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Case Report

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Shoulder Coccidioidomycosis Masquerading as Synovial Chondromatosis

Sergio Ilic[1]; Siddharth B Joglekar[2]*

[1]Orthopedic Surgeon, VA Central CA Health Care System, USA.

[2]Chief Orthopedic Surgeon, VA Central CA Health Care System, Assistant Professor, UCSF Fresno Orthopedic Surgery, USA.

Abstract

Coccidioidomycosis is an endemic fungal infection in arid areas of Southwestern US and Northern Mexico. It is difficult to diagnose and treat, may take years to make the correct diagnosis, may leave sequelae and could even be fatal. It may mimic other infectious or neoplastic pathology and can be frequently misdiagnosed. The patient in our case report was misdiagnosed and treated for 17 years as synovial chondromatosis. Coccidioidomycosis requires a high index of suspicion to make the correct diagnosis and addition of cultures of biopsy specimens in case of unusual pathology may help make the correct diagnosis.

Background

Coccidioidomycosis is an endemic fungal infection in arid areas of Southwestern US and Northern Mexico. It is difficult to diagnose and treat, may take years to make the correct diagnosis, may leave sequelae and could even be fatal. This patient was misdiagnosed and treated for 17 years as synovial chondromatosis. Coccidioidomycosis frequently mimics other diseases, and a high index of suspicion is needed to make the correct diagnosis.

Case

A 61-year-old, male, diabetic, Filipino patient presented in 2016 complaining of intermittent right shoulder pain since 1999 and an enlarging mass in right shoulder area since 2002. He was evaluated at another hospital with MRI and an open biopsy and diagnosed and treated as Synovial Chondromatosis. He underwent arthroscopic synovectomy and debridement in May 2010. The operative report mentioned no loose bodies in the GH joint but hundreds of osseocartilaginous bodies were evacuated from the subacromial space. Because of continuing pain another arthroscopy was done in Nov 2015, for synovial chondromatosis. Multiple osseocartilaginous bodies were again excised from the subacromial space.

He came to our clinic in July 2016 with continued pain and mild swelling. X-rays were normal, MRI findings were compatible with multiple loose bodies and a partial Rotator Cuff tear (Figure 1). Exam showed mild decrease in ROM and mild weakness. There were no masses, cellulitis, draining sinuses or increased warmth.

He underwent arthroscopy in October 2016 which showed multiple cartilaginous, pedunculated, and free bodies were found in the subacromial and subdeltoid space and similar bodies were detected in the glenohumeral joint which were removed and sampled for histopathology (Figure 2).

Histopathology sections demonstrated eosinophilic fibrous nodules, with chronic granulomatous inflammation with multinucleated giant cells. Studies were negative for amyloid, acid fast

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Correspondance: *Siddharth B Joglekar, Chief Orthopedic Surgeon, VA Central CA Health Care System, 2615 E Clinton Ave, Mail Code 112, Fresno, CA 93703, USA. Email: Siddharth.joglekar@va.gov*

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organisms, foreign body giant cells and cartilage or bone. Gomori's methenamine and Periodic Acid–Schiff–Diastase (PASD) stains showed the presence of yeast forms (Figure 3).

The specimen was further studied for fungal identification by ribosomal focus DNA sequencing using PCR which was positive for Coccidioides immitis and posadasii [1].

Upon questioning, patient remembered having had an episode of Coccidioidomycosis in 1994 when he had an upper respiratory infection with low grade fever, myalgias, cough and positive serology for Cocci. No treatment was given, he was lost to follow-up.

CT of the thorax showed 3.5 cm left upper lung nodule. Bone scan was negative for any other involvement. He was treated with Fluconazole 600 mg daily and later with Voriconazole. Serology remained positive at last follow-up. Repeat MRI showed no recurrence, but patient remains with mild pain and stiffness.

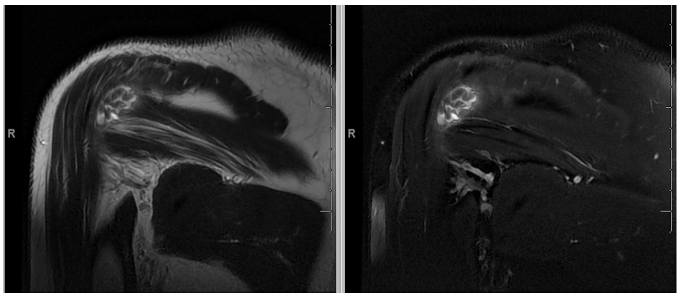

Figure 1: Coronal MRI images T1 and T2 sequence showing presence of multiple loose bodies in the shoulder sub acromial and subdeltoid space.

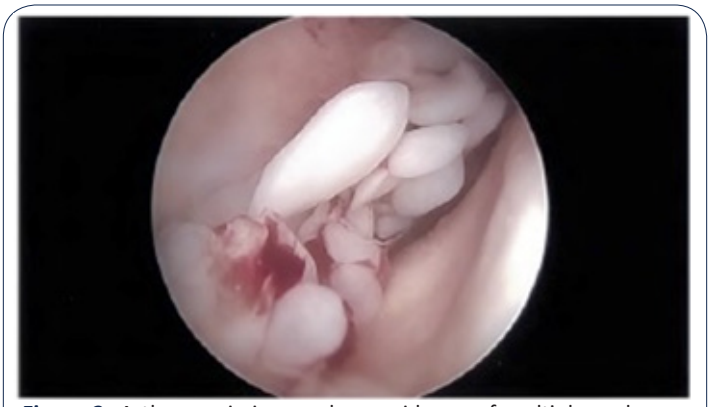

Figure 2: Arthroscopic image show evidence of multiple pedunculated loose bodies in the subacromial and subdeltoid spaces.

Discussion

Coccidiodes Immitis and Posadasii is a dimorphic fungus that is found in the soil of arid regions of the Southwest USA, California Central Valley, Northern Mexico, and parts of South America. It was first reported by Alejandro Posadas, an Argentinian pathologist in 1892. Spores are inhaled during soil disturbances by earthquakes, dust storms, farming or construction. It is commonly known as Valley Fever or Desert Rheumatism in the CA Central Valley.

It is estimated that 80% of persons living in endemic areas become infected within 5 years. It is also estimated that more than 100,000 cases occur annually. Immunocompromised patients are

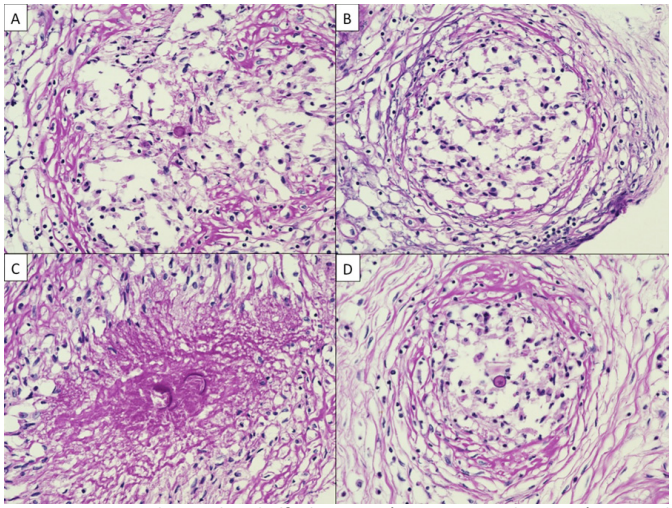

Figure 3: Periodic acid–Schiff–diastase (PAS-D, PAS diastase) stained sections showing fungal forms within necrotizing granulomas in synovial tissue fragments. Fungal forms shown include hyphae, including one budding from a spherule and branching forms (panel A), isolated hyphae (panel B), and spherules (panels C and D).

at risk for dissemination as are people from Philippines, Mexico, African Americans, residents of endemic zones and women in their 3rd trimester of pregnancy. 60% of the cases may have only flu like symptoms which resolve spontaneously without treatment, and it is never diagnosed.

The life cycle starts as a mold in the soil with branching septate hyphae and long filaments. The mold is the least infectious form. Its growths rapidly in the rainy season and the filaments break off into arthrospores which become airborne when the dry soil is disturbed. These disarticulate into single rectangular arthroconidia which are the infective particles which go to the alveoli, become round and begin the yeast stage of their life cycle. Internal division starts within 24 to 72 hours; they become filled with thousands of endospores which are released to form new spherules.

Dissemination occurs in 1% of cases and the most common sites are the lungs, pleura, lymph nodes and skin. In 10 to 50% of these cases skeletal infection occurs with osteomyelitis, septic arthritis, and meningitis. The spine, bony prominences and the epiphysis of long bones are a frequent site of infection. Recurrences are frequent once the treatment is stopped.

The diagnosis is frequently delayed, and misdiagnosis is common [2-5]. Serology is diagnostic but, in many cases, the definitive diagnosis is made either by DNA sequencing on biopsy specimen, I&D of abscesses or aspiration of fluid from the site with fungal cultures. Complement fixation is usually positive and if greater than 1:16 indicates dissemination. IgM antibody titers reach the maximum level at the 3rd week of the disease, can cross react with other fungal diseases. Serology is negative in about 20 to 25% of immunocompromised patients.

Articular findings are usually synovitis, effusion, periarticular bone destruction, osteomyelitis, erosions, and osteopenia [3,5-8]. In our case there were no bony erosions but multiple pedunculated and loose bodies that resembled synovial chondromatosis in the subacromial space and in the glenohumeral joint.

Infection of synovial tissue has been reported previously in the knee. Pollock et al reported four cases which showed findings similar to those in our case, but hyphal forms were not reported [9]. The presence of hyphal forms of C. immitis in tissue specimens is rare and has been reported in cases of meningitis and pulmonary infection but reported for the first time in synovial tissue in our case [4,10]. The primary infection is usually not treated since it is seldom diagnosed and generally is self-limiting with no residual symptoms. Antifungals such as Fluconazole are usually recommended for 6 to 12 months and may be recommended for life in the immunocompromised patient. Amphotericin B is given IV for disseminated disease and intrathecal for meningitis. Synovectomy, I&D, sequestrectomy, arthrodesis, even amputation is sometimes necessary.

Conclusions

Coccidioidomycosis is an endemic disease that is rarely diagnosed in its acute phase when it spontaneously resolves as a flu-like episode. It is also frequently misdiagnosed because it can mimic other diseases. A high index of suspicion is essential when there has been exposure in endemic areas. The treatment can be difficult, and it can frequently recur once medical treatment is discontinued.

Lessons from this case are to get a detailed history, have a high index of suspicion and always consider cultures of biopsy specimens.

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