacteria. The first result was negative. We send another sample containing 25 ml of serous aspect obtained by needle aspiration from another formation situated at the superior pole of the incision. The microscopic examination showed the absence of Koch bacteria.



**Figure 3:** Liquid from the right deltoid region (20 ml).

We injected contrast substance in the formation situated at the superior pole of the incision and a route between it and the right shoulder joint is observed.



At one month from admission we performed suture of the remaining cavity with separate stitches. During admission the patient had initially a Hb of 6.3 g/dl for which we administrated 1 IU of red blood cell, followed by another 2 IU of red blood cell. The patient was discharged and guided toward a rheumatologic department, to receive proper investigations and treatment according to the progression of the disease.

## Discussions

Taking in consideration the long history of immunosuppressants, their possible side effects and the lack of inflammatory signs, our first thought was toward a cold abscess. The most common infection in this kind of treatment is the tuberculosis. We tried to exclude any infections by performing different investigation: blood tests, ultrasonography, radiography for chest and shoulder, anatomopatologic exam tissue, secretion examination. The different diagnosis was made with: folliculitis, cellulitis, actinomycosis, fungal infection and infected tumor. The most relevant investigation was the radiography with contrast substance made in real time, that showed the path of the substance from the place of injection to the bursae of the right shoulder. This guided us to a rheumatological disorder and not to an infectious one. The shoulder girdle consists of the glenohumeral, acromioclavicular and sternoclavicular joints. The proliferative synovitis of rheumatoid arthritis commonly involves not only the glenohumeral joint, but also the other joints and bursae of the shoulder complex, particularly the subacromial-subdeltoid bursa. Persisting proliferative synovitis at the glenohumeral joint may result in cartilage destruction, erosive changes of the joint margins, and eventual rupture of the rotator cuff and biceps tendons [9].

As the biceps tendon sheath is merely an extension of the synovial membrane of the joint, generalized joint effusion in arthritis can lead to fluid in the sheath [10]. According to the earlier studies, as many as 90% of RA patients suffer shoulder symptoms at some point [11]. Although the shoulder joint seems to become involved later than the other upper limb joints in RA, the shoulder has been found to be involved already during the first two years in nearly 50% and during 14 years in 83% of patients [12].

## Conclusions

The radiologic image showing the route of the contrast substance from the incision point to the shoulder was clarifying for the diagnosis of shoulder inflammation with synovial fluid production. We couldn't find any related cases in the medical literature regarding synovial effusion from the shoulder joint through the biceps tendon sheath between the smooth muscle fibers of the deltoid region. The differential diagnosis was initially made with bacteria infection (Koch bacteria), actinomycosis, leprosy, fungal infection and even infected tumors. A plain anteroposterior radiography of the shoulder is necessary to assess any progression of rheumatoid disease and to predict functional outcome in the long term [13]. In the past 25 years, ultrasonography and magnetic resonance imaging (MRI) have been introduced into the clinical practice of diagnosing shoulder problems. MRI is superior to ultrasonography in depicting effusions, synovial thickening and cartilage damage, but in comparison with ultrasonography, MRI is less available, more expensive and time consuming, and less convenient to the patients [14,15].

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