Case Report

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Stenotrophomonas maltophilia infection: an unusual complication of total elbow arthroplasty

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Introduction: This case report describes the secondary infection of a total elbow prosthesis with the uncommon Gram-negative organism *Stenotrophomonas maltophilia*. To our knowledge, this is the first reported case of an elbow arthroplasty being affected by this organism.

Case presentation: A 57-year-old male had a right total elbow replacement in 2009, which subsequently became infected with *Stenotrophomonas maltophilia*, eventually leading to an above-elbow amputation.

Conclusion: This case highlights the importance of a combined surgical and microbiological approach for the difficult treatment of deep joint replacement infections. Although *Stenotrophomonas maltophilia* is currently a rare pathogen in infection following arthroplasty, given the increasing number of multidrug-resistant organisms, it may represent a future challenge to both surgeons and microbiologists.

Keywords: Arthroplasty; elbow; infection; multi-drug resistance; *Stenotrophomonas maltophilia*.

Introduction

Approximately 125 231 joint replacements and 442 total elbow replacements were performed in England and Wales in 2013 according to data collected by the National Joint Registry (2014). Elbow replacements have a deep infection rate ranging from 1 to 12 % (Ewald *et al.*, 1993; Kasten and Skinner, 1993), with an estimated mean of 5 % (Little *et al.*, 2005).

Staphylococcus aureus and *Staphylococcus epidermidis* together account for approximately 50 % of the agents isolated from infected joint prostheses, making them the most common causative bacteria (Tattevin *et al.*, 1999). Bacteria responsible for elbow prostheses infections include *Staphylococcus aureus*, *Staphylococcus epidermidis*, *Klebsiella* spp. and *Enterobacter cloacae* (Yamaguchi *et al.*, 1998).

Stenotrophomonas maltophilia is a Gram-negative aerobe. It is naturally found in aqueous environments and is part of an emerging class of multidrug-resistant organisms (Amin and Waters, 2012). It is associated with respiratory infections in patients with cystic fibrosis, often in conjunction with *Pseudomonas aeruginosa* (Amin and Waters, 2012; Brooke, 2012). Isolates have been reported in a number of implantable devices including venous lines, prosthetic aortic valve replacements and subclavian catheters (Mehta *et al.*, 2000; Ratnalingham *et al.*, 2002; Shah and Feinfeld, 2000). It has also been cited as the responsible agent in one case of deep arthroplasty infection (Tattevin *et al.*, 1999) and has been isolated from hip joint prostheses using PCR (Dempsey *et al.*, 2007).

Case report

Our patient was a 57-year-old, right-hand-dominant male who, in November 2009, underwent a right total elbow replacement for osteoarthritis and presence of a loose body. The patient was an arteriopath, having had a previous coronary artery bypass graft and was a lifelong smoker. He had no diabetes or immunosuppression. At no time did he become neutropenic during antibiotic therapy.

Within a few weeks of the original operation, he developed signs of an infection. The wound was discharging and the joint was red, hot and swollen. In February 2010, the implant was removed in the first of a two-stage revision procedure. The second stage was performed in January 2011, but between the first and second stages, the patient had numerous washouts. Within 3 weeks of the second-stage revision, the new joint prosthesis became infected. In February 2011, the implant was removed and the patient had further washouts. Unfortunately, the patient developed a chronic wound over the incision site that would not heal. He also developed complete radial nerve palsy after the revision surgeries.

He was referred to a regional plastic surgery department where on 3 February 2012 he underwent washout, debridement and insertion of a cement spacer, plus both local and anterolateral thigh flap surgery. Fig. 1 summarizes the time line of surgical procedures and microbiological findings. The patient was put on intravenous meropenem 2 g three times daily and gentamicin

Fig. 1. Timeline of surgical procedures against microbiological findings.

Fig. 2. Plain radiograph of elbow once the prosthesis had been removed.

5 mg kg⁻¹ once daily following surgery. This was changed to ertapenem 1 g once daily to go home to the community. Unfortunately, continuing signs of infection, including fever and spreading cellulitis, meant he was re-admitted in April 2012 and put onto tazobactam/piperacillin and gentamicin intravenously. He had a further washout and bony debridement on 12 April 2012 and on 18 April 2012 his flap dehisced and was washed out, with shortening of the distal humerus. He underwent debridement one further time in April.

By May 2012, the elbow was still infected and on 11 May 2012 he had a final washout of frank pus from the humerus and application of a vacuum-assisted closure dressing (see Figure 2). The pus sample grew *Stenotrophomonas maltophilia* and he was placed on ceftazidime with colistin and finally ticarcillin/clavulanate intravenously.

Finally, despite therapy with intravenous ticarcillin/clavulanate and vacuum-assisted closure therapy, the patient showed evidence of continuing deep infection, including rising inflammatory markers. In November 2012, the patient underwent an above-elbow amputation, from which he has recovered well.

Discussion

The reported incidence of deep infection is <1 % overall for all arthroplasties (Lidgren, 2001). It presents a difficult

complication to treat, both surgically and microbiologically. The method of treatment varies greatly, from suppression therapy with antibiotics to revision arthroplasty. The choice of treatment is dependent on both patient factors and surgical preference or experience.

Single-stage revision arthroplasty involves removal of the infected prosthesis, washout and debridement, followed by reinsertion of a new prosthesis, all in one surgical sitting. Single-stage revisions require meticulous debridement and washout intra-operatively, with appropriate peri-operative antibiotic treatment.

The advantages of single-stage revision arthroplasty include: lower morbidity rates, a faster return to mobility, reduced expense and greater overall limb stability (Miley *et al.*, 1982). Success rates of 83–87 % can be achieved with single-stage revision (Saleh *et al.*, 2002). Of 1299 hip arthroplasties analysed (Jackson and Schmalzried, 2000). The following factors, as summarised in Table 1, were identified as being predictive of the outcome for single stage revisions.

Two-stage revision arthroplasty involves removing the prosthesis and inserting an antibiotic-impregnated spacer (or other material such as beads), followed by a variable period of antibiotic therapy with a combination of oral and intravenous antibiotics. The second stage is then to remove the antibiotic-loaded device and insert a new prosthesis. Two-stage revision has an overall success rate of approximately 91 % (range 89–100 %; Robbins *et al.*, 2001).

Stenotrophomonas maltophilia represents a challenge to both surgeons and microbiologists. Surgically, as it is a Gram-negative infection, two-stage revision is indicated. From a microbiological perspective, there is no suitable antibiotic-loaded device for this organism and therefore the challenge is to prevent its occurrence.

To our knowledge, this is the first reported case of *Stenotrophomonas maltophilia* in an infected elbow arthroplasty. This case highlights both the difficulties in treating deep joint replacement infections and the importance of continuing to sample microbiologically. Unfortunately, the outcome for this patient was poor, but perhaps, had the collaboration between microbiology and surgeons been closer earlier in his treatment, antibiotic use could have been more focused and the ultimate appearance of *Stenotrophomonas maltophilia* in his wound avoided.

Table 1. Predictors of successful outcome from single-stage revision arthroplasty

Successful revision	Unsuccessful revision
Meticillin-sensitive <i>Staphylococcus aureus</i> infection	Meticillin-resistant <i>Staphylococcus aureus</i> infection
<i>Streptococcus</i> infection	Polymicrobial infection
Healthy patient	Gram-negative infection
Bone cement impregnated with sensitive antibiotic	
No initial wound complications	

The *Stenotrophomonas maltophilia* isolated was resistant to all antibiotics except ceftazidime, co-trimoxazole and ticarcillin/clavulanate. *In vitro* synergy testing identified ticarcillin/clavulanate and ciprofloxacin to be the most active combination against this isolate (see Fig. 1).

Intravenous antibiotics failed because the patient had a chronic wound. Chronic wounds are associated with biofilms (Wolcott *et al.*, 2013). *Stenotrophomonas maltophilia* has an inherent ability to adhere to foreign materials and chronic wounds, forming a biofilm and rendering protection from host defences and antimicrobial agents. Factors contributing to this behaviour include its positively charged surface and fimbrial adhesions. In addition, *Stenotrophomonas maltophilia* is intrinsically resistant to all β -lactam antibiotics, including carbapenems, and aminoglycosides.

This patient was interesting in that he did not show features frequently associated with *Stenotrophomonas maltophilia*, namely debilitation, immunosuppression and neutropenia (Brooke, 2012; Looney *et al.*, 2009; Samonis *et al.* 2012). The organism emerged as a result of profound antibiotic pressure, as a result of long-standing infection. This case highlights the continuing need for sound antimicrobial stewardship to avoid such bacteria emerging and becoming commonplace.

Stenotrophomonas maltophilia is so far uncommon in deep infection of prosthetic joints; however, with increased antimicrobial resistance, it may represent an important challenge for the future.

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