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Prosthetic Hip Loosening Due to Brucellar Infection: Case Report and Literature Review

Tebourbi A[1], Hadhri K[2], Salah M[2], Bouzidi R[1], Kooli M[2]

Abstract

Context: Brucellosis is actually considered to be the commonest zoonotic infection worldwide; conversely prosthetic infection due to brucella is extremely rare. Although diagnostic is easily achieved, management of such situations is extremely challenging.

Aims: To report the case of prosthetic hip loosening due to brucellar infection, discuss management manners and to summarize data about 19 cases reported in the literature.

Methods: We report the case of a 73-year-old woman with brucellar prosthetic hip loosening treated with 2-stage exchange of the prosthesis and prolonged double antibiotherapy

Results: At two years follow up the patient is pain free with total functional recovery and no clinical and radiographic signs of prosthetic loosening

Conclusions: Brucella should be evocated as a cause of total joint arthroplasty infection especially in patients from endemic regions and with occupational exposure. Antibiotic treatment alone can be followed if there are no signs of implant loosening. Tow stage revision should be considered in other cases

Keywords: *total hip arthroplasty; prosthetic joint infection; Brucellosis*
Level of Evidence: *AAOS Therapeutic Level IV*

Introduction

Brucellosis is now considered to be the commonest zoonotic infection worldwide with more than 500.000 new cases annually [1,2]. According to the World Organization for Animal Health, North Africa has been traditionally considered endemic. Brucellosis affects several types of animals, including cows, sheep, goats, deer, pigs and dogs. Six species of gram-negative bacteria belonging to the genus Brucella are responsible for infection: B. melitensis, B. abortus , B. suis, B. canis, B. ovis, and B. neotomae. Only the first four are able to infect humans by ingestion of con-

1 Anis Tebourbi, Ramzi Bouzidi
Department of Orthopedics and Traumatology, Mongi Slim Hospital, La Marsa, Tunisia
2 Khaled Hadhri, Mohamed Ben Salah, Mondher Kooli
Department of Orthopedics and Traumatology, Charles Nicolle Hospital, Boulevard 9 Avril, 1006 Tunis, Tunisia
(Direct reprint requests to Khaled Hadhri)

taminated food or drinks, inhalation of the organism, or inoculation through skin abrasion.

Osteoarticular infection is the most common complication and has been described with rates of 10% – 85% in patients infected by brucella [3,4]. It affects the large joints, especially the sacro-iliac joint. Spondylitis, bursitis, tenosynovitis and osteomyelitis have been also described [5-11].

Conversely prosthetic infection due to brucella is extremely rare. In the literature there are 19 cases of total joint arthroplasty infected by Brucella involving only 9 hips.

In the present report we describe a case of prosthetic hip loosening due to this infection and review the literature.

Case Report

73-year-old woman presented to our hospital in March 2002 with two years history of hip pain. After radiographic examination, the patient was diagnosed with osteoarthritis of the hip. The peripheral white blood cell (WBC) count was 5500cells/mm3, the erythrocytes sedimentation rate (ESR) was 50mm/h, and the C-reactive protein (CRP) level was 4, 8mg/l.

She underwent cemented Charnley total hip arthroplasty through a posterior approach. Culture of specimen from the synovium obtained during surgery was negative and microscopic examination showed degenerative changes.

The hip was pain free and annual postoperative radiographs were satisfactory until the 10th year.

On February 2012 the patient was hospitalized for L2-L3 and T6-T7 brucellar spondylitis with discovertebral needle biopsy showing brucellar granuloma, positive Rose bengal test and 1/640 brucella serum antibody titer . She received standard chemotherapy with oral Doxyciline and Rifampicine.

In February 2014 the patient presented complaining of hip pain that had been present for the previous 8 months. There were no external signs of infection and no previous episod of fever. Radiographic examination showed evident signs of prosthetic loosening [Figure 1]. The inflammatory biomarkers included blood leukocytes count (8700cells/mm3) and C- reactive protein level (7,6 ng/l). A revision total hip arthroplasty (THA) was planned because of suspected asceptic loosening.

During the surgical procedure, purulent white fluid poured out of the joint with abundant necrotic tissue and marked loosening of the prosthesis.

At this stage revision total hip arthroplasty was abandoned and we proceed to resection arthroplasty through a large femorotomy [Figure 2] Several bacteriologic culture specimen and materials for pathologic examination were taken.

Fig. 1: Twelve year control x-ray demonstrating bipolar loosening of a total Charnley prosthesis of the left hip.

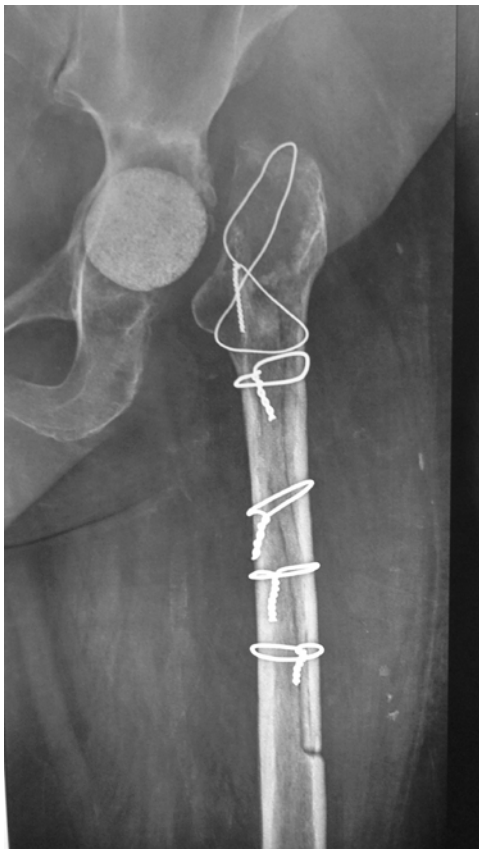

Fig. 2: Postoperative control x-ray after femorortomy and prosthesis extraction.

Empirical antibiotic treatment with Rifampicine 600mg and Ofloxacine400 was started.

The bacteriological culture were negative but serum antibody titer for Brucella was 1/320 and microscopic ex-

Fig. 3: Radiographic examination at two year follow up.

amination showed a brucellian granuloma. The antibiotic chemotherapy was maintained for 3 months, Ofloxacine wasn't changed because a history of digestive disagreement during the previous uses of Doxcycilline

A second stage of THA reimplantation was performed at the end of the antibiotic treatment period. At two years follow up the patient is pain free with total functional recovery and no clinical and radiographic signs of prosthetic loosening [Figure 3].

Discussion

Infection is considered to be the most devastating of prosthesis-related complication, leading to prolonged hospitalization, repeated surgical intervention. The incidence of prosthetic joint infection (PJI) is 1–2.5% for primary hip or knee replacements and 2.1 – 5.8% for revision surgeries [12-14].

The majority of infections (65%) are caused by aerobic gram-positive cocci, most commonly Staphylococcus aureus, coagulase negative staphylococci and enterococci. Aerobic gram-negative bacilli, anaerobes and mycobacterial infection are far less frequent [15]. PJI due to Brucella is an extremely rare condition, and only 19 cases have been reported in the literature [16-28]. The demographic characteristics of the 19 cases and our patient are summarized in Table 1 [Table 1]. Among these patients, there were 12 men with a mean age of 59 years. Most patients had occupationnal exposure to brucella history of unpastorised milk products consumption and lived in areas where brucellosis is endemic. The Hip was involved in 10 patients. Knees were involved in 8 patients with two bilateral cases. Systemic symptoms of brucellosis as fever, headache, weakness, sweetness, profuse sweating, splenomegalia, adenopathy are non specific and were present in only four cases. Local symptoms as night pain, swelling, local inflammation, sinus tract formation and restriction of the joint movement were present in nearly all cases. Radiographic signs of loosening were found in nine cases. The rate of isolation of brucella in patients with osteoarticular brucellosis oscillates between 33% and 77% [3]. In the reviewed cases culture of synovial fluid sample or tissue sample recovered brucella in 17 cases (85%).In one case the germe was isolated from a sinus tract discharge [28] and from blood samples in two cases [18,23]. Laboratory culture of brucella is often unsuccessful because of the slow-growing nature of these microorganisms and the requirement for special media and high Co2 tension. So the culture period should be made longer than usual and clinicians should notify the laboratory staff if there is a suspicion of brucellosis [29]. Negative joint culture result does not rule out osteoarticular brucellosis and the diagnosis can be made through the detection of specific antibodies in serum. In active brucellosis, high titers of IgM antibodies can be detected by standard agglutination and Rose Bengal tests, which are followed by an increase of IgG and IgA antibodies in chronic stage of the disease [30]. In our review 18 patients (90%) had positive titers of specific antibodies (>=80) . Generally joint prothesis can become infected through three different routes: Implantation, hematogenic infection, and reactivation of latent infection [31]. In Brucellar PJI most of the authors support the hematogenous route [24]. Previous spinal brucellosis in our patient supports this septic pathogenesis of the articular involvement. Because of the rare occurrence of PJI caused by Brucella, there is no consensus on its management. The most accepted course is antibiotic treatment with removal or retention of prosthetic components [23]. A variety of drugs have activity against brucella, however the results of in vitro susceptibility tests do not always predict clinical efficacy [32].The intracellular localization of brucella is believed to offer some protection against antimicrobials, and drugs with good penetration into cells are thought necessary for cure. Monotherapy for brucellosis has generally been considered inadequate due to unacceptably high relapse rate. Of the 20 cases that we reviewed, 14 were treated with double antibiotic therapy [16-18,20-23,25-28], the most used association was Doxycylline and Rifampicine (8 cases) this association offers the advantage of an all-oral treatment and was recommended by the World Health Organization (WHO) in 1986 [33] and by Consensus " Loannina Recommendations" for

Table1: Summuary of the reported cases in the literature

	Year	Age / sex	Implant	Exposure	Symptoms	Radiology	Surgical treatment	Antibiotic treatment	Recurrence	Follow up
Jones et al (16)	1983	54 /M	THA	Professional	Systemic	No Loosening	Surgical debridement	Doxycycline 6w/ Streptomycine6w	No	3y
Agarwall et al (17)	1991	24/f	02 TKA	No	local	No Loosening	NO	Rifampicine 19m/ cotrimoxazole19m	NO	19m
Malizos et al (18)	1997	74 /M	02TKA	Professional	Systemic	No Loosening	NO	Streptomycine 3w/ Doxycycline 5m	yes	2y
Orti et al (19)	1997	60 /M	TKA	Professional	local	No Loosening	NO	Doxycycline 6w Rifampicine 6w Streptomycine 3w	NO	8m
Ortega et al (20)	2002	63/M	THA	Professional	local	Prosthetic loosening	2 stage replacement	Streptomycine 3w Doxycycline 3m	NO	6m
Weil et al (21)	2003	38/M	THA	Unpastorised milk consumption	local	Prosthetic loosening	2 stage replacement	Doxycycline 12w Rifampicine12w	NO	1y
Weil et al (21)	2003	61/M	TKA	Unpastorised milk consumption	local	Prosthetic loosening	2 stage replacement	Doxycycline 12w Rifampicine 12w	NO	1y
Weil et al (21)	2003	67/M	TKA	Unpastorised milk consumption	Systemic	Prosthetic loosening	2 stage replacement	Doxycycline 12w Rifampicine12w	NO	1y
Kasim et al (22)	2004	54/F	THA	No	local	Prosthetic loosening	1 Stage replacement	Vibramycin 5m Rifampicine5m	NO	4y
Cairo et al (23)	2006	50/M	THA	Professional	local	NR	NO	Doxycycline 26m Streptomycine 2w	NO	5y
Cairo et al (23)	2006	21/M	THA	Professional	local	NR	1 Stage replacement	Doxycylline 6m Rifampicine 6m Streptomycine 10d	NO	3y
Tena et al (24)	2007	56/M	THA	Professional	local	Prosthetic loosening	2 Stage replacement	Doxycylline 2m Rifampicine 2m Streptomycine 2w	NO	4y
Ruiz-Iban (26)	2006	66/F	THA	Contact with Cattle	local	Prosthetic loosening	2 Stage replacement	Doxycylline 6w Rifampicine 6w	NO	5,5y
Ruiz-Iban (26)	2006	71/M	THA	Professional	local	No Loosening	Surgical debridement	Doxycylline 6m Rifampicine 6m Strptomycine 1m	NO	5y
Tassinari et al (25)	2008	68/M	TKA	NR	Systemic	No Loosening	NO	Doxycylline 8w Rifampicine 6w	NO	1y
Wunschel et al (28)	2011	64/F	TKA	NR	Local	Prosthetic loosening	1 Stage replacement	Doxycylline 8w Rifampicine 6w	NO	NR
Erdogan et al (27)	2010	63/F	TKA	Unpastorised milk consumption	local	No Loosening	NO	Doxycylline 4m Rifampicine 4m	NO	3y
Our Case	2014	73/F	THA	Unpastorised milk consumption	Local	Prosthetic loosening	2 Stage replacement	Rifampicine 6m Ciprofloxacine 6m	No	6m

M= Male, F= Female ; TKA= Total Knee arthroplasty, THA= Total Hip arthroplasty ; y= years, m= Months, w= Weeks,

the Treatment of Human Brucellosis in 2006 [34] . The most used antibiotics are Doxycyclline in 15 cases, Rifampicine in 14 cases, Streptomycine in 8 cases, Vibramicyne in 1 case, cotrimoxasole in 1 case and ciprofloxacine in 1 case. In our patient, because of the digestive disagreement, we used flouroquinolone which is considered as an acceptable alternative to doxycycline [34]. The total duration of antibiotic therapy necessary for eradication of the infection is unknown. In the review antibiotic treatment lasted from 6 weeks to a maximum of 26 months. Six Weeks is the duration recommended by the WHO and the Loannina Consensus. In six cases out of twenty the infection resolved with the sole use of the antibiotic therapy without having to resort to surgical revision [17,18,19,23,25,27] in this patient there was no radiographic evidence of implant loosening. A single stage prosthetic revision was done in 3 cases but only because the infection had not been suspected from the beginning [22,23,28]. In 7 cases a two stages revision was done, this procedure is believed to be the treatment ofchoice for loosened total joint arthroplasty infected with brucella. In brucellosis , even with effective drug treatment, relapses occur in 5–10% of patients, usually in the early post-treatment period [34], in our review the infection was recurrent in only one patient out of twenty [18].

Conclusions

Brucella should be evocated as a cause of total joint arthroplasty infection especially in patients from endemic regions and with occupational exposure. Antibiotic treatment alone can be followed if there are no signs of implant loosening. Tow stage revision should be considered in other cases.

Disclosure

The authors declare that there is no conflict of interest regarding the publication of this paper. For full disclosures refer to last page of this journal.

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