

## CASE REPORT

# TUBERCULOSIS OF THE CLAVICLE PRESENTING AS AN EXPANSILE LYTIC LESION: A CASE REPORT

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**An unusual case of skeletal tuberculosis, presenting as an expansile osteolytic lesion in the lateral end of the clavicle is presented. Diagnostic confusion delayed appropriate medical therapy, leading to development of a discharging sinus with secondary infection, which further confused the picture. With re-emergence of tuberculosis as an important infection worldwide, and the ability of this disease to mimic many skeletal pathologies, this has to be included in the differential diagnosis, especially at unusual sites.**

**Keywords :** tuberculosis ; osteolytic lesion ; clavicle.

**Mots-clés :** tuberculose ; lésion ostéolytique ; clavicule.

## INTRODUCTION

With the resurgence of tuberculosis as an important cause of osteoarticular infection worldwide, more and more cases are being reported with unusual features. The clavicle is an uncommon site of skeletal tuberculosis, reportedly accounting for less than one percent of cases (11), and the presentation at this site may frequently be atypical, leading to diagnostic confusion, and to delays in appropriate therapy. In the few reports available, the lateral end of the clavicle has been found to be less frequently diseased than the medial end (6, 8, 11). Additionally, tuberculosis has been known to mimic all types of lesions, and the absence of pulmonary lesions and other concomitant features, in addition to an atypical radiographic picture may not bring the diagnosis primarily to mind. A recent report highlighted a case of tuberculosis of the

medial end of the clavicle in a patient undergoing dialysis (2), which was thought to be a neoplasm. Our experience with an unusual case involving the lateral end of the clavicle prompted this report. Here too the radiologic characteristics mimicked those of a giant cell tumor and posed diagnostic difficulties.

## CASE REPORT

A 30-year-old female developed pain in her right shoulder after sustaining trivial trauma, when she fell out of bed. She got it massaged by an unqualified practitioner, subsequent to which she perceived crepitus and increased pain around the shoulder. A general practitioner requested shoulder xrays, which were interpreted by him as showing a fracture of the lateral end of clavicle, and she was treated in an arm pouch sling for two months. The pain in the shoulder persisted, and she developed a gradually progressive localized swelling over the right shoulder in the next few weeks. Radiographs were repeated at this stage ; they were interpreted by the radiologist as well as the treating physician as a giant cell tumor of the lateral end of clavicle,

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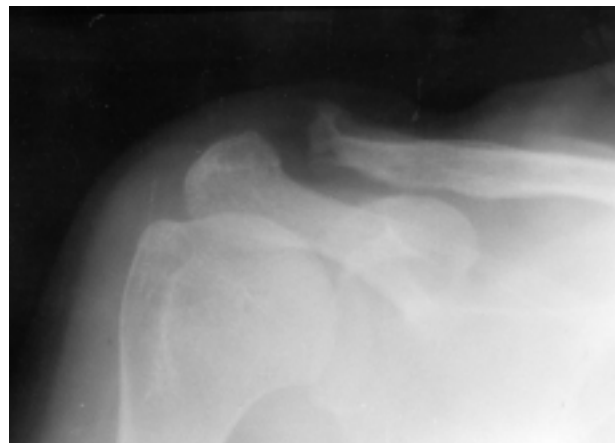
and she was advised to undergo surgical excision. However, a discharging sinus developed over the swelling before any surgical intervention was done ; she was prescribed broad-spectrum antibiotics, without bacterial culture. The sinus reportedly healed, but the discharge recurred after four months, and the patient was then referred to our tertiary care center for diagnostic evaluation and treatment.

Clinical examination of the shoulder revealed a soft tissue swelling (2.5 × 2.0 cms) and two discharging sinuses over the lateral end of the clavicle. There was no evidence of cervical or axillary lymphadenopathy, and the lung fields were essentially clear. The anteriorly placed sinus showed signs of inflammation, and pale granulation tissue was observed at its mouth.

Laboratory tests showed severe anemia (hemoglobin 7 g/dl), a total leucocyte count of 9000 cells/mm<sup>3</sup> with lymphocytosis (54%) and eosinophilia (8%). The ESR (Westergren) was 140 mm/hour. The discharge from the sinus revealed Gram - positive cocci on Gram staining ; the Ziehl Nielsen staining for acid fast bacilli was negative. Culture of the discharge revealed growth of *Staphylococcus aureus*. Anteroposterior radiographs of the right clavicle showed multiple, well-defined lytic lesions, with a soap bubble appearance, involving the lateral third of the clavicle in addition to a periosteal reaction along the superior border (fig. 1). There was acromioclavicular joint subluxation and soft tissue swelling on the superior aspect of the shoulder joint. A clinical diagnosis of chronic osteomyelitis with discharging sinuses, superimposed on a pre-existing giant cell tumor was entertained, and an incisional biopsy was done. The histopathologic report revealed evidence of chronic granulomatous inflammation suggestive of tuberculosis (fig. 2). Special staining and culture of the curettings for acid fast bacilli (AFB) were negative. The patient was started on a multidrug anti-tubercular therapy regimen (ATT) consisting of four drugs (rifampicin 450 mg/day, isoniazid 300mg/day, pyrazinamide 20-25 mg/kg/day and ethambutol 15-25 mg/kg/day) for three months. The sinuses healed within one month of starting ATT, and the swelling decreased in size. The



**Fig. 1.** — Xray photograph (anteroposterior view) of the clavicle, showing an expansile lesion with soap bubble appearance at the lateral end.



**Fig. 2.** — Xray photograph (anteroposterior view) of the same case 2 years after anti-TB therapy, showing evidence of healing.

patient was subsequently put on a maintenance therapy of two drugs (rifampicin, INH), that were continued for a period of 15 months.

Within four months, the patient recovered full range of shoulder movements, with complete resolution of the swelling. She was asymptomatic at two-year follow-up, except for occasional pain and minor discomfort during terminal abduction of the shoulder. Radiographs taken at this time revealed healing of the lesion with deformation of the lateral clavicular end, and widening of the acromioclavicular joint (fig. 3).

Table I. — Literature review –Tuberculosis of the clavicle

Series/year	Total number	Systemic TB	Lateral end	Age range ( in years)	Sex M:F	Side Rt:Lt	Radiologic Type	Surgical Treatment
Sorrel, 1932	One	*	One	*	Male	*	Spina ventosa	*
Sirkin, 1936	30	11	*	10-72	18:11*	11:12*	*	Excision/ Curettage-28 Conservative-2
Lafond, 1958	1/230	*	*	*	*	*	*	*
Jensen, 1959	5	2	3	9-73	3:2	4:1	Destructive-4 Spina ventosa-1	Excision
Tuli, 1969	7	*	One	*	*	*	Destructive	*
Srivastava, 1974	12	8	5	23.7 Average	1:2	6:4 Both-2	Destructive-4 Cystic rarefaction-3 Sequestrum-2 Irregular thickening-3 Proliferative-1	Curettage-7 ATT-5
Tucker, 1990	One	Nil	2	28	Female	Both	Destructive-Both	Curettage
Rasool, 1991	4	*	One	3-12	*	*	Irregular expansion, destruction & periosteal reaction	Curettage
Gerscovich, 1994	One	One	-	65	Female	Right	Lytic & sclerotic	Excision Biopsy
Fang, 1996	One	Nil	-	34	Female	Left	Apparent neoplasm	Resection
Present case	One	Nil	One	30	Female	Right	Soap bubble type Apparent neoplasm	Curettage

\* Details not available.

## DISCUSSION

Extrapulmonary tuberculosis presenting with involvement of the skeleton accounts for approximately 1 to 4% of cases. Most of these cases are in the spine, and involvement of the appendicular skeleton is unusual. With the problems of immunocompromised patients, a significantly older population, and the emergence of strains that are multidrug resistant, the problem of osteoarticular tuberculosis is becoming more prominent. The rarity of the problem, and the ability of tuberculosis to mimic other diseases, combined with a lack of awareness by the treating orthopedist, especially in communities where the disease is not routinely prevalent, often leads to diagnostic delays. This is unfortunate, as in many cases the disease starts as a bony focus, and with the passage of time there is

development of either a sinus or involvement of the adjacent joint. Both these stages have a prognosis that is worse than the purely osteomyelitic stage, in the form of either superimposed infections or decreased joint motion after healing.

Although the disease is known to mimic all types of osseous pathology, the problem is compounded when it presents at unusual sites. This frequently poses diagnostic difficulties (3), as the clinician may not keep the possibility of TB primarily in mind. Tubercular involvement of the clavicle is extremely rare, especially when it involves the lateral end (6, 11). A literature review revealed fewer than 20 cases of clavicular TB reported in the last 50 years, and only seven of these involved the lateral end (1-4, 6-11). Infection at this site has been reported to be more frequently of the destructive or proliferative type and may also be complicated by

a pathologic fracture (4, 6). Even the joint space reduction and the surrounding rarefaction, a feature typical of osteoarticular TB elsewhere in the body, is not so commonly observed at this site.

Diagnostic problems are confounded by the fact that this site is more prone to the development of neoplasms as compared to infections (1, 2). Fang *et al.* (2) reported a case of an apparent neoplasm of the medial end of the clavicle in a dialysis patient, which ultimately turned out to be tuberculosis. This again highlights the fact that in elderly patients, or those who are immunocompromised, or those from endemic areas, destructive swellings of bone have to be looked at suspiciously for the possibility of tubercular infection. Even in nonendemic areas, the problem may become significant, as large-scale population migrations have led to a mixing up of different ethnic groups.

Osteoarticular TB is almost always secondary to a primary focus elsewhere in the body (2, 7), and a definite attempt should be made to screen the pulmonary, GIT and renal systems for evidence of disease. The basis for an accurate diagnosis of TB is obtaining representative tissue from the focus or the isolation of *Mycobacterium tuberculosis* on culture. In our experience with such cases, as well as that of leading workers in this field (8, 11), there is a high incidence of false-negative culture reports. Osteoarticular tuberculosis is a paucibacillary disease in comparison to the number of bacilli seen in pulmonary lesions. This makes demonstration of AFB on staining or on culture from the skeletal lesions extremely difficult. Nevertheless a determined attempt at culture or staining for AFB is warranted. In most of the cases, however, the diagnosis has to be suspected by clinical features, concomitant pathology, histopathological evidence of granulomatous tissue, and a high index of suspicion (1, 2). Abdelwahab *et al.* (1) have even suggested that any confusing bone lesion in a non-white patient can potentially be caused by tuberculosis. The literature is also replete with reports about how tuberculosis of the skeleton can mimic any lesion. However a certain degree of complacency has crept into the minds of physicians worldwide about the decreasing incidence of this disease, and often TB is not considered in the differential

diagnosis of skeletal lesions. This is unfortunate, as diagnostic delays may lead to the development of sinuses, which may become secondarily infected, and will further confuse the picture. On the other hand, since TB is thought to be primarily a joint disease, purely bony involvement at an unusual site, with some kind of an expansile or destructive radiological picture will bring to mind the more commonly encountered tumors. The acromioclavicular joint does not show much joint space reduction, as the weight of the arm tends to distract it. In the present case too there was some joint subluxation, and the pathologic fracture, absence of systemic manifestations, and soap bubble appearance in a metaphyseal location on radiographs were certain peculiarities that confused the picture.

The treatment of skeletal TB is medical, and surgical intervention is needed only for the purpose of obtaining tissue for diagnosis. Once the appropriate anti-TB therapy is started, the symptoms resolve within 6 to 8 weeks, and most discharging sinuses heal. We recommend a four-drug regimen for a minimum period of 3-4 months, and once the clinical features settle, the patient should be maintained on a two-drug regimen for an additional 9 to 12 months. The therapy has to be prolonged even after there is a significant reduction of symptoms, to eliminate the persistent bacilli which are small populations of bacteria that lie dormant and are metabolically inactive in the initial phases.

In conclusion, we would like to emphasize the fact that a radiological picture of a soap bubble pattern in the lateral end of clavicle should include tuberculosis in its differential diagnosis. A high index of suspicion should be maintained in cases of gradually progressive swellings around the shoulder, even if they lack systemic features of tuberculosis. AFB culture is not essential for diagnosis, and once granulomatous tissue is obtained on histopathology, multidrug medical therapy for a prolonged period will cure the disease.

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### SAMENVATTING

*P. L. BASANAGOUDAR, P. N. GUPTA, R. BAHADUR, M. S. DHILLON. Tuberculosis van de distale clavicula voorkomend als een expansief lytisch letsel : een geval.*

Een ongewone tuberculeuze aantasting van de distale clavicula bij een 30 jarige vrouw wordt beschreven.

De diagnose was vertraagd door verwarrende klinische en radiografische gegevens, zodat uiteindelijk een gesur-infecteerde sinusdrainage ontstond met nog meer ver-warring tot gevolg. de diagnose werd uiteindelijk gesteld door biopsie. Behandeling met tuberculostatica in triple associatie was afdoende.

Gezien de actuele wereldwijde tuberculosis opflakke-ring moet bij skeletale afwijkingen ook deze mogelijkheid difentieel diagnostisch worden over-wogen.

### RÉSUMÉ

*P. L. BASANAGOUDAR, P. N. GUPTA, R. BAHADUR, M. S. DHILLON. Tuberculose de la clavicule se présen-tant comme une lésion lytique expansive : présentation d'un cas.*

Les auteurs présentent un cas inhabituel de tuberculose osseuse qui se présentait sous forme d'une lésion ostéolytique expansive au niveau de l'extrémité latérale de la clavicule. Une confusion diagnostique initiale a eu pour résultat l'instauration tardive du traitement médical approprié, ce qui a permis le développement d'une fis-tule productive avec surinfection, qui est venu compli-quer encore le tableau clinique. Dans le monde entier, la tuberculose apparaît à nouveau comme une infection avec laquelle il faut compter, et elle peut donner le change avec de nombreuses pathologies osseuses ; elle doit donc toujours être prise en considération dans le diagnostic différentiel, en particulier dans des localisa-tions inhabituelles.