Case report

Sternoclavicular joint septic arthritis and osteomyelitis caused by Aggregatibacter aphrophilus

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Case presentation

A 62-year-old lady was referred to the Infectious Diseases outpatients clinics with a 3-week history of right shoulder pain and a 1-week history of progressive swelling and erythema of the soft tissue overlying the right sternoclavicular joint (SCJ). No systemic symptoms were reported. No triggering event could be recalled and there was no recent travel, no animal exposure and no known contact with tuberculosis.

On examination, temperature was normal and the patient appeared systemically well. There was an indurated, tender, erythematous, warm swelling over her right SCJ with associated decreased range of movement of the right shoulder. Cardiac examination revealed a soft systolic murmur. No other peripheral stigmata of endocarditis were identified. The patient was admitted for investigation and work up of a suspected SCJ septic arthritis arising either as a consequence of an undocumented bacteraemia or due to a contiguous extension from overlying skin and soft tissue infection.

Initial laboratory investigations revealed a normal white cell count and differential. Inflammatory markers were elevated with an ESR of 94 mm/h (<36) and a CRP of 84 mg/l (<5). Serial blood cultures were obtained prior to therapy commencement. A targeted ultrasound (US) of the SCJ was performed urgently. This identified a small pocket of fluid adjacent to the right SCJ, intimately associated with the joint space. This fluid was aspirated under US guidance. Treatment comprising high-dose intravenous (IV) flucloxacillin and benzylpenicillin was subsequently commenced.

Imaging with computed tomography and magnetic resonance (MR) revealed marked soft tissue oedema surrounding the SCJ associated with oedema and inflammatory changes in the adjacent bone and parietal pleura (Figure 1a and b). A transthoracic echocardiogram did not identify any features suggestive of endocarditis. This finding was later confirmed with a trans-oesophageal echocardiogram.

Laboratory methods

Gram stain of the SCJ aspirate revealed few pus cells, and no organisms were seen. The fluid was inoculated onto chocolate and blood agars incubated under 10% CO₂; fastidious anaerobic agar with neomycin incubated anaerobically; and McConkey agar incubated in air. After 48 h, growth was detected on the chocolate agar only. The colonies appeared small, round and opaque with no requirement for X and V factor. The organism was identified as Aggregatibacter aphrophilus using an API NH®. Formal identification was confirmed by the Haemophilus reference unit at Colindale utilizing additional sugar fermentation tests. Testing for β-lactamase production using the hodge method proved negative. Minimum inhibitory concentrations (MICs) were carried out using the Liofilchem® MIC test strip and the organism was found to be susceptible to ampicillin (0.064 mg/l), amoxicillin/clavulanate (0.125 mg/l), cefotaxime...
(0.008 mg/l), trimethoprim (0.064 mg/l), ciprofloxacin (0.004 mg/l) and tetracycline (0.5 mg/l). All blood culture sets remained culture negative.

Upon identification of \textit{A. aphrophilus}, the anti-microbial regimen was altered to ceftriaxone 2 g IV daily and gentamicin 80 mg IV 8 hourly. Gentamicin was discontinued after 2 weeks and the patient was treated as per a clinical diagnosis of septic arthritis and osteomyelitis. A 6-week course of IV ceftriaxone was completed utilizing an outpatient parenteral anti-microbial therapy programme. The patient had an excellent clinical response with complete resolution of symptoms and the local soft tissue induration. Inflammatory markers had normalized by Day 14. A follow-up MR of neck and SCJ at 8 weeks identified radiological progression of the bone and joint abnormalities confirming the extent of local involvement (Figure 2a). Following the IV phase of treatment a decision was made to prolong therapy with oral ciprofloxacin 500 mg twice daily to complete 12 weeks of treatment in total. The patient remains well and symptom free on follow-up with no evidence of local disease recurrence.

Figure 2. Follow-up magnetic resonance image. Decreased signal intensity consistent with marrow oedema and a joint effusion with extension into the periarticular soft tissues as well as the extrapleural soft tissues adjacent to the apical right lung.

\textbf{Discussion}

\textit{Aggregatibacter aphrophilus} is a small, fastidious, Gram-negative, capnophillic coccobacillus. This organism, formerly known as \textit{Haemophilus aphrophilus}, was renamed following a review of the classification of it and other related organisms in 2006.\textsuperscript{1} It was first described in 1940 as the causative pathogen in a case of fatal infective endocarditis and is more renowned as a member of the HACEK (\textit{Haemophilus, Actinobacillus, Cardiobacterium, Eikenella, Kingella}) group of organisms, collectively causing 3\% of cases of infective endocarditis.\textsuperscript{2,3} Despite being an oral flora commensal, clinical infection is rare. Indeed it is a very rare cause of septic arthritis. We report the first case of a SCJ septic arthritis due to \textit{A. aphrophilus} and only the fifth reported case of septic arthritis due to \textit{A. aphrophilus}.

The carriage prevalence of \textit{A. aphrophilus} is 30–45\%.\textsuperscript{4} Despite its ubiquity in the oral cavity, infection is rarely encountered. This reflects both the lower virulence, and the very fastidious nature of this organism. Within the laboratory, isolation and identification of \textit{A. aphrophilus} from clinical specimens is particularly challenging.

While renowned as a pathogen causing infective endocarditis, historical series of infections due to this organism identified the most common entities (21 of 42 cases) to be wound infections and/or abscesses.\textsuperscript{5} This series contained no reports of bone or joint infection and only five cases of endocarditis. More recently Huang \textit{et al.} reviewed 28 cases of infection between 1990 and 2003. The clinical presentations were very diverse; however, the series did include seven cases of infective endocarditis, and nine with musculoskeletal sites of infection.\textsuperscript{6}
SCJ septic arthritis

The SCJ is a diarthroidal saddle type synovial joint articulated by the inferior medial clavicular head, superior lateral notch of the manubrium and the cartilage of the first rib. SCJ infections are a rare entity, accounting for 0.5–1% of the septic arthritis in the general population.7–10 Risk factors for SCJ infection include, but are not limited to, IV drug abuse, diabetes mellitus, trauma, central venous catheter infection, chronic renal failure, alcoholism, corticosteroid use, retroviral infection, malignancy, cirrhosis and rheumatoid arthritis.7–9

In a review of 180 cases of SCJ septic arthritis from 1970 to 2004 by Ross and Shamsuddin, the predominant pathogen identified was Staphylococcus aureus (49%), followed by Pseudomonas aeruginosa (10%), Brucella melitensis (7%) and Escherichia coli (5%). Bacteraemia was present in 62% with culture positive joint aspiration in 77%. Patients predominantly presented with chest (78%) and shoulder pain (24%).7

Septic arthritis due to A. aphrophilus has rarely been reported. In all only four cases of septic arthritis due to A. aphrophilus have been described in the medical literature to date. Page and King reported the first two cases of A. aphrophilus septic arthritis involving the left shoulder joint and the acromioclavicular joint in 1966. The organism was cultured from the synovial fluid and blood cultures, respectively, and both patients had favourable outcomes following treatment.11 Merino et al.12 reported a septic arthritis of the knee in 1993, which isolated A. aphrophilus on culture of synovial fluid aspiration. Chesterfield et al.13 isolated A. aphrophilus from a wash out of an infected left hip prosthesis in 2008.

Osteomyelitis, secondary to A. aphrophilus, has been encountered more frequently. Vertebral involvement constitutes the majority and these cases have been variably associated with oral cavity pathology or recent dental manipulation.6,14 Interestingly there was no such recent history in our patient.

Anti-microbial therapy

A number of anti-microbials have been used to treat infections caused by A. aphrophilus. Historically, penicillin therapy was first line; however, due to concern regarding β-lactamase production a third generation cephalosporin is now recommended for the treatment of serious infections.15,16 Other agents utilized successfully include gentamicin, macrolides and fluoroquinolones.6,14 Disk diffusion for assessment of anti-microbial susceptibility of A. aphrophilus has been reported to be unreliable, and MIC testing is the preferred method as outlined above.

Conclusion

We describe an unusual presentation of septic arthritis and osteomyelitis, caused by an organism rarely isolated from clinical specimens. The indolent nature of presentation and the low-grade findings on initial imaging raised the possibility of an atypical infecting organism of reduced virulence. This characteristic of A. aphrophilus as an ‘opportunist’ pathogen, combined with its fastidious nature and stringent requirements for isolation in the laboratory, may partly account for the low frequency with which A. aphrophilus is encountered in clinical practice. Central to an optimal clinical outcome in cases such as these is prompt diagnostic evaluation (prior to anti-microbial therapy), and meticulous laboratory methodology.

Conflict of interest: None declared.

References


