## **CASE REPORT**

# MULTIPLE FRACTURES OF THE SYMPHYSIS PUBIS DUE TO TUBERCULOUS OSTEOMYELITIS

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#### INTRODUCTION

Osteoarticular tuberculosis, the most common site being the spine, accounts for approximately 2-3% of cases of tuberculosis (Moon *et al*, 1990). The diagnosis of tuberculosis of the symphysis pubis is difficult and rarely made. The differential diagnosis for pubic pain is broad and there are many difficulties in isolating and culturing *Mycobacterium tuberculosis* from this site. We present a case of tuberculous osteomyelitis of the pubic symphysis followed by a review of the literature.

#### CASE REPORT

A 74 year old African American woman with no significant past medical history presented with generalized weakness and progressive lower back pain for three months. She denied fever or history of trauma. Magnetic resonance imaging (MRI) of her lumbar spine showed multilevel degenerative changes, L2 and L3 compression fractures, and diffuse abnormal signal intensity in the sacrum and adjacent soft tissues which was considered to be from osteoporosis. Her tuberculin skin test was also positive. The erythrocyte sedimentation rate and C-reactive protein were 93 mm/hr and 26.5 mg/l, respectively. Those values prompted a rheumatologic work-up which revealed normal thyroid function tests, normal C3/C4 levels, negative extractable nuclear antigen antibodies, negative anti-SSA/SSB antibodies, an anti-nuclear antibody titer of 1:40 (speckled), and a rheumatoid factor of 17 (normal 0-15). She was started on treatment for osteoporosis and discharged from the hospital on calcium and alendronate. Eight months later, she was re-admitted with complaints of a painful, bead-sized right groin swelling for several months that had begun draining approximately two weeks prior to admission. The swelling was associated with progressive inability to walk. She denied fever or cough; however, she admitted to a gradual weight loss of 45 kg over three years. She had normal vital signs and was afebrile.

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Examination was notable for a 2 cm fistulous tract in the right inguinal region with mild fluctuance and purulent, sanguinous drainage. Laboratory examination included a white blood cell count of 5,970/µl, a hematocrit of 29%, and platelet count of 295,000/ mm<sup>3</sup>. Chest radiography demonstrated old granulomatous disease. A plain film of the pelvis demonstrated osteopenia, a wide symphysis pubis, soft tissue emphysema, and mixed sclerosis of the sacrum consistent with chronic osteomyelitis. Computed tomography (CT) of the pelvis (Fig 1) demonstrated extensive bony destruction and sclerosis of the symphysis pubis with a fistula to the right groin, consistent with osteomyelitis or cellulitis.

Empiric treatment with vancomycin and ceftriaxone was begun. A wound culture from the fistulous tract grew a few methicillin sensitive *Staphylococcus aureus*, a few diphtheroids, and a few group B Streptococci. A follow-up bone biopsy grew rare coagulase-negative *Staphylococcus*. A smear for acid-fast bacilli (AFB) was negative. The patient was discharged home on intravenous ertapenem for presumed bacterial osteomyelitis, calcium supplements for osteoporosis and daily oral isoniazid of 300 mg daily for treatment of latent TB.

Despite the intravenous ertapenem, the drainage and pain in the right groin persisted. The patient was admitted again the next month, after sustaining a right femoral neck fracture due to a fall that required a right hip arthroplasty. On examination, she had ongoing drainage from a 1.5 cm fistula in her right inguinal region. A CT of the pelvis was similar to the prior month's image and showed extensive bony destruction and a fistulous tract. She was again diagnosed with pubic symphysis osteomyelitis and antibiotic coverage was changed to nafcillin and cefepime since the patient had *S. aureus* bacteremia. During this admission, the AFB



Fig 1–CT scan of pelvis showing extensive bony destruction and sclerosis of the symphysis pubis with a fistula to the right groin, consistent with osteomyelitis or cellulitis.

culture (sensitivity 82-85%) from her previous admission returned positive for *Mycobacterium tuberculosis* and the patient was placed on four-drug therapy with isoniazid, rifampin, pyrazinamide, and ethambutol with the final diagnosis being TB osteomyelitis. At last follow-up, she was doing well with resolution of the fistulous tract and pelvic pain.

We performed a systematic review of cases of tuberculosis of the pubis reported from 1955 until February, 2007. We included both English and non-English articles and cases reported before 1966. The search was done with PUBMED, using the terms "Tuberculosis", "pubis", and/or "pubic symphysis" in the title or abstract, or as keywords. Then, we performed a hands-on review of the articles to evaluate which reported cases of tuberculous osteomyelitis of the pubis. The last search was performed in February 2007.

We identified 17 references with 33 cases reported in the literature (Table 1). From our

		Cases reporte	d with tuberculous o	osteomyelitis o	f symph	ıysis pubis.		
References	Age/ sex	Presenting complaint	Imaging	Treatment <sup>a</sup>	Outcome	Pathology and culture	Prior history of TB	Other sites of TB infection
Bayrakci et al, 2006 Ohkouchi et al, 2004	36/F 56/M	Abdominal & groin pain, limp Groin pain, fever	XR- erosion XR- erosion	Surgical, medical Medical, surgical	Healed Healed	Soft tissue cx- MTB Sputum cx- MTB	None None	None Lungs
Humer et al, 2001 Ramakrishnaiah et al, 2000	38/F 29/F	ытол раит, swening, што раит Draining sinus	Miki-erosion CT- pus in retropeirtoneum, erosion	meancal, surgical Medical	Healed	Soft tissue bx- granulation tissue	Lungs Lungs	lvone Sacro-iliac joint
Benbouazza et al, 1997	21/F	Groin pain, limp, weight loss, inguinal lymphadenopathy	XR-erosion and sclerosis	Medical	Healed	LN bx- giant follicles	None	Lung
Benbouazza et al, 1997	19/F	Groin pain, weight loss	XR- erosion, bone scan- fixation of pubis	Medical	Healed	LN bx- giant follicles	None	Cervical LN
Moujtahid et al, 1995	34/F	Groin pain & swelling, fever, weight loss	XR- erosion	Surgical, medical	Healed	Drainage cx- AFB	None	Rib, humerus, ilium
Tsay et al, 1995	57/F	Limp, fever	CT- erosion	Medical, surgical	Healed	Soft tissue bx- caseous necrosis	None	Elbow
Manzaneque et al, 1992	79/F	Groin swelling	CT-cystic mass in pelvis, erosion	Surgical	Healed	Soft tissue bx- MTB	None	None
Khazov et al, 1992	62/F	Groin swelling	XR- erosion	Medical			Lungs	None
Rozadilla et al, 1991	58/M	Groin pain	XR-erosion	Medical	Healed	Soft tissue bx- MTB	Spine	Lungs
Moon et al, 1990	5/M	Limp	XR-erosion	Surgical, medical	Healed	Soft tissue bx-MTB	None	None
Moon et al, 1990	8/M	Groin swelling, limp	XR- erosion	Surgical, medical	Healed	Soft tissue bx-MTB	None	None
Moon et al, 1990	26/F	Groin pain	XR-erosion	Surgical, medical	Healed		Peritoneum	None
Moon et al, 1990	5/M		XR- erosion	Surgical, medical	Healed		None	None
Moon et al, 1990	8/M		XR- erosion	Surgical, medical	Healed		None	None
Bronner et al, 1990	70/F	Groin pain	XR-erosion, increased symphysis width	Medical	Died	Drainage - MTB	None	None
Bronner et al, 1990	56/M	Groin pain, fever		Medical	Healed	Soft tissue cx- MTB	Lungs, elbow	None
Ker. 1986	41/F	Multiple groin sinuses	XR- erosion & sequestrae	Surgical, medical	Healed	Soft tissue cx- MTB	None	None
Vogelzang et al, 1983	56/M	Groin swelling	CT- large abscess, erosion	Surgical	Healed	Bone bx-MTB	None	Lungs
Ellis, 1974	14/M	1	XR- erosion	Surgical, medical	Died	Soft tissue bx- giant cells	None	Spine, LN
Nicholson, 1958	7/M	Groin pain, limp	XR-erosion	Medical	Persistent	Inguinal node bx -MTB	None	None
Nicholson, 1958	19/M	Thigh swelling	XR- erosion	Aspiration, surgical	Healed	Drainage- MTB	Lungs	None
Nicholson, 1958	18/M	Groin pain, thigh swelling	XR- erosion	Surgical	Persistent	Drainage- MTB	None	None
Nicholson, 1958	22/M	Groin pain & swelling,	XR-erosion, sequestrum	Surgical	Persistent		Lungs, elbow	None
		draining sinus						
Nicholson, 1958	26/M	Groin pain	XR-erosion	Surgical, medical	Persistent	Drainage- MTB	None	None
Nicholson, 1958	24/M	Groin pain & swelling, limp	XR- erosion	Surgical	healed	Drainage- MTB	None	None
Nicholson, 1958	29/F	Limp, groin pain & swelling	XR-erosion with sequestrum	Surgical, medical	healed	Drainage- tuberculous	None	None
Nicholson, 1958	30/M	Groin swelling, draining sinus	XR- erosion with sclerosis	Surgical	Persistent	Drainage- tuberculous	TB peritonitis	None
Nicholson, 1958	36/F	Painless groin swelling	XR- erosion with sequestra	Surgical, Medical	healed	Drainage- tuberculous	None	Lungs
Nicholson, 1958	68/F	Groin pain & swelling, limp	XR- erosion with sequestra	Medical , surgical	Persistent	Drainage- MTB	None	None
Nicholson, 1958	55/M	Groin pain, groin swelling	XR-erosion with sequestra	Surgical	Persistent	Drainage- tuberculous	None	None
Bevan, 1955	23/F	Groin swelling	XR- erosion	Surgical, medical	healed	Drainage cx- MTB	None	None

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<sup>a</sup>Medical treatment: Antituberculous drug therapy XR, x-ray of pelvis; MTB, Mycobacterium tuberculosis; AFB, Acid-fast bacilli; cx, culture; bx, biopsy; LN, lymph nodes

review, we found the disease occurs mostly in younger individuals (39% between 16 to 30 years old, 15% in those less than 10 years old). There was no gender difference with a male to female ratio of 1.06:1. No age was found to be immune. Commonly reported symptoms included: groin swelling (48%), groin pain (48%), a limp (24%) and a draining sinus (15%). Groin swelling generally indicated abscess formation in the groin.

Thirty percent of cases had a previous history of tuberculosis. Only 30.1% of cases had concurrent tuberculosis at other sites. and of these, half had pulmonary disease. For most patients, osteomyelitis of the pubis was the only site of tuberculous infection. On imaging, most had osteoarticular erosion of the pubis. CT did not seem to have any benefit over traditional radiography for diagnosis, but made delineation of the abscess easier. Erythrocyte sedimentation rate was elevated in ten of twelve cases and tuberculin skin test was positive in four of five. Thirty-three percent of cases were diagnosed by culture of aspirated pus and 27% by culture of biopsy tissue. Twenty-one percent of reported cases were diagnosed on the basis of granulomas on biopsy of either the lesion or lymph nods without culture verification.

Twenty-four percent of patients received anti-tuberculous drug therapy without surgical intervention. Twenty-four percent of patients received only surgical treatment. The rest were managed with a combination of medical and surgical therapies. Medical treatment was generally continued over a period of 6-12 months. Surgical treatment generally consisted of debridement or bone grafting. In most patients (73%), the lesion healed regardless of treatment modality. In 21% of patients, the lesion developed into a draining sinus which persisted for a few months despite treatment. Most of these patients received only surgical management. In one case, the abscess recurred within three months but eventually healed with medical and surgical management.

## DISCUSSION

Tuberculosis of the pubis is rare; only two cases have been reported in the Englishlanguage literature in the past decade (Hunter and Sim, 2000; Bayrakci *et al*, 2006). To our knowledge our patient is the first reported case in the United States of tuberculous osteomyelitis of the pubis with extensive fractures without coexisting active TB disease at another site.

Because of the indolent nature of the disease, patients often present with chronic symptoms ranging from two weeks to one year. It is thought that osteoarticular tuberculosis is a result of hematogenous or lymphatic spread from a reactivated latent focus, usually pulmonary; however, evidence of previous infection is not always encountered.

Delays in the diagnosis of tuberculous osteomyelitis of the pubis are common. Our review, highlights several clues that should raise the clinician's index of suspicion. These include chronic pain with groin swelling with or without a draining sinus, progressive difficulty walking, and a current or previous history of disease compatible with TB. Diagnosis is best made by biopsy and culture of the affected site. In the proper clinical context with pathological evidence of granulomatous disease, one need not wait for culture verification to initiate treatment.

Wide surgical excision in combination with antituberculous chemotherapy has been suggested as the best course of treatment (Moon *et al*, 1990). However, depending on the clinical context, surgical or chemotherapeutic regimens alone may suffice. Our review indicates most patients do well with treatment.

In conclusion, osteomyelitis of the pubis is a rare presentation of tuberculous infection.

Its indolent nature and difficulties in culturing the organism have made this process a difficult one to diagnose and complications can ensue as exemplified by our patient. Our case highlights the importance of early tissue specimen culturing in all patients presenting with groin pain and swelling. Treatment should be started in patients with a high degree of suspicion without waiting for final cultures to prevent complications.

### REFERENCES

- Bayrakci K, Daglar B, Tasbas BA, Agar M, Gunel U. Tuberculosis osteomyelitis of symphysis pubis. *Orthopedics* 2006; 29: 948-50.
- Benbouazza K, Allali F, Bezza A, *et al.* Pubic tuberculous osteo-arthritis. Apropos of 2 cases. *Rev Chir Orthop Reparatrice Appar Mot* 1997; 83: 670-2.
- Bevan PG. Tuberculosis of pubis presenting as a cold abscess of the thigh. *Br Med J* 1955; 2(4943): 832-3.
- Bronner U, Lindquist L, Svanbom M. Tuberculous infection of the symphysis. *Lakartidningen* 1990; 87: 388-9.
- Ellis W. Multiple bone lesions caused by Avian-Battey mycobacteria. Report of a case. *J Bone Joint Surg Br* 1974; 56: 323-6.
- Hunter DC, Sim D. Pelvic tuberculosis: a case of delayed diagnosis. *Acta Obstetr Gynecol Scand* 2001; 80: 281-2.
- Ker NB. Tuberculosis of the pubic symphysis. *J R Soc Med* 1986; 79: 429-30.
- Khazov PD. Congestive abscess masking a soft tissue tumor. *Problemy Tuberkuleza* 1992; (5-6): 55-6.

- Manzaneque L, Marin I, Garcia-Bragado F, Beiztegui A, Dastis C, Sanchez-Matas P. Osteoarticular tuberculosis of the symphysis pubis presenting as a hypogastric cystic mass in a woman with primary Sjogren's syndrome. *Br J Rheumatol* 1992; 31: 495-6.
- Moon MS, Ok IY, Ha KY, Sihn JC. Tuberculosis of the ischiopubic ramus. A report of five cases. *Int Orthop* 1990; 14: 175-7.
- Moujtahid M, Essadki B, Lamine A, Bennouna D, Zryouil B. Multifocal bone tuberculosis: apropos of a case. *Rev Chir Orthop Reparatrice Appar Mot* 1995; 81: 553-6.
- Nicholson OR. Tuberculosis of the pubis; report of eleven cases. *J Bone Joint Surg Br* 1958; 40-B: 6-15.
- Ohkouchi M, Inase N, Yasui M, Miura H. Case of pubic tuberculous osteomyelitis and pericarditis during anti-tuberculosis chemotherapy. *Kekkaku* 2004; 79: 531-5.
- Ramakrishnaiah VP, Jain V, Choon AT, Rao BH. Retroperitoneal cold abscess with tuberculosis of sacro-iliac joint and pubic bone: a case report of unusual presentation of osteoarticular tuberculosis. *J Indian Med Assoc* 2000; 98: 128-9.
- Rozadilla A, Nolla JM, Rodriguez J, Del Blanco J, Roig Escofet D. Tuberculosis of the pubis symphysis. *J Rheumatol* 1991; 18:1271-2.
- Tsay MH, Chen MC, Jaung GY, Pang KK, Chen BF. Atypical skeletal tuberculosis mimicking tumor metastases: report of a case. J Formos Med Assoc1995; 94: 428-31.
- Vogelzang RL, Hendrix RW, Neiman HL. Computed tomography of tuberculous osteomyelitis of the pubis. *J Comput Assist Tomogr* 1983; 7: 914-5.