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Short Communication

A Case of Tuberculous Pyomyositis That Caused a Recurrent Soft Tissue Lesion Localized at the Forearm

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SUMMARY: We present the case of a 20-year-old male who had a non-traumatic soft tissue lesion (4 × 3 cm) with recurrent discharge at his right posteromedial antebrachial muscles; the patient underwent surgery twice, and antibiotic therapy was administered, but no cure was achieved with these treatments. The patient underwent surgery at our medical center. There was no history of pulmonary, gastrointestinal, or genitourinary tuberculosis (TB). Due to suspected pulmonary, genitourinary, and gastrointestinal TB, radiography and computed tomography scans were performed, and these studies disclosed no evidence of a primary origin. The erythrocyte sedimentation rate and the results of purified protein derivate testing were normal. We also detected submandibular lymphadenopathy (LAP) (2×3 cm) localized at a submandibular site in our patient 4 months after his first visit to our clinic. Smears were stained with Ehrlich Ziehl Neelsen (EZN) stain and cultures were grown for *Mycobacterium tuberculosis* complex (MTC); the samples used for these assays had been obtained by incisional biopsy of the forearm lesion and by aspiration of the submandibular lymph node, and they were found to be MTC-positive. Then, a culture for MTC, derived from an induced sputum sample, was found to be positive, despite the negative results obtained with a sputum smear subjected to EZN staining. According to these results, the primary focus of the tuberculous pyomyositis and the submandibular LAP was the lungs. The lesion and submandibular LAP were both treated successfully by the administration of antituberculous chemotherapy.

Tuberculous pyomyositis (TPM) is an extremely rare condition that has previously been described in both immunocompetent as well as immunosuppressed individuals (1-4). It is the least frequent location of extraspinal musculoskeletal tuberculosis (TB) reported in the literature (5). TMP is typically caused by invasion from adjacent structures rather than by primary infection, lymphatic spread, or hematogenous seeding (3).

A 20-year-old male patient complaining of a recurrent soft-tissue lesion (4 × 3 cm) that had exhibited continuous discharge at the right posteromedial antebrachial muscles was admitted to our hospital. The patient had been healthy prior to these discharge episodes, and had been suffering from this non-traumatic wound for 2 years. The patient had undergone irrigation and debridment surgeries at different medical centers. Following the second operation, there had been a pathological diagnosis of granulomatous inflammation. The patient had not postoperatively taken any tuberculostatic medications due the lack of a specific diagnosis.

The patient was afebrile with normal vital signs. Physical examination revealed seropurulent and hemorrhagic discharge with an irregularly marginated lesion located 8 cm distal to the elbow joint. There was hyperemia over the mass, which was soft on presentation (Fig. 1A). Movement of the elbow joint was painful. Radiographs of the antebrachium and humerus showed no bone lesion. The patient had a painless, submandibular lymphadenopathy (LAP) with a diameter 2 × 3 cm; this LAP was clearly observable in the neck during the patient's fourth month of hospitalization (Fig. 1B).

There was no history of pulmonary, gastrointestinal, or genitourinary TB. X-ray images were obtained and computerized tomography (CT) scans of the pulmonary,

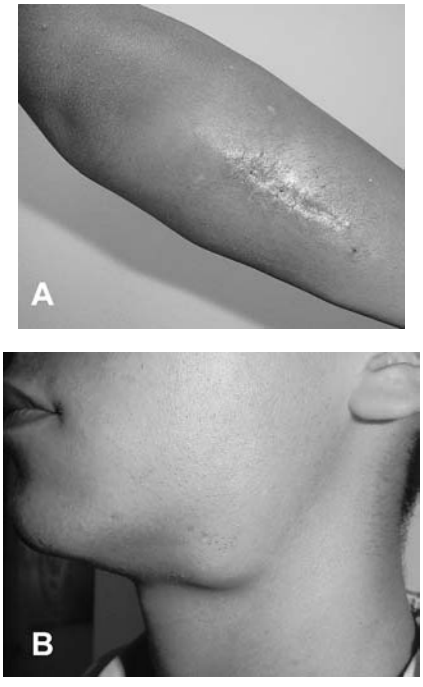

Fig. 1. (A) Incision scatris after the first surgical exposure, and the discharge site located at the line of incision. (B) Submandibular painless lymphadenopathy (2 × 3 cm) in the patient's neck.

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gastrointestinal, and genitourinary systems were performed, and these imaging studies disclosed no evidence of primary foci of the disease. In addition, respiratory function test results were found to be within the normal range.

Immunoserological tests, the whole blood count, routine biochemical marker levels, and urine analysis were all reported to be normal (Table 1). Magnetic resonance imaging (MRI) revealed subcutaneous edema and inflammation of the fatty tissue that started just distal to the elbow joint and continued through one-third of the antebrachium, and was located between the subcutaneous fatty tissue and the extensor muscle planes of the posteromedial antebrachium (Fig. 2A-C).

Surgical exposure was performed and after incision and drainage of the lesion, the purulent and caseous material was discharged. Three specimens were curetted from a deep site in the wound. The histopathological report for the specimen confirmed the diagnosis of caseous granulomatous inflammation (Fig. 3A). During the same procedure, puncture of the left submandibular LAP site was performed. Smears

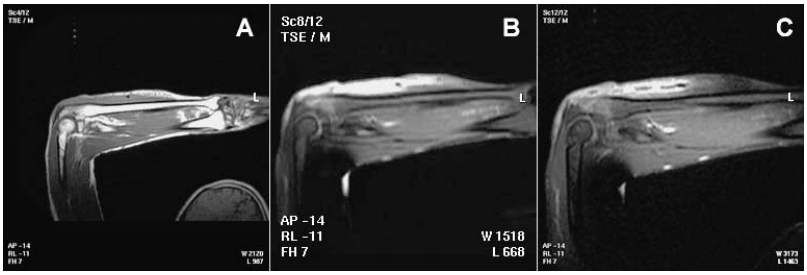

Fig. 2. (A) A T1-weighted sagittal MRI reveals subcutaneous inflamed tissue as isointense to the subjacent muscles. The neighboring ulna were preserved, including its cortical integrity. (B) A T2-weighted sagittal MRI shows the intense edema of the subcutaneous tissue as a highlighted area (hyperintense with respect to the subjacent muscles). (C) A post-contrast (gadolinium) T1-weighted sagittal MRI with fat suppression shows heterogeneous enhancement of the diseased subcutaneous tissue, indicating co-existing inflammation with edema.

Table 1. The results of immunoserological, routine biochemistry tests, whole blood count and urine analysis performed to the patient

Immunoserological tests				Urine analysis			
Test	Result	Reference	Unit	Test	Result	Reference	Unit
PPD skin test	8	0-10	mm/72h	pH	6.5	5-8	–
RF quantitative	10	0-15	IU/ml	Density	1.010	1.005-1.030	–
CRP	11	0-6	mg/L	Glucose	Negative	Negative	–
IgG	9	7-16	g/L	Protein	Negative	Negative	–
IgM	1.0	0.4-2.3	g/L	Bilirubin	Negative	Negative	–
IgA	2.3	0.7-4.0	g/L	Urobilinogen	Normal	Normal	–
Total IgE	35	<100	IU/L	Ketone	Negative	Negative	–
C3	1.1	0.9-1.8	g/L	Nitrite	Negative	Negative	–
C4	0.25	0.1-0.4	g/L	Leukocyte	0	0-4	In each field
Anti-HIV 1+2	Negative	Negative	–	Erythrocyte	0	0-4	In each field
HBs Ag	Negative	Negative	–	Epithelia	0	0-4	In each field
Anti-HCV	Negative	Negative	–				
Routine biochemical tests				**Whole blood count**			
Test	Result	Reference	Unit	Test	Result	Reference	Unit
AST	19	10-40	U/L	WBC	6.09	4.3-10.3	× 10.e3/microL
ALT	7	10-40	U/L	RBC	5.51	4.38-5.77	× 10.e6/microL
ALP	265	38-155	U/L	HGB	14.3	13.6-17.2	g/dL
GGT	13	10-49	U/L	HCT	41.6	39.5-50.3	%
Serum amilase	51	25-90	U/L	MCV	75.5	80.7-95.5	fL
LDH	400	220-450	U/L	MCH	25.9	27.2-33.5	pg
Creatine kinase	59	24-190	U/L	MCHC	34.4	32.7-35.6	g/dL
Protein (total)	6.6	6.4-8.3	g/dL	RDW	14.7	11.8-14.3	%
Albumine	4.2	3.5-5.5	g/dL	PLT	220	156-373	× 10.e3/microL
Serum creatinine	0.9	0.6-1.2	mg/dL	MPV	10.2	6.9-10.8	fL
Uric aside	4.1	3.6-8.2	mg/dL	%NEUT	57.2	41.0-73.0	%
Serum glucose	87	65-107	mg/dL	%LYMPH	31.2	19.4-44.9	%
Urea	22	15-44	mg/dL	%MONO	7.88	5.1-10.9	%
Serum sodium	142	135-145	mmol/L	%EOS	3.3	<6	%
Serum potassium	4.4	3.5-5.5	mmol/L	%BASO	0.387	<1.2	%
ESR	9	0-15	mm/h				

CRP, C-reactive protein; PPD, Purified protein derivate; AST, Aspartate amino transferase; ALT, Alanine amino transferase; ALP, Alkaline phosphatase; GGT, Gamma glutamil transferase; LDH, Lactate dehydrogenase; ESR, Erythrocyte sedimentation rate.

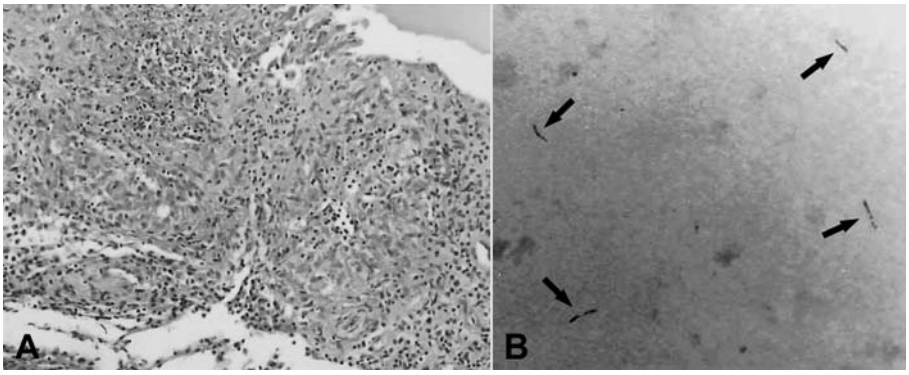

Fig. 3. (A) Hematoxylin-eosin stained sections prepared from the curettage material of a lesion sent for pathology examination. The soft-tissue samples showed granulomatous inflammation with central caseous necrosis. (B) Acid-fast bacilli in a smear stained with EZN, which had been obtained from an incisional biopsy sample.

obtained from this site were stained with Ehrlich Ziehl Neelsen (EZN) stain, and the smears were found to be positive for acid-fast bacilli (AFB) (Fig. 3B). Both conventional (Löwenstein Jensen medium; Salubris Inc., Istanbul, Turkey) and radiometric (BACTEC 460TB culture system; Becton Dickinson Diagnostic Instruments, Sparks, Md., USA) culture examinations of the incisional biopsy specimens of the forearm lesion revealed growth of *Mycobacterium tuberculosis* complex (MTC). Differentiation between MTC and non-tuberculous mycobacteria was achieved by selective inhibition of the MTC in the presence of 5 μl/mL of p-nitro-α-acetyl-amino-β-hydroxypropiophenone (NAP), according to the BACTEC manual. The material aspirated from the submandibular lymph node was positive according to all of these procedures. In addition, a culture for MTC of an induced sputum sample was found to be positive, despite a negative stained smear result obtained with a sample from this induced sputum.

After growth of the MTC was observed in the mycobacteriological culture from the incisional biopsy material and following the intensive radiological and microbiological analyses, the submandibular LAP and TPM in our patient were revealed not to be originated from either a gastrointestinal and genitourinary systems. Although the radiological tests (chest X-ray and thorax CT) performed to visualize the lungs did not reveal any pathology, we concluded that the primary origin of the TPM and the submandibular LAP was the lungs, as based on the growth of the MTC in the mycobacteriological culture of an induced sputum sample.

The MTC strain was found to be sensitive to isoniazid (INH), rifampicin (RIF), streptomycin, and ethambutol (ETB), according to BACTEC 460TB anti-TB drug susceptibility testing. Treatment includes drainage of the lesion, either by aspiration or surgery, as well as specific anti-TB chemotherapy (ATCT). In cases of TPM, as in cases of pulmonary TB, ATCT should be continued for at least 6 months. The patient was administered INH (300 mg/day), ETB (2 g/day), pyrazinamide (2 g/day), and RIF (600 mg/day). After 2 months of this regimen, dual treatment with INH and RIF was initiated and was continued for an additional 10 months. High-dose pyridoxine (10 mg/day) was prescribed to prevent INH-related neuropathy during ATCT. The patient tolerated the ATCT well and no complications occurred. The lesion and the submandibular LAP in this patient were both treated successfully by surgical drainage and subsequent ATCT. The diagnosis of TPM was also confirmed by the success of the treatment.

Pyomyositis is a term used to describe a bacterial infection and the formation of an abscess of the skeletal muscle. The etiology of pyomyositis has not yet been well defined. Pyomyositis is frequently seen in tropical countries, and it can therefore often be defined as tropical pyomyositis or as myositis tropicans. Turkish weather conditions are much less conductive to pyomyositis than are tropical conditions. Thus, the low incidence in Turkey may pose a diagnostic problem (1,6). In general, *M. tuberculosis* is an extremely rare organism determined to be responsible for pyomyositis in imunocompetent patients such as our patient. In our search of the literature written in English, we found a few examples of case reports regarding cases of TPM. This diagnosis is made with increasing frequency in immunodeficient patients, and it has been causally associated with the use of corticosteroids, the presence of HIV infection, the administration of cancer chemotherapy, and renal failure (3,4). However, as noted in this case, TPM can also develop in immunocompetent person (3,7). Logginess processing is a characteristic feature of TPM, which is also associated with extreme to total loss of muscle contractility. Marked edema can develop in cases of TPM as well. The common symptoms are typically mild and include fever, night sweats, malaise, and weight loss (2).

As in the case presented here, the focus of infection was the submandibular lymphadenitis, but this focus occurred after development of the lesion at the pyomyositic origin; it is thought that this course of disease might account for the lack of positive findings upon clinical and radiological evaluation to determine the primary site of TB infection. An alternative etiology might have been some traumatic injury at the site of the upper extremity, which may have led to inoculation of the organism into the muscle or the adjacent tissue. In the present case, however, the patient did not report experiencing any traumatic event involving his upper extremities.

The basic component of a diagnosis of TPM remains clinicians suspicion. Thus, the diagnosis of TPM still poses a diagnostic challenge in the clinical setting, as it requires of clinicians a high degree of awareness of potential, yet equivocal sings. Differential diagnosis can be based on tests of fluid samples acquired by surgical intervention or by fine needle aspiration of the skeletal muscle; relevant differential diagnoses would be expected to include bacterial, tuberculous or fungal abscesses, as well as hematomas and neoplasms. A subacute presentation and the slow progression of this disease could lead to suspicion of TB as well as to a diagnosis involving a non-infectious cause (3,8). A positive tuberculin

skin test is indicative of a TB diagnosis; but a negative test, as in our case, cannot be used to exclude the presence of active TB. The laboratory abnormalities in patients with TB may include an elevated erythrocyte sedimentation rate, significant anemia, leukocytosis, and elevated creatine phosphokinase levels; however, none of these is specific to this particular diagnosis, and none are seen in each case (3,9). The persistence and/or progression of lung infiltration, and/or the inefficacy of broad-spectrum antibiotic therapeutic treatment are potentially predictive of TB. Initial TB investigations were not performed in our case due to the rarity of this disease, which initially confounded diagnostic efforts. To date, some authors have reported that the establishment of a diagnosis of extrapulmonary TB is more difficult when a patient is alive than after a patient has died (9). It should be noted that imaging studies are of great significance in the diagnosis of pyomyositis. The location, extent, and consistency of a mass can be determined by imaging studies such as ultrasound, CT, and MRI, which are most frequently used in the process of establishing a differential diagnosis of TPM (3,6).

The diagnosis of TPM must be confirmed by positive culture results or by histological evidence from incisional biopsy samples. It is essential to obtain fluid samples in order to confirm a diagnosis of TB and to exclude other possibilities; such samples are also important for determining the susceptibility of the organism to ATCT. Specimens obtained from an abscess should be sent for histological examination. A pathological report of granulomatous inflammation is beneficial in the early diagnosis of TPM, even if the AFB stain is negative (3).

The appearance of this type of lesion on MRI and the granulomatous inflammation in the forearm lesion of our patient led us to suspect a diagnosis of TPM, and this diagnosis was confirmed by mycobacteriological testing. In most cases receiving a timely diagnosis, effective drainage, and appropriate ATCT, the prognosis for a functional recovery is good. In order to avoid diagnostic delays, TPM should be kept in mind as a possible etiological factor when attempting to arrive at a differential diagnosis of cases of pyomyositis; this line of reasoning applies even in case involving immunocompetent persons and in the absence of any osseous involvement.

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