Osteonecrosis of the hip in sickle-cell disease associated with tuberculous arthritis

A review of 15 cases

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Summary. *We report a study of 15 cases of tuberculous hips with sickle-cell disease who presented during 1991–1993. Although the osteonecrosis was long-standing, biopsy was nearly always required to reveal the more recent tuberculous infection. Management consisted of 6 months of anti-tuberculous chemotherapy with appropriate palliative surgery 5–8 weeks after the start of drug treatment. The operative techniques which we used are described. The results were good both post-operatively, and in 12 patients followed-up at an average of 3 years. We recommend this combined management for the treatment of secondary tuberculous infections of hips previously damaged by sickle-cell disease.*

Résumé. *Cette étude concerne 15 cas d'arthrite tuberculeuse compliquant une ostéonécrose drépanocytaire de hanche observés de 1991 à 1993. La nécrose étant antérieurement connue, le diagnostic de tuberculose fut affirmé sur l'évolutivité septique et la biopsie. Nous avons opté pour une chirurgie palliative, intégrée à la chimiothérapie antituberculeuse et plus adaptée aux lésions, à la fragilité constitutionnelle et au jeune âge des patients. Ce traitement a abouti, avec une faible morbidité post-opératoire, à 12 résultats fonctionnels satisfaisants avec un recul de 3 ans. Face à l'agression tuberculeuse de ces hanches précaires, notre choix thérapeutique a constitué une excellente alternative.*

Introduction

In our region, bacterial infection is the dominant cause of arthritis in adults [11]. In hospital practice,

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however, an increasing number of tuberculous hips is presenting, often in patients with pre-existing bone necrosis due to sickle-cell disease. The combination of these two problems increases the loss of joint function [8, 16].

This study was designed to analyse the principal epidemiological and anatomical features, and to describe our method of management.

Patients and methods

Fifteen patients (8 men and 7 women) presented with sickle-cell disease of a hip complicated by tuberculous infection between 1991 and 1993 (Table 1). Their average age was 29 years (18–58 years). Five were homozygous (SS) and had involvement of the hip joint; the others comprised a heterogeneous group of whom 7 were composite heterozygotic (SC) and 3 thalassaemic (Sthal) with avascular necrosis of the femoral head of an average duration of 8 years.

Diagnostic criteria

These included clinical and radiological examination with biological and histological investigation (Table 1).

Clinical and radiological findings

Six patients had no previous relevant history, but among the others there were 2 with pulmonary tuberculosis, 1 with osteomyelitis of the mandible, and 6 with discharging sinuses from hip infections for which the cause had not been revealed at previous arthrotomies. These sinuses had been present for an average of 8 months (4–9 months).

The presenting symptoms and signs were a painful limp, a reduced walking distance and deterioration in the general condition of the patient. There were 2 with persistent groin sinuses (case nos. 5 and 16). Clinical examination revealed a flexion contracture of 45–90° (12 cases), and complete ankylosis in 3 cases. There was concomitant muscle wasting.

Table 1. The patients

Case	Age (yrs)	Sex	Geno-type	Period of necrosis (years)	Hip	Duration of infection (months)	Previous	ESR/Hb mm/q per l	IDR/HIV	Biopsy	Treatment
1	58	M	SS	15	R Ankylosis Destruction of head and neck	8	Arthritis & sinus	102/10.4	-/+	Caseous Tb & Tb bacilli	Girdlestone excision
2	25	F	SThal	9	L Stiffness	6	None	25/ 9.4	+++/-	Tb synovitis	Arthrodesis
3	36	F	SC	20	Sclerotic OA hip L Stiffness	19	Arthritis & sinus	60/10.2	+++/-	Granulomatous synovitis	Judet CTI
4	28	M	SS	3	Destruction of trochanter R Stiffness	12	Pulmonary Tb	28/10.7	++/-	Tb synovitis	Judet CTI
5	41	M	SC	8	Destruction of trochanter R Stiffness	8	Arthritis & sinus	90/ 9.7	++/-	Non-specific	Arthrotomy
6	25	M	SC	5	Sclerotic OA hip L Ankylosis	17	Arthritis & sinus	10/11.4	+/-	Non-specific	Arthrotomy
7	18	M	SC	3	Sclerotic OA hip R Stiffness	12	None	25/12	+++/-	Tb synovitis	Girdlestone excision
8	21	M	SS	10	Destruction of head and neck R Stiffness	5	Osteomyelitis of mandible	40/10.2	-/+	Tb synovitis	Girdlestone excision
9	28	M	SC	12	Destruction of head and neck L Ankylosis	7	Pulmonary Tb	27/10.1	+/+	Caseous Tb & Tb bacillus	Girdlestone excision
10	30	F	SC	6	Destruction of head and neck R Stiffness	5	None	20/10.1	+++/-	Granulomatous synovitis	Girdlestone excision
11	27	F	SThal	5	Sclerotic OA hip R Stiffness	9	Arthritis & sinus	30/ 9.8	++/-	Tb synovitis	Judet CTI
12	30	F	SS	9	Destruction of trochanter R Stiffness	5	None	19/10.2	+/+	Granulomatous synovitis	Girdlestone excision
13	21	F	SS	11	Destruction of head and neck R Stiffness	4.5	None	35/ 9.8	++/-	Non-specific	Girdlestone excision
14	23	M	SThal	3	Destruction of head and neck L Ankylosis	6.5	None	12/ 9.5	+++/-	Caseous Tb & Tb bacillus	Arthrodesis
15	22	F	SC	2	Sclerotic OA hip R Stiffness	6	Arthritis & sinus	15/11	+/-	Non-specific	Judet CTI

CTI: Coaptation Trochantero-Iliaque; R: right side; L: left side

Fig. 1a–c. SS patient, 31 years old, 5th month of evolution: **a** Bilateral femoral osteonecrosis at presentation; **b** Sclerotic coxarthrosis with right cephalic fragmentation, 6 weeks later; **c** 2 years after a Girdlestone excision

Radiographs showed evidence of rapid and major joint destruction: 5 revealed sclerosis or fragmentation of the femoral head, 6 destruction of the head and neck (with or without subluxation), and 4 destruction of the trochanter.

Laboratory investigations

In 12 patients the sedimentation rate was either normal or marginally raised, and in the others there was a marked increase. Five patients had a haemoglobin level of 9.4–9.8 grams% (3 Sthal, 1 SC, 1 SS). The intradermal reaction (IDR) was positive in 13, and negative in 2. There was little evidence of pre-

Fig. 2a–c. SS patient, 28 years old, 12th month of evolution: **a** Anterior-posterior view showing bilateral osteonecrosis; **b** Lysis of the right femoral head and neck; **c** CTI result at 5 years

vious BCG vaccination. Four patients who had received a blood transfusion were HIV positive (case nos. 1, 8, 9, 12).

Histology

In 75% of the patients, typical tuberculous lesions were found in synovial biopsies, but this investigation was not conclusive in 4. Koch's Bacillus was only cultured from material removed from HIV positive patients. Thus in many of the patients the diagnosis of tuberculosis depended on a 'positive' biopsy as there was no evidence of a generalised tuberculous infection elsewhere.

Management

The aim of our treatment was to eradicate the infectious process and to preserve as much bone as possible.

Medical treatment included a 6 months regimen of anti-tuberculous chemotherapy with Rifampicin, Isoniazid and Pyrazinamide for 2 months followed by Rifampicin and Isoniazid for 4 months. Surgery was performed during the first 6–8 weeks. A prophylactic regimen against sickle-cell crisis and venous thrombosis was carried out 10–15 days after operation using an anti-inflammatory drug (keprofen), a vasodilator (pentoxifyllin) and an anticoagulant (calcium nadroparin).

Four types of operation were undertaken, but no attempt was made to insert a prosthesis on account of the poor bone quality resulting from the septic osteolysis associated with sickle-cell disease. In all cases a thorough debridement was performed under general anaesthesia.

Two hips in which bone destruction was not extensive were treated by arthrotomy and excision of the fistula. Two with stiff joints, but with reasonable bone and joint congruity and no abscesses, had an intra-articular arthrodesis using a screw but no bone graft. Girdlestone excision arthroplasties (RTC) were performed on 7 patients with destroyed and subluxated hips with or without sinuses, followed by traction for 3 weeks, and then 6 weeks of 'non-weight-bearing' (Fig. 1). Four septic hips, which had previously been operated upon and where there was considerable bone loss of the head and trochanter, were treated by Judet's trochanteric-iliac coaptation, and were temporarily stabilised by screw or pin fixation (Fig. 2).

A plaster hip-spica was used for all patients for 6–8 weeks except those treated by a Girdlestone excision. This immobilisation was followed by physiotherapy and appropriately increasing weight-bearing.

Results

The patients were assessed after operation and at follow-up.

Post-operative results (Table 2)

None of the patients with SS disease developed a postoperative occlusive vascular crisis as a result of a possible hypoxia during anaesthesia. Superficial infection with staphylococcus, streptococcus, colibacteria, or klebsiella occurred in 5 cases (nos. 1, 3, 5, 13, 15). All these infections resolved within 10–15 days, with little influence on wound healing which occurred on average in 3 weeks. Satisfactory bone healing or fusion, together with considerable relief of

symptoms, occurred within 46 days for the arthrotomies, and 85 days for the arthrodeses.

Results at follow-up (Table 3)

At an average of 3 years after operation (range: 2.5 to 4 years) there was no evidence of active tuberculosis, and all the patients were as mobile as before treatment. Functional assessment was based on the criteria of Merle d'Aubigne [17]:

- Pain.** The operated hip was still painful in 3 cases (1 RTC, 2 CTI). There were 3 painful knees distal to pain-free hips, 5 contra-lateral hips were painful, and 2 patients had low back pain after hip arthrodesis.
- Joint mobility.** No hip regained more than 100° of flexion, but 70–90° was found after hip excision (RTC). However, 11 hips were very stiff, with 7 ankylosed.
- Stability.** All patients were able to stand on one leg except for those with a hip excision. Four patients were totally unable to crouch, and for another 4 this manoeuvre was difficult to do.
- Walking.** Shortening of 2 cm or more was always associated with limping, and this was more marked when the discrepancy was greater than 4 cms. An ankylosed hip made it difficult to put on shoes, and the majority of the patients used 1 (10 cases) or 2 (2 RTC) walking sticks.

Discussion

When considering the aetiology and pathology it is important to remember that an old tuberculous focus in a limb or elsewhere is often reactivated in HIV positive patients [5]. The tubercle bacillus appears to have an affinity for both healthy and 'damaged' hip joints [15, 16]. Hips with avascular necrosis resulting from sickle-cell disease provide an ideal location for the tubercle bacillus [2, 7, 10].

In our series, tuberculous infection occurred in young sickle-cell patients (average age 29 years) for whom medical treatment had been able to control the avascular necrosis for some years (average 8). This helps to explain the late presentation, and the severity of their lesions when they did seek surgical help.

Table 2. Immediate post-operative results

	Arthrotomies (2 cases)	Girdlestone excision (7 cases)	Arthrodesis (2 cases)	CTI of Judet (4 cases)
Sepsis	<i>Staphylococcus aureus</i> + <i>Streptococcus</i> (1)	<i>Staphylococcus aureus</i> + <i>Klebsiella pneumoniae</i> (1) <i>Streptococcus</i> (1)	0	<i>Staphylococcus aureus</i> (1) Colibacilli (1)
Wound healing	22 days	20 days	20 days	23 days
Consolidation	46 days	74 days	85 days	67 days

Table 3. Results at follow-up

	Pain	Hip joint mobility				Stability		Walking		Follow-up (years)				
		Operated hip	Opposite hip	>100 v. good	90–70° good	<70° poor	Ankylosed bad	Squatting	Standing on one leg	Shortening	No walking stick	1 walking stick	2 walking sticks	
Arthrotomy (2 cases)	0	0	0	0	0	1	1	2 difficult	2	2–3 cm: 1 4–5 cm: 1	1	1	2	3.5
Girdlestone excision (7 cases)	1	2	0	4	3	0	0	7 possible	0	4.5 cm: 1 >5 cm: 2	0	5	2	3
Adese (2 cases)	0	2	0	0	0	2	2	2 difficult	2	2–3 cm: 2	1	1	0	2.5
CTI of Judet	2	1	0	0	0	4	0	0	4	2–3 cm: 2 4–5 cm: 2	1	3	0	4

The diagnosis of tuberculosis in a previously necrotic hip is difficult, partly due to its insidious onset, and also because of the changes already resulting from the underlying sickle-cell disease [8]. This resulted in an average delay of 9 months. Persistent pain, previous infection and bone destruction suggested the diagnosis, and the intradermal reaction (IDR) was positive in 9 patients. However, in endemic regions, a weakly positive or a negative reaction does not rule out a tuberculous infection [9]. Three quarters of our patients had infection confirmed by synovial biopsy [4]. The rarity of positive culture of the tubercle bacillus is really only relevant in sero-positive patients when examination has failed to reveal other sites of tuberculosis [20, 21].

The efficacy of 6 month courses of appropriate chemotherapy in tuberculous hips is well established [1, 3] and we have opted for palliative surgery with major lesions or when there was significant loss of function. We considered that an arthroplasty [22] in these septic situations was too risky, and we preferred the arthrodesis, the CTI of Judet, or a Girdlestone excision, all of which are well established in the management of septic hip lesions [13, 14]. Furthermore, these procedures all allow further surgery [12, 18] often with good results [19, 20]. The results we have obtained from relatively simple surgery have on the whole been acceptable and somewhat unexpected, with consolidation of bone and no evidence of disease or re-appearance of infection. At an average follow-up of 3 years the functional results were satisfactory in spite of 3 hyper-mobile hips, and 7 which were ankylosed. Twelve patients said that they were better with less disability, less pain and increased general mobility. Only 3 patients were considered to be candidates for a future replacement. Tuberculous arthritis of the hip associated with sickle-cell disease is probably related to the damage produced by avascular necrosis which has produced an ideal site for the activities of the tubercle bacillus. Although this combination as a cause of a tuberculous hip is relatively rare [6] when compared to the incidence of uncomplicated tuberculosis of the adult hip, it does accentuate various important points:

- The possibility of a tuberculous hip infection in a sickle-cell patient should be considered in areas where these two conditions are relatively common, and where there is rapid extension of previous necrotic lesions;
- Specific chemotherapy greatly improves the results of surgery;
- Appropriate 'palliative' surgery with preservation of as much of the femoral head as possible will help to allow replacement in the future.

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