ORIGINAL ARTICLE

Unusual locations of osteoarticular tuberculosis in children: A report of 12 cases

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Accepted: 15 October 2012

KEYWORDS
Osteoarticular tuberculosis; Unusual location; Child

Summary
Introduction: Unusual locations of osteoarticular tuberculosis (OA-TB) raise diagnostic issues due to their atypical and non-suggestive clinical and radiological presentation.
Objectives: The present retrospective study analyzed the various clinical, radiological and therapeutic aspects.
Patients and methods: A retrospective series included 12 children (mean age, 7 years 4 months; sex-ratio, 0.7), treated in our department between 1980 and 2010. Knee, hip and spine locations were excluded.
Results: Mean time to diagnosis was 32 months. Active TB infection was identified in 42% of cases. Pain was the presenting symptom in 83% of cases, with a preponderance of osteitis. Bone loss was the main radiological sign. Phemister’s triad was found in two cases of combined articular and bone infection. Diagnosis was confirmed on histology in 92% of cases. All patients were managed according to the Moroccan national TB protocol. Surgery was indicated in five cases, comprising abscess drainage with or without bone surgery (notably for joint dislocation). Four patients showed orthopedic sequelae, including two with associated spinal locations.
Discussion: Rare osteoarticular tuberculosis locations often cause diagnostic problems. Any chronic clinical presentation or suspected atypical bone lesion should suggest a diagnosis of osteoarticular tuberculosis.
Level of evidence: Level IV. Retrospective study.
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Introduction

Osteoarticular tuberculosis (OA-TB) is defined by a set of pathological signs secondary to involvement of locomotor osteoarticular structures by Koch’s bacillus [1]. It represents 3 to 5% of TB cases and about 15% of extrapulmonary cases. It may involve any bone segment [2,3], but the most common
locations are those of Pott’s disease (spine) or TB of the hip or knee. There have been few studies of infrequent atypical locations in children. The present study analyzed the clinical, radiological and therapeutic aspects of rare locations, which raise many diagnostic problems.

Patients and methods

A retrospective study included 12 unusual OA-TB locations observed over a 30-year period from 1980 to 2010. Usual locations were excluded: tuberculous spondylitis or Pott’s disease, and TB of the hip (coxalgia) or knee. TB was diagnosed on histology or by presumption: history of TB, TB in another location, impaired general health status, fever and night sweats. For each patient, history, clinical, biological and radiological signs and diagnostic factors were recorded, as well as treatment and evolution data (Table 1).

Results

The series comprised five boys (42%) and seven girls (58%). Mean age was 7 years 4 months (range, 10 months to 14 years). Only one patient had history of TB. TB infection was found in five patients (42%). Two children (16%) had not had their BCG vaccination; the others had all been vaccinated at birth. Two patients had traumatic osteitis. Mean time to consultation was 32 months (range, 2 months to 5 years). Ten patients (83%) complained of pain at first consultation. Six (50%) showed functional impotence. General signs were always accorded secondary importance. Nine patients (75%) showed local limb tumefaction. Three (25%: patients 1, 4 and 11) already presented with fistula at diagnosis. A second location was found in three cases: lumbar spine (patient 6), dorsal spine (patient 7) and skin (patient 1). Patients 3 and 5 (17%) had osteoarthritis, whereas the other 10 (83%) had tuberculous osteitis without joint involvement. Both cases of OA-TB concerned the knee. Osteitis locations were calcaneus (two case), ulna (two cases), iliac bone (two cases), femoral neck (one case), lateral cuneiform (one case), greater trochanter (one case), radius (one case), talus (one case), tibia (one case) and 4th rib (one case). In OA-TB, radiologic signs comprised joint-line impegement, and irregular edges and subchondral cysts (Fig. 1), whereas in osteitis the radiological images systematically showed bone loss (Fig. 2). CT, performed in three cases, gave a better view of the bone lesions seen on X-ray (Fig. 3). One patient had an MRI of the forearm, showing radial diaphysis bone loss (Fig. 4). No patients underwent bone scintigraphy. Nine patients showed elevated sedimentation rate (10–100 mm at hour 1). Total blood count was performed in 11 patients and found hyperleukocytosis in two cases (patients 4 and 6) and moderate anemia in four (patients 2, 3, 4 and 6). Tuberculin intradermal reaction was performed in 10 patients and was positive in six and negative in four. Diagnosis was confirmed on histology of the joint synovial membrane, harvested bone or abscess wall, performed in 11 cases and systematically finding epithelioid-giant-cell granuloma; in the remaining case (of cutaneous TB with foot tumefaction, fistulas and lateral cuneiform bone-loss on plain X-ray), diagnosis was founded on history-taking, clinical and radiological findings.

Figure 1 Lateral right-ankle X-ray in a 9-year-old girl (patient 3), showing talocrural joint-line impingement with subchondral osteocondensation.

All patients were managed according to the Moroccan national TB protocol, which has been regularly updated, with the most recent version dating from April 2011. For OA-TB, the protocol comprises four anti-TB agents (isoniazid, rifampicin, pyrazinamide and streptomycin) for a 9-month course. In five cases, surgery was associated: five abscess drainages, with one associated hip dislocation reduction (patient 2, with TB of the femoral neck).

All patients were followed up at least once; two were lost to follow-up after 3 and 5 months’ treatment respectively. Mean follow-up was 37.5 months (range, 3 months to
### Table 1  The 12 cases.

<table>
<thead>
<tr>
<th>Age</th>
<th>Sex</th>
<th>Location</th>
<th>TB infection</th>
<th>BCG vaccination</th>
<th>Trauma</th>
<th>General signs</th>
<th>Pain</th>
<th>Tumefaction</th>
<th>Fistulas</th>
<th>Other locations</th>
<th>Time to diagnosis</th>
<th>IDR (mm)</th>
<th>SR at hour 1</th>
<th>Plain X-rays</th>
<th>CT/MRI</th>
<th>EGCG on biopsy</th>
<th>Antibiotics</th>
<th>Surgery</th>
<th>Evolution</th>
<th>FU</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>6 yrs</td>
<td>M</td>
<td>Left lateral cuneiform</td>
<td>+</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>Skin</td>
<td>1 yr</td>
<td>18</td>
<td>30</td>
<td>Cyst</td>
<td>Not performed</td>
<td>Skin biopsy</td>
<td>+</td>
<td>+</td>
<td>Favorable</td>
<td>1 yr</td>
</tr>
<tr>
<td>2</td>
<td>6 yrs</td>
<td>M</td>
<td>Left femoral neck</td>
<td>+</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>+</td>
<td>+</td>
<td>—</td>
<td>3 months</td>
<td>18</td>
<td>75</td>
<td>Osteolytic sequencers</td>
<td>Joint-line impingement, irregular edges</td>
<td>Osteolysis</td>
<td>&quot;&quot;</td>
<td>+</td>
<td>+</td>
<td>Favorable</td>
</tr>
<tr>
<td>3</td>
<td>9 yrs</td>
<td>F</td>
<td>Talocrural joint</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>5 yrs</td>
<td>15</td>
<td>55</td>
<td>—</td>
<td>—</td>
<td>Favorable</td>
<td>Ankle ankylosis</td>
<td>8 months</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>13 yrs</td>
<td>F</td>
<td>2nd ray</td>
<td>Left ulna Right calcaneus</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>3 months</td>
<td>15</td>
<td>88</td>
<td>Skin bistuity</td>
<td>Skin lesion</td>
<td>+</td>
<td>+</td>
<td>Favorable</td>
<td>1 yr</td>
<td></td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>8 yrs</td>
<td>F</td>
<td>Left talonavicular joint</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>1 yr</td>
<td>—</td>
<td>Normal</td>
<td>Irregular edges + subchondral lysis</td>
<td>Bone loss + subcapital cartilage damage</td>
<td>Osteolysis</td>
<td>&quot;&quot;</td>
<td>+</td>
<td>+</td>
<td>Calcaneo-valgus foot</td>
<td>1 yr</td>
</tr>
<tr>
<td>6</td>
<td>6 yrs</td>
<td>F</td>
<td>Ulna Calcaneus</td>
<td>+</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>L2-L4 spine</td>
<td>5 months</td>
<td>—</td>
<td>100</td>
<td>Bone loss + destruction of left L2 pedicle</td>
<td>Lumbar stiffness</td>
<td>3 months</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>14 yrs</td>
<td>M</td>
<td>Left 4th rib</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>D9-D10 spine</td>
<td>1 yr</td>
<td>Not performed</td>
<td>10</td>
<td>Bone loss + subcapital cartilage damage</td>
<td>Dorsal kyphosis</td>
<td>3 yrs</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>4 yrs</td>
<td>M</td>
<td>Right greater trochanter</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>+</td>
<td>+</td>
<td>—</td>
<td>5 months</td>
<td>11</td>
<td>Normal</td>
<td>—</td>
<td>—</td>
<td>Favorable</td>
<td>6 yrs</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>9</td>
<td>11 yrs</td>
<td>F</td>
<td>Sup. extremity of left radius</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>3 months</td>
<td>17</td>
<td>69</td>
<td>Bone loss</td>
<td>NRB: radial diaphysis lysis</td>
<td>+</td>
<td>+</td>
<td>Favorable</td>
<td>3 yrs</td>
<td></td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>9 yrs</td>
<td>F</td>
<td>Iliac wing</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>2 months</td>
<td>—</td>
<td>50</td>
<td>Osteolysis ilium + acetabulum</td>
<td>CT: iliac wing + acetabulum + soft tissue collection</td>
<td>Osteolysis</td>
<td>&quot;&quot;</td>
<td>+</td>
<td>+</td>
<td>Favorable</td>
<td>5 yrs</td>
</tr>
<tr>
<td>11</td>
<td>6 yrs</td>
<td>M</td>
<td>Talus</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>6 months</td>
<td>—</td>
<td>28</td>
<td>Talus lysis</td>
<td>CT: Talus lysis</td>
<td>+</td>
<td>+</td>
<td>Good clinical evolution</td>
<td>6 months</td>
<td></td>
<td></td>
</tr>
<tr>
<td>12</td>
<td>10 months</td>
<td>M</td>
<td>Left acetabulum, inf. femoral and sup. and inf. tibial extremities</td>
<td>+</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>2 months</td>
<td>Not performed</td>
<td>6</td>
<td>Osteolysis acetabulum + sup. and inf. tibial extremities</td>
<td>CT: Osteolysis at 4 sites</td>
<td>Osteolysis at 4 sites</td>
<td>+</td>
<td>+</td>
<td>Good clinical evolution</td>
<td>5 months</td>
<td></td>
</tr>
</tbody>
</table>

EGCG: epithelioid-giant-cell granuloma; IDR: Intradermal reaction; TB: tuberculosis; M: male; F: female.
Peripheral osteoarticular tuberculosis constitutes nearly 1 to 5% of cases of TB, taking all locations together. Unusual locations other than the spine (Pott’s disease), knee or hip have been little studied and raise problems of differential diagnosis, notably with granulomatous and metastatic pathologies. Risk factors for OA-TB include trauma activating a pre-existing osteoarticular disorder [4,5], and economic insecurity and low socio-economic level inducing immunodepression which increases the risk of OA-TB even in vaccinated subjects [6]. All of the present patients, in fact, were from socially disadvantaged backgrounds, and showed a 42% rate of TB infection. OA-TB is rare in developed countries, thanks to generalized BCG vaccination and, above all, improved living standards. The absence of BCG vaccination in 16% ($n = 2$) of the present cases is to be explained by the fact that both were longstanding cases, dating back 30 years to the very beginnings of the national vaccination program. Trauma was found in 16% of cases. Koch’s bacillus can reach the joint and bone by extension from an osteitis site or hematogenically from another, usually pulmonary, site [7].

Positive diagnosis of atypical and unusual forms of OA-TB is difficult, often requiring a range of clinical, biological and radiological findings. Symptomatology is typically chronic and insidious. Clinical signs such as pain, functional impotence, fever and night sweats are systematically reported [8]. In the present series, the typical insidious onset with pain and functional impotence was found in 83% of the children. General signs are often put in the background by parents. Few studies have reported series of unusual OA-TB locations. Teklali [1] reported rare locations: elbow (10 cases) ankle (10 cases), shoulder (one case), wrist (one case), skull, trochanter and ribs, with some double (three cases) or even multiple involvement (five cases). Unlike in the present series, most cases were of osteoarthritis. Plain X-ray is very useful for diagnosis, but may be normal at early stages [9]. OA-TB presents a Plemister triad, regardless of location, associating osteoporosis next to the joint, bone erosion at the peripheral site and progressive joint-line impingement; the radiological aspect, however varies according to stage at diagnosis [10]. The radiological aspects of tuberculous osteitis vary, although some may be highly suggestive; the most frequent is of a clearly or sometimes irregularly contoured osteolytic lesion, sometimes surrounded by a thin area of osteocondensation [10,11] (Fig. 2). In forms involving the periphery of the bone, there is early destruction of cortical bone, so that infection and bone debris spread to adjacent soft tissue with development of an abscess [10]. In long bones, periosteal reaction is possible. CT provides detailed analysis of the joint line and adjacent bone and visualization of microcalcifications within abscesses highly suggestive of tuberculous etiology [12]. In OA-TB, CT is useful in certain locations: sacro-iliac, sternoclavicular and pubic symphysis. In tuberculous osteitis, it is useful in case of involvement of flat bones such as the pelvis, ribs or sternum [13]. MRI is the optimal imaging examination for diagnosis and follow-up of tuberculous osteitis; it may reveal synovial pannus, joint effusion, cartilage destruction, bone erosion, bone fragments, peri-articular abscess, peri-articular inflammation and bone edema [14]. Technetium bone scintigraphy allows exploration for other clinically silent OA-TB locations [15]. Biologically, inflammation and leukocyte assessment is often normal. Intraepidermal reaction is usually positive, but when negative does not rule out diagnosis: it was positive in 69% of Teklali’s cases [1] and 87% of Hsing’s [16]. Diagnosis is confirmed by Koch’s bacillus identified on the various samples or on histology of synovial or bone biopsy. In the present series, diagnosis was provided by anatomopathology in 92% of cases (11 biopsies).

Treatment is primarily and essentially medical, with surgery reserved for certain situations or complications. Early antituberculosis therapy is very effective, providing complete...
ostearticular recovery, ensured by associating several anti-TB agents. In most reports, associated surgery was considered mandatory [7,16], and was usually early, with a two-fold objective: backing up antibiotherapy in controlling the tuberculous infection to conserve joint function, and to achieve bone stability. Indications for early surgery are: evolved osteoarthritis, bone sequestration, presence and size of soft-tissue abscesses, joint dislocation and non-response to anti-TB antibiotherapy over the first 3 to 6 months [2].

In the present series, abscess evacuation and sequestrectomy were the main indications; pathologic joint dislocation reduction concerned only one patient.

“Late” surgery has a functional objective of reconstruction or stabilization, with a choice between arthrodesis and implant. Rest is essential during the initial phase, to conserve joint function and enable rapid resolution of pain: for a few weeks in OA-TB, and at least 3 months in tuberculous osteitis [17].

Conclusion

Rare and unusual OA-TB locations often pose a problem of differential diagnosis, notably with granulomatous and metastatic disease, and especially when locations are multiple. All chronic clinical presentations or suspect bone lesions of atypical aspect should suggest a diagnosis of OA-TB. This allows early management of the pathology, limiting morbidity.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References