Acta Orthop. Belg., 2011, **77**, 294-298

Primary subacute osteomyelitis of the talus in children A case series and review

Ravinder PABLA, Saket TIBREWAL, Manoj RAMACHANDRAN, Matthew BARRY

From the Department of Trauma & Orthopaedics, Royal London Hospital, London

Subacute haematogenous osteomyelitis of the talus in children is a rare condition. All previously reported cases have been managed by hospital admission with surgical debridement and antibiotics or by intravenous antibiotic therapy followed by oral antibiotics. This case series documents the management of the condition at our institution and reviews the current published literature. We conclude that with appropriate patient selection, primary subacute haematogenous osteomyelitis of the paediatric talus can be managed on an out-patient basis with oral antibiotic therapy.

Keywords : osteomyelitis ; subacute ; talus ; children.

INTRODUCTION

Subacute osteomyelitis is a haematogenous infection of the bone characterised by an insidious course with local symptoms and few clinical signs. In its primary form, it is a distinct clinical entity defined by the absence of an initial acute osteomyelitis or preceding systemic antibiotic therapy.

We report three cases of primary subacute osteomyelitis of the talus in children and review our management of these cases. We wish to emphasise the need for awareness, early diagnosis and implementation of appropriate treatment of the condition in order to avoid potentially unnecessary morbidity. In particular, we highlight that with careful case selection, one can avoid the need for hospital admission, prolonged intravenous antibiotic therapy or surgery. The use of oral antibiotics alone as a treatment modality has not previously been reported in the literature.

CASE SERIES PATIENTS

Case 1

A 2-year old girl (LW) presented with a six week history of limping, intermittent swelling to the right ankle and low grade pyrexia. There was no history of trauma. She had been systemically well throughout. On examination she walked with a mild limp. There was no localised bony tenderness on palpation. She had a good, pain-free range of movement in her ankle and sub-talar joints.

Inflammatory markers and blood cultures were normal. Initial ultrasound scanning (USS) revealed

■ Ravinder Pabla, Core Surgical Trainee.
■ Saket Tibrewal, Specialist Registrar.
■ Manoj Ramachandran, Consultant.
■ Matthew Barry, Consultant.
Department of Trauma & Orthopaedics, Royal London Hospital, London, U.K.
Correspondence : Manoj Ramachandran, Department of Trauma and Orthopaedics, Royal London Hospital, Whitechapel, London E1 1BB, U.K.
E-mail : manoj.ramachandran@bartsandthelondon.nhs.uk
© 2011, Acta Orthopædica Belgica.

No benefits or funds were received in support of this study

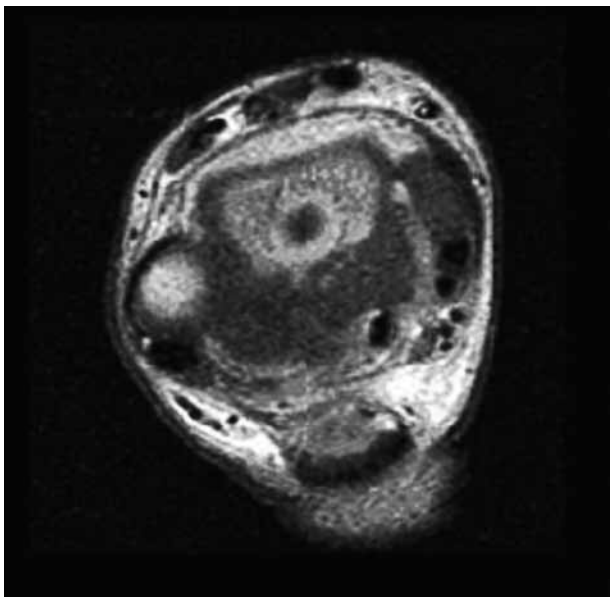

Fig. 1. — Penumbra sign of primary subacute osteomyelitis on T1-weighted axial MRI images of case 1 (patient LW) at diagnosis.

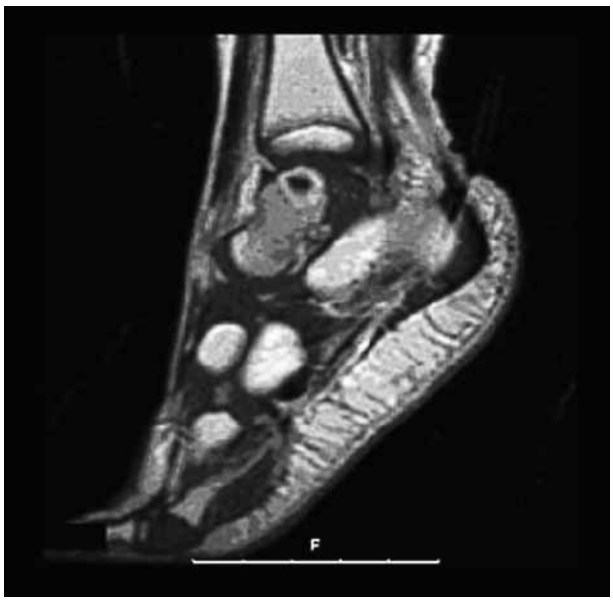

Fig. 2. — Penumbra sign of primary subacute osteomyelitis on T1-weighted sagittal MRI images of case 1 (patient LW) at diagnosis.

an ankle joint effusion but no apparent disruption of the talus. Radiographs showed a progressive lytic lesion in the posterior aspect of the talus. Magnetic resonance imaging (MRI), pre- and post-contrast, demonstrated a characteristic penumbra sign, showed talar collapse, a small collection and an associated ankle joint effusion (Figs. 1-2).

At surgical exploration of the talus, there was a bony cavity but without pus. Specimens were sent for microscopy, culture and sensitivity but yielded no positive results. The ankle was immobilised in plaster for four weeks. The girl was allowed to weight bear as tolerated. Oral flucloxacillin was continued for a total of eight weeks.

Six months after surgery, LW was systemically well with no palpable tenderness or swelling. Ankle and sub-talar joint movement was normal and pain free. Repeat radiographs at this time showed progressive healing with residual sclerosis and lucency of the talus. At 30 months, LW complains of occasional ache and mild scar tenderness but has a full range of ankle movement and maintains a normal, unrestricted level of activities.

Case 2

A 15-month old boy (NM) presented to our Emergency Department with a short history of a painful swelling to his left ankle. There had been a possible twisting injury to his left ankle three months earlier but no acute bony injury was noted at that time. He was systemically well. His left foot was noted to be held in an inverted position. The ankle was swollen and all movements were restricted by pain.

White cell count (WCC) was normal but the C-reactive protein (CRP) was moderately raised at 66 mg/L. Radiographs revealed a lytic area in the posterior aspect of the talus. USS confirmed an ankle joint effusion with possible synovial thickening. MRI confirmed these findings. Admission to hospital with administration of intravenous benzylpenicillin and flucloxacillin resulted in improvement of his clinical symptoms and inflammatory markers. CRP at discharge was less than 5 mg/L. Antibiotics were continued orally for a total course length of eight weeks.

At clinical review five months later his symptoms had settled. Inflammatory markers were normal and he was weight bearing. He had a full range of pain-free movement in his ankle. Radiographically, there was evidence of resolution of the lytic area within the talus. At 28 months, NM remains asymptomatic with no evidence of complications.

Case 3

A 2-year old girl (MC) was referred by an Emergency Department to our fracture clinic with a four week history of a limp associated with pain and possible swelling of the left foot. There was no history of trauma and the child was well with no local or systemic symptoms. On examination she walked with a limp and tended to invert the foot. Hip and knee examination were normal. There was no swelling or bony tenderness of the ankle or foot. The left hindfoot was perhaps slightly warmer than the right hindfoot. She had normal ankle, sub-talar and mid-foot movements bilaterally.

Radiographs showed a lytic/sclerotic lesion in the posterior aspect of the left talus. Serological analysis revealed a normal WCC and CRP but the erythrocyte sedimentation rate (ESR) was mildly raised at 28 mm/hr.

Oral flucloxacillin and penicillin V were prescribed for a total of seven weeks. Symptomatic relief was reported within a couple of weeks of commencing antibiotic therapy. Repeat radiography seven months after initial presentation showed no progression of bony change. At two year follow-up MC continues to be asymptomatic and without complication from her non-operatively managed primary subacute talar osteomyelitis.

DISCUSSION

Haematogenous osteomyelitis in children is well recognised. A decline in incidence had been noted over previous decades (*3,6*). More recently, however, the overall incidence of acute and subacute forms has started to rise. Increasing numbers of cases of the acute disease have been related to the emergence of methicillin-resistant Staphylococcus aureus (*2,10*). Rates of subacute osteomyelitis have not changed significantly, although the trend has been upwards from 1 to 2 per 100,000 population per year from 1991 to 1997 (*3*).

The distinction between acute and subacute osteomyelitis is arbitrary with infection diagnosed as subacute if presentation was two weeks or more after onset of symptoms (*1,6,15*). The primary form of this condition is characterised by the absence of an initial acute osteomyelitis, systemic illness, directly preceding trauma or recent course of antibiotic therapy.

Primary subacute osteomyelitis of the paediatric talus is rare. Subacute osteomyelitis of the paediatric talus is rare. Our search of the published literature yielded only 19 explicit cases (*1,9,19-20,22*).

Like all subacute osteomyelitis, it is characterised by an insidious course with local symptoms and few clinical signs. It can therefore be difficult to diagnose and delayed diagnoses result in delayed initiation of appropriate treatment. Mean diagnostic delays of up to 158.5 days have been reported (*12*). Clinical features and laboratory investigations are usually non-specific in nature. WCC, CRP and ESR are often within normal limits or, as in our cases, only mildly raised (*14,17,20-22*). Radiographic features are present at diagnosis and plain radiographic features have been classified into six types (*16,18*) (Fig. 3).

MRI is used as a second line investigation and demonstrates a characteristic, though not pathognomonic, penumbra sign (*21*). The penumbra sign is the picture of a transitional zone of high intensity between areas of radiodense sclerosis and radiolucent abscess on T1-weighted MRI scans (Figs. 1-2). It correlated to a layer of vascularised granulation tissue surrounding the abscess (*21*). Bone scintigraphy is usually positive with areas of high uptake at the affected site or sites (*9,12,19*).

The majority of reports of primary subacute osteomyelitis in the literature are a mix of adult and paediatric cases. In reports focusing on children the lesions are most commonly found within the long bones of the lower limb, with tibia and femur being the most frequently affected sites (*17,20-21*). Primary subacute osteomyelitis affecting other bones is more unusual but sites reported to have been affected

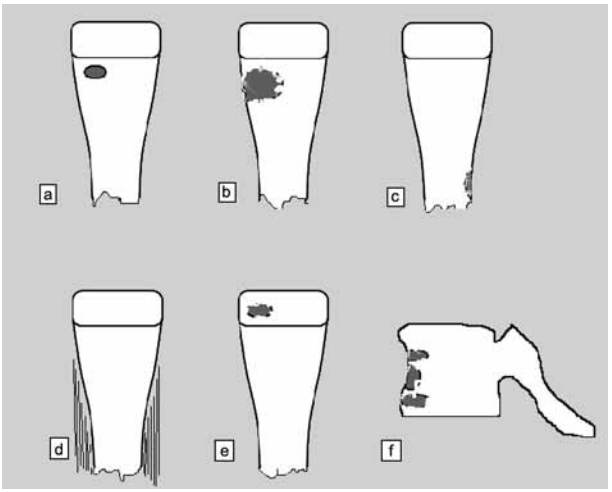

Fig. 3. — Classification of subacute osteomyelitis : a. indicates lucency type ; b. metaphyseal with loss of cortical bone ; c. diaphyseal ; d. onion-skinning ; e. epiphyseal ; and f. spine (adapted from McCarthy *et al* (*16*)).

include the clavicle, humerus, radius, ulna, calcaneum, talus and metatarsal (*20*) ; navicular (*4*) ; and spine (*7*).

An important differential diagnosis that must be excluded is presentation of benign or malignant (primary or metastatic) bone tumours. Therefore, where clinical suspicion is sufficient, a surgical approach is advocated in order to obtain tissue samples for the exclusion of neoplastic bone disease. Potential diagnostic difficulties are demonstrated by the frequent misdiagnoses of subacute osteomyelitis as Ewing's sarcoma (*5,21*), chondroblastoma (*5*), osteosarcoma (*5,21*), eosinophilic granuloma (*5*), fibrosarcoma (*19*), giant cell tumour (*5,21*), osteoid osteoma (*21*), metastatic neoplasm (*21*) and bone tumour of unknown origin (*5*).

Surgical intervention is also appropriate in advanced and/or aggressive cases where there is suspicion of a collection of subperiosteal pus or intra-articular pus in order to washout the collection and to obtain microbiology samples. Although causative organisms are infrequently identified, *Staphylococcus aureus* is the most commonly isolated microorganism (*14,17,20,22*). Other agents that have also been implicated include *Staphylococcus epidermidis* and *Streptococcus* (*3*) ; *Pneumococcus, Klebsiella* and *Kingella* (*17*) ; *Peptostreptococcus, Escherichia coli* and *Enterobacter* (*21*) ; *Salmonella* (*4*) ; and *Propionibacterium acnes* (*7*). Typical histological features are of a cellular and vascular inflammatory tissue with bony necrosis and destruction and an infiltrate containing neutrophils, macrophages and lymphocytes (*17,22*).

There is controversy regarding the management of this condition. Traditionally, authors have advocated surgical intervention in the form of curettage and biopsy with post-operative immobilisation and antibiotics for 6 weeks (*21*). Some authors have reported successful resolution of lesions after adequate surgical debridement without any antibiotics at all (*11*) though others recommend continuing treatment for anything up to 4 months (*5*) post-operatively. Treatment with antibiotics alone has been advocated with surgical intervention reserved for those cases where there is diagnostic uncertainty or that do not respond to initial antibiotic therapy (*8-9,12-14,20*). Successful outcomes have been reported with antibiotic therapy for a minimum total of 4 weeks (*9*) but more typically of 6 weeks duration. All regimens reported to date have consisted of initial intravenous antibiotics followed by prolonged courses of oral therapy.

Complications through inadequate treatment or progressive forms of osteomyelitis are rare in children. These may include recurrence, growth disturbance, pathological fracture and conversion to a chronic osteomyelitis. A survey of 50 patients treated over a period of 8 years revealed a complication rate of 12% but none in the subgroup of patients diagnosed with subacute osteomyelitis (*3*).

Our case series illustrates the various options available in the management of primary subacute osteomyelitis of the paediatric talus. Both surgical management and non-operative treatment, using antibiotics administered either intravenously or orally, was effective. In addition, it highlights that (i) operative management can have complications, some of which may be persistent ; and (ii) oral antibiotics alone can be sufficient for successful treatment.

Careful case selection is important in the choice of non-operative management. As well as having a

good local blood supply and a high propensity to bony healing, we consider the following as essential : i) unequivocal diagnosis ; ii) no joint involvement ; iii) no previous treatment. Where there is doubt about the diagnosis, or treatment to date has been unsuccessful, tissue and microbiological samples should be obtained for further analysis, in order to exclude neoplasm and to guide antimicrobial therapy. Surgical debridement may also be required.

In view of our experiences we wish to highlight that, in appropriately selected cases, intravenous antibiotics, hospital admission and/or surgical intervention are not always necessary for the successful management of this condition. Oral antibiotics alone can be sufficient for the successful treatment of primary subacute osteomyelitis of the paediatric talus.

REFERENCES

1. **Antoniou D, Connor AN.** Osteomyelitis of the calcaneus and talus. *J Bone Joint Surg* 1974 ; 56-A : 338-345.
2. **Arnold SR, Elias D, Buckingham SC *et al.*** Changing patterns of acute hematogenous osteomyelitis and septic arthritis : emergence of community-associated methicillin-resistant Staphylococcus aureus. *J Pediatr Orthop* 2006 ; 26 : 703-708.
3. **Blyth MJ, Kincaid R, Craigen MA, Bennet GC.** The changing epidemiology of acute and subacute haematogenous osteomyelitis in children. *J Bone Joint Surg* 2001 ; 83-B : 99-102.
4. **Cakmak Celik F, Sayli TR, Ocguder DA, Bozkurt M, Okdemir D.** Primary subacute Salmonella osteomyelitis of the navicular bone in a child with normal immunity. *J Pediatr Orthop* 2009 ; 18-B : 225-227.
5. **Cottias P, Tomeno B, Anract P, Vinh TS, Forest M.** Subacute osteomyelitis presenting as a bone tumour. A review of 21 cases. *Int Orthop* 1997 ; 21 : 243-248.
6. **Craigen MAC, Watters J, Hackett JS.** The changing epidemiology of osteomyelitis in children. *J Bone Joint Surg* 1992 ; 74-B : 541-545.
7. **Do TT, Strub WM, Witte D.** Subacute Propionibacterium acnes osteomyelitis of the spine in an adolescent. *J Pediatr Orthop* 2003 ; 12-B : 284-287.
8. **Ezra E, Cohen N, Segev E *et al.*** Primary subacute epiphyseal osteomyelitis : role of conservative treatment. *J Pediatr Orthop* 2002 ; 22 : 333-337.
9. **Ezra E, Wientroub S.** Primary subacute haematogenous osteomyelitis of the tarsal bones in children. *J Bone Joint Surg* 1997 ; 79-B : 983-986.
10. **Gafur OA, Copley LA, Hollmig ST *et al.*** The impact of the current epidemiology of pediatric musculoskeletal infection on evaluation and treatment guidelines. *J Pediatr Orthop* 2008 ; 28 : 777-785.
11. **Gledhill RB.** Subacute osteomyelitis in children. *Clin Orthop Relat Res* 1973 ; 96 : 57-69.
12. **González-López JL, Soleto-Martín FJ, Cubillo-Martín A *et al.*** Subacute osteomyelitis in children. *J Pediatr Orthop* 2001 ; 10-B : 101-104.
13. **Hamdy RC, Lawton L, Carey T, Wiley J, Marton D..** Subacute hematogenous osteomyelitis : are biopsy and surgery always indicated ? *J Pediatr Orthop* 1996 ; 16 : 220-223.
14. **Hayes CS, Heinrich SD, Craver R, MacEwen GD.** Subacute osteomyelitis. *Orthopedics* 1990 ; 13 : 363-366.
15. **Jones NS, Anderson DJ, Stiles PJ.** Osteomyelitis in a general hospital. A five-year study showing an increase in subacute osteomyelitis. *J Bone Joint Surg* 1987 ; 69-B : 779-783.
16. **McCarthy JJ, Dormans JP, Kozin SH, Pizzutillo PD.** Musculoskeletal infections in children – Basic treatment principles and recent advancements. *J Bone Joint Surg* 2004 ; 86-A : 850-863.
17. **Rasool MN.** Primary subacute haematogenous osteomyelitis in children. *J Bone Joint Surg* 2001 ; 83-B : 93-98.
18. **Roberts JM, Drummond DS, Breed AL, Chesney J.** Subacute hematogenous osteomyelitis in children : a retrospective study. *J Pediatr Orthop* 1982 ; 2 : 249-254.
19. **Robertson DE.** Primary acute and subacute localized osteomyelitis and osteochondritis in children. *Can J Surg* 1967 ; 10 : 408-413.
20. **Ross ER, Cole WG.** Treatment of subacute osteomyelitis in childhood. *J Bone Joint Surg* 1985 ; 67-B : 443-448.
21. **Shih HN, Shih LY, Wong YC.** Diagnosis and treatment of subacute osteomyelitis. *J Trauma* 2005 ; 58 : 83-87.
22. **Skevis XA.** Primary subacute osteomyelitis of the talus. *J Bone Joint Surg* 1984 ; 66-B : 101-103.