CASE REPORT

Kingella kingae sternoclavicular osteoarthritis

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SUMMARY

Sternoclavicular arthritis is an unusual osteoarticular infection and can be associated with severe complications. Cases in a paediatric population are infrequently reported, making this approach challenging. *Kingella kingae* is an agent of increasing recognition in paediatric invasive infections, principally below 2 years of age. A case of *K. kingae* osteoarthritis in a 17-month-old child is described with a review of the literature.

BACKGROUND

Kingella kingae is a gram-negative coccobacillus, with an increasing role in paediatric invasive disease for the last 20 years.[1] Known as a fastidious bacterium, its prevalence in osteoarticular infections is believed to be underestimated as cultures seldom yield positive results.[2] However, recent and more advanced methods of culture as well as new polymer chain reactions (PCR)-based assays revealed it to be one of the major pathogens involved in osteoarticular infections in children <4 years of age.[2]

The sternoclavicular (SC) joint is an unusual site of infection and is associated with particular risk factors such as immunosuppression, diabetes, oncological disease, secondary sites of infection and intravenous drug use. In a case series, Ross and Shamsuddin reported only 180 cases between 1970 and 2004.[3] Few cases have been reported in the paediatric population, with the youngest being 7 years old by the time of the diagnosis.[4]

We present a rare case of *K. kingae* septic osteoarthritis in an otherwise healthy 17 months child.

CASE PRESENTATION

A 17-month-old overweight male child presented to the emergency department with a 5-day history of fever, limitation of the right superior limb movement and ipsilateral neck deviation. He had history of a fall onto his outstretched arms and an upper respiratory infection in the previous week. He had no personal or familiar medical relevant history.

Physical examination revealed right SC swelling with local inflammatory signs, conditioning right upper limb functional limitation. Laboratory analysis showed a white cell count within normal range, a blood C reactive protein (CRP) of 0.22 mg/dL and a sedimentation rate of 54 mm/1st hour. Two blood cultures were obtained. Initial plain radiograph of the affected limb was normal. Sternoclavicular ultrasound revealed a heterogeneous SC joint effusion, with thickening of the joint capsule and surface irregularities of the clavicle. MRI scan confirmed moderate volume joint effusion and bone marrow oedema of the clavicle and manubrium (figure 1).

The patient was admitted with a probable diagnosis of sternoclavicular osteoarthritis and therapy with IV cefuroxime was initiated. Prolonged incubated blood cultures did not yield any growth. Ultrasound-guided arthrocentesis was performed in the third day after admission, with no pathogen isolation from culture of the joint aspirate, but PCR with universal 16S ribosomal RNA primers detected DNA of *K. kingae*.

OUTCOME AND FOLLOW-UP

He evidenced a rapid clinical and analytical improvement, maintaining antibiotherapy with cefuroxime for 4 weeks (2 weeks IV and 2 weeks *per os*) with no surgical approach.

He has made a good functional, analytical and radiologic recovery to date.

DISCUSSION

Sternoclavicular arthritis is an uncommon infection, accounting for <1% of all osteoarticular infections.[3] The mean age of presentation is approximately 45 years old,[3] with very few cases reported in the paediatric population. Some cases of sternum, manubrium-xiphoidal and sternocostal infection have been reported in infants and toddlers,[5] although to our knowledge this is the first reported case affecting the sternoclavicular joint in this age range.

The anatomic proximity of this joint to vital structures like the great vessels, trachea, oesophagus, mediastinum, vagus and phrenic nerves can lead to serious complications like empyema and mediastinitis,[6] requiring prompt intervention. Its superficial position could lead to an early recognition. However, the intra-articular disk and the strong joint capsule tend to slow the build-up of the effusion delaying its presentation,[7] probably explaining the high likelihood of concomitant osteomyelitis (69%).[3] In this case, mild joint swelling in an overweight child was unnoticed by the parents, perhaps delaying the presentation and leading to the associated manubrium osteomyelitis.

Because the plain radiography infrequently shows alterations, CT or MRI should always be obtained to confirm the diagnosis and exclude complications. When performed by an experienced professional, ultrasound can be useful in diagnosis and to assist fluid aspiration in a minimal invasive approach.[7]

K. kingae is a commensal oropharyngeal pathogen in children, and it is postulated that musculoskeletal infections may follow a period of bacteraemia associated with a breach in the respiratory epithelium, after an often-reported preceding upper airway

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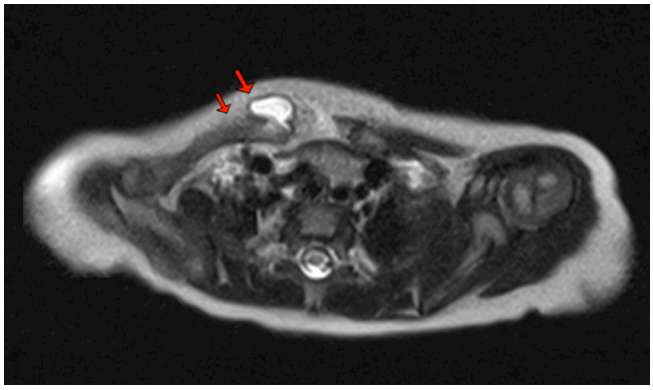

Figure 1 MRI showing SC joint effusion and bone marrow oedema of the clavicle and manubrium. SC, sternoclavicular.

infection.[2] In this case, the previous upper respiratory infection may have triggered a bacteraemia that allowed pathogen seeding into a previously traumatised SC joint.

The clinical presentation of *K. kingae* articular infections is usually subtle, less pronounced than with other common pathogens like *Staphylococcus aureus*.[2] Also, blood parameters are generally equivocal. William *et al* reported a total of 27 *K. kingae* osteoarticular infections with a mean CRPof 2.4 mg/dL.[2] Therefore, although *S. aureus* has been identified as the most common pathogen of SC arthritis,[3] the patient's age, previous upper airway infection, non-toxic appearance and blood parameters, suggested *K. kingae* as the offensive pathogen.

Synovial fluid was inoculated into an aerobic blood culture phial, as it can significantly improve the yield of cultures.[1] Also, we highlight the importance of collecting sample before antibiotic therapy. Even so, in order to prevent severe complications related to the location of the infection, prompt empirical antibiotherapy was initiated without previous synovial cultures. Therefore, a second tube sample was obtained for PCR-based assays. These methods not only enhance the identification of fastidious pathogens, but also allow their detection in synovial fluid up to 6 days after initiation of antibiotics.[8] The use of 16 s rRNA allows identification of multiple micro-organisms and has been proven useful in the work-up of paediatric septic arthritis.[9]

Difficulties in sampling synovial fluid from a sternoclavicular joint have been described in adults due to the paucity of fluid and the presence of an intra-articular disk.[7] In this case of a 17-month overweight child, this process was one of the major difficulties, as the small size of the joint enabled us to collect only 0.5 mL of effusion.

Because of the rarity of SC arthritis, its optimal therapy has not yet been defined.[6] If image studies show limited disease even with concomitant osteomyelitis, minimal invasive aspiration with medical therapy may be successful. However, if extensive bone destruction, mediastinitis or other local complications are present, joint resection is indicated.[7] The risks associated with this procedure and the absence of reports in children justified our attempt for conservative management despite some reports of recurrence with this method.[6]

Early transition to oral antibiotic therapy in osteoarticular infections is increasingly advocated nowadays.[10] Nevertheless, attending to the risk of severe complications like mediastinitis and the absence of reports in children, we decided to maintain intravenous therapy for 2 weeks, despite the rapid clinical improvement.

Learning points

- In conclusion, we describe an uncommon site of osteoarticular infection (OAI), even more unusual in the paediatric population.
- The risk of serious complication and lack of studies regarding this pathology in children made its approach challenging.
- The age, anamnesis, clinical status and blood parameters should support empirical antibiotherapy with *K. kingae* coverage, after collecting samples for microbiological study.
- Finally, we underline the importance of routine use of molecular methods when the diagnosis of *K. kingae* osteoarticular infection is likely.

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