Case Report

Case of pulmonary sarcoidosis relapsed as unilateral carpal tunnel syndrome: A rare case report

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ABSTRACT

Sarcoidosis is a multisystemic chronic inflammatory disease with variable clinical manifestations and having a tendency to relapse of the disease in the same organ and other organs as well. Extrapulmonary relapse is rarely reported in the literature. Here, we are reporting the case of a 56-year-old female with a known case of pulmonary sarcoidosis for 21 years, which was asymptomatic since then, now presented as unilateral carpal tunnel syndrome secondary to extrapulmonary sarcoidosis relapse. Diagnosis was confirmed by open biopsy, which revealed noncaseating granulomas in resected tissue. She was treated successfully by surgical intervention and systemic steroids.

Key words: Carpal tunnel syndrome, Neuro-sarcoidosis, Noncaseating granulomas

Sarcoidosis is a granulomatous disease that affects multisystems and is characterized by the formation of non-caseating granulomas. That can affect any organ of the body, but the lungs and intrathoracic lymph nodes are involved in over 90% of cases, extrapulmonary involvement occurs in approximately 30% of patients and can be the initial or only manifestation [1]. Prevalence of sarcoidosis is 10–40 patients/one lack population, but in India, this is <5 cases/lack population, and most of the time diagnosed as tuberculosis. The highest prevalence of sarcoidosis is in Sweden. Common extrapulmonary sites include the skin, eyes, liver, spleen, heart, nervous system, and musculoskeletal system. Clinical presentation varies widely depending on the organ involved. Extrapulmonary sarcoidosis highlights the systemic nature of the disease. Timely recognition and treatment are critical, particularly for potentially life-threatening manifestations like cardiac or neurosarcoidosis. Sarcoidosis is naturally having a relapsing tendency in the same organ or other organs. Overall relapse in sarcoidosis is 30–50%. In various studies, this has been pointed out that there is a need to be more vigilant among patients with corticosteroid-induced remissions, because of the high rate of relapses in this context [2]. In contrast, disease progression or clinical relapse is infrequent among patients who spontaneously complete remission of the disease [3]. Clinical manifestations are wide and nonspecific, which creates diagnostic confusion, especially in release cases. If the diagnosis is not suspected timely, then this causes significant damage to that particular organ. Management of relapse cases is usually more difficult and depends on the severity and organ systems involved, with corticosteroids being the first-line treatment. Immunosuppressive agents such as methotrexate or tumor necrosis factor inhibitors are considered in steroid-refractory cases or for long-term management. Extrapulmonary relapse as carpal tunnel syndrome (CTS) is not reported in literature.

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CASE PRESENTATION

A 56-year-old female patient presented to us with a history of pain in the left wrist joint which was progressive and aggravated by wrist movement. She took local symptomatic treatment, but there was insignificant improvement. Since last 1 month, she developed a small tender swelling over the wrist joint, swelling was progressively increasing and is now associated with painful movement and tingling sensation in the distal part of the hand.

On examination, she has mild systemic hypertension with a blood pressure of 156/100, and the rest of the systemic examinations were normal. On review of old records, she was diagnosed and treated for pulmonary sarcoidosis about 27 years back. On examination of the wrist, there was a small swelling of about 3–4 cm, painful with no superficial signs of inflammation or infection. The swelling was not fixed to the underlying bone.

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Based on these clinical findings, she was suspected as a case of unilateral CTS.

We investigated here on the same line by nerve conduction velocity (NCV), X-ray, and magnetic resonance imaging (MRI) of the wrist joint. NCV suggests slowing of conduction in the median nerve, and X-ray does not suggest any significant bony etiology. MRI of the wrist joint revealed soft-tissue showing, hypertense signal on T2, and short-tau inversion recovery sequence is visualized around the flexor tendons suggesting florid synovial thickening (blue arrow) which leads to accentuated waist at the level of carpal tunnel on coronal images (Fig. 1a) and is causing compression over the median nerve within the carpal tunnel (red arrow) making it inconspicuous on axial images (Fig. 1b).

The reports of investigations were correlated with clinical findings, and the diagnosis of carpal tunnel was confirmed. As she was very much symptomatic and there was significant neurological involvement considering, this a surgical intervention was planned.

Carpal tunnel decompression was done through an extended carpal tunnel release approach. Intraoperatively, the median nerve and flexor tendons were encased by hypertrophied synovium, which was extended in the carpal tunnel as well. It was causing mass effect and compressing the median nerve. Extensive synovectomy was done around the median nerve and flexor tendons (Fig. 2).

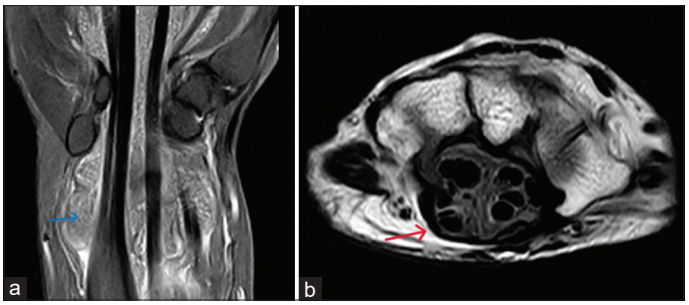

Figure 1: (a) Magnetic resonance image at the level of carpal tunnel level on coronal images (b) and is causing compression over median nerve within the carpal tunnel, making it inconspicuous on axial images

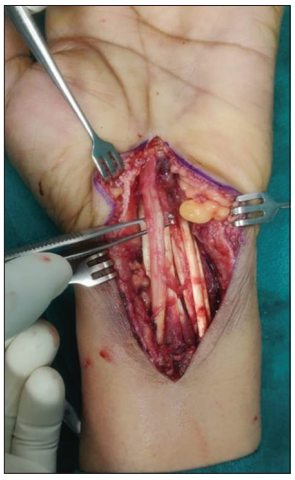

Figure 2: Preoperative picture after extensive synovectomy

The biopsy of resected tissue of the transverse carpal ligament revealed fibrocollagenous and fibroadipose tissue lined partly by synovial cells and showed infiltration by noncaseating epithelioid cell granulomas with Langerhans-type giant cells. Few nerve bundles, lymphoplasmacytic inflammation, and a few congested capillaries were also seen. No necrosis was seen. A diagnosis of granulomatous synovitis was given (Fig. 3). Since the patient was a known case of sarcoidosis and GeneXpert and acid-fast bacilli staining were negative for *Mycobacterium tuberculosis*, a final diagnosis of synovial sarcoidosis was made.

After the tissue diagnosis, he was put on oral systemic steroid therapy with a prednisolone dose of 0.5 mg/kg of body weight and other supportive measures, and responded well. The steroid was tapered within 1 year. There was complete recovery of the lesion.

DISCUSSION

Extrapulmonary sarcoidosis refers to the manifestation of sarcoidosis outside the lungs, which can involve any organs, such as the skin, eyes, lymph nodes, liver, spleen, heart, and bones. Relapse of extrapulmonary sarcoidosis occurs when the symptoms or signs of the disease reappear or worsen after a period of improvement or remission. The relapse can involve the same organ previously affected or a different one. The exact cause of a relapse is not always clear, but it may be related to factors such as, immune system triggers, medication issues, or disease. Reported relapse rates of sarcoidosis range from 13% to 75% depending on the population studied. These relapses typically occur 1 month–1 year after therapy is tapered or discontinued [4].

Sarcoidosis patients treated with infliximab appear to have a very high likelihood of relapse of their disease when the drug is discontinued. In one study in which the drug was withdrawn after 1 year or less, 90% of patients had a recurrence of their symptomatic disease [3]. Patients with sarcoidosis treated with systemic steroids have a higher incidence of relapse in the future; the exact explanation is not given in the literature, but this is hypothesized that patients might have severe disease on clinical presentation [5].

Neuro-sarcoidosis can involve various parts of the nervous system as an isolated organ or in combination with pulmonary sarcoidosis. Neurological manifestation depends upon the organ involved, which include the brain, spinal cord, cranial nerves, and peripheral nerves. Median nerve involvement by sarcoidosis

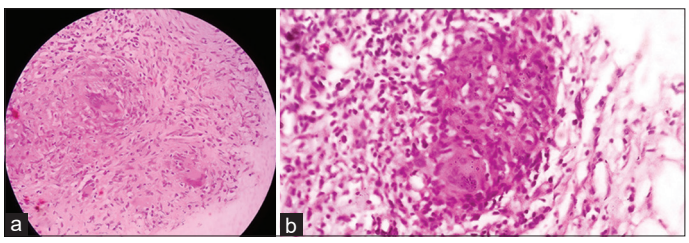

Figure 3: (a and b) Hematoxylin and eosinstained sections reveal large, well-formed non-caseating epithelioid cell granulomas with Langhans giant cells (400×)

and causing CTS is a very rare presentation. CTS occurs when the median nerve in the wrist is compressed due to swelling in the carpal tunnel. Some possible causes of CTS include trauma, arthritis, obesity, metabolic disorders, and hypothyroidism. CTS due to sarcoidosis is a condition where sarcoidosis causes compression of the median nerve as it passes through the carpal tunnel in the wrist. This is relatively less common compared to other manifestations of sarcoidosis but can occur, especially in cases of peripheral neuropathy associated with the disease.

One study tried to correlate the association of CTS and sarcoidosis by doing a nerve conduction study, but the result was not statistically significant. The pathological mechanism of neurologic involvement in sarcoidosis is not clear. This is proposed that granulomatous inflammation of nerve layers, secondarily caused by vasculitic neuropathy, demyelination, panangiitis, and compression by sarcoid tissue, or thick edema under perineural tissue may be the cause [6].

Patients of CTS present with numbness in the thumb, index finger, middle finger, and part of the ring finger and tingling sensation, often described as “pins and needles” in the same fingers. Sometimes, there is a significant weakness in the muscle of the wrist, leading to difficulty with gripping or pinching, and possibly muscle wasting at the base of the thumb. A similar case has been reported in the literature, but this was the first presentation of carpal tunnel syndrome [7].

CONCLUSION

Sarcoidosis is a disease with a relapsing tendency that can involve any pulmonary and extrapulmonary organ. Its clinical differential diagnosis is tuberculosis. In India, tuberculosis is a common disease which causes sarcoidosis to be wrongly diagnosed most of the times and treated as tuberculosis. The case presented here emphasizes that carpal tunnel syndrome due to sarcoidosis relapse is extremely rare and imitate tuberculosis. We should always suspect this etiology whenever a patient develops new organ involvement in pre-existing sarcoidosis so that early diagnosis can be made and therapeutic intervention can be optimized.

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