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CASE REPORT

Subacute osteomyelitis due to *Staphylococcus caprae* in a teenager: A case report and review of the literature

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Abstract

BACKGROUND

Staphylococcus caprae (*S. caprae*) is a human commensal bacterium which can be detected in the nose, nails, and skin. It can be responsible for heterogeneous infections such as bacteremia, endocarditis, pneumonia, acute otitis externa, peritonitis, and urinary tract infections. Bone and joint infections due to *S. caprae* have also been reported, but most of them resulted from the infection of orthopedic devices, especially joint prostheses and internal osteosynthesis devices. Rare cases of primary osteoarticular infections caused by *S. caprae* have been described, including osteitis, arthritis, or spondylodiscitis.

CASE SUMMARY

We report an unusual case of subacute osteomyelitis in a toe phalanx caused by *S. caprae* in a 14.5-year-old girl.

CONCLUSION

Subacute *S. caprae* osteomyelitis is a little-known and probably underestimated community-acquired infectious disease. This microorganism's pathogenicity should be seen as more than a classic nosocomial orthopedic device infection.

Key Words: Subacute; Osteomyelitis; *Staphylococcus caprae*; Teenagers; Case report

Core Tip: *Staphylococcus caprae* (*S. caprae*) is a human commensal bacterium which can be detected in the nose, nails, and skin. It may be responsible for heterogenous infections such as bacteremia, endocarditis, pneumonia, acute otitis externa, peritonitis, and urinary tract infections. Bone and joint infections due to *S. caprae* have also been reported but most of them resulted from orthopedic device infections, including above all joint prosthesis infections and internal osteosynthesis device infections. Only any rare cases of primary osteoarticular infections caused by *S. caprae* have been described and consisted in osteitis, arthritis, or spondylodiscitis. We report here the case of an unusual subacute osteomyelitis of a toe phalanx caused by *S. caprae* in a 14.5-year-old girl.

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INTRODUCTION

Coagulase-negative staphylococci (CoNS) are ubiquitous human and animal commensals and form an integral part of healthy human skin microbiota. CoNS are frequently discovered in clinical samples and often considered contaminants because they can become opportunistic pathogens in certain situations. CoNS associated with nosocomial infection are typically characterized by their pronounced antimicrobial resistance, including methicillin-resistant and multidrug-resistant isolates. However, they do not have as much pathogenic potential as coagulase-positive staphylococci such as *Staphylococcus caprae* (*S. caprae*).

S. caprae is a commensal coagulase-negative *Staphylococcus* known to colonize the skin and mammary glands of goats [1], occasionally causing mastitis[2]. In humans, commensal *S. caprae* can be detected in the nose, nails, and skin[3,4], and it can be the initial cause of heterogeneous infections such as bacteremia[4-13], endocarditis[6], pneumonia[7], acute otitis externa[3,5], peritonitis[14], and urinary tract infections[6,15]. Bone and joint infections due to *S. caprae* have been reported, but, fortunately, they remain rare[3,8,16-27]. Most osteoarticular *S. caprae* infections are the result of infected orthopedic devices, especially infected joint prostheses[3,8,17,18,21-27] and internal osteosynthesis devices[17,19,27]. Only very rare cases of primary osteoarticular *S. caprae* infections have been described, including osteitis[3,8], arthritis[16], or spondylodiscitis[27,28]. It is commonly accepted that the prevalence of human *S. caprae* infections is underestimated since conventional phenotypic identification systems incorrectly identify many *S. caprae* strains[3,27,28]. Molecular techniques have improved their identification[9,14,17,20,29,30], becoming essential when standard cultures give negative results[25]. We report a rare and unusual case of subacute osteomyelitis caused by *S. caprae* in a toe phalanx of a 14.5-year-old girl.

CASE PRESENTATION

Chief complaints

A 14.5-year-old girl was referred to our department by her pediatrician due to persistent pain in her fourth right toe for over 3 mo.

History of present illness

Persistent pain in the fourth right toe for over 3 mo.

History of past illness

There was no past illness for this patient.

Personal and family history

There was a history of trivial trauma but no suggestion of broken skin or fever.

Physical examination

At admission, the patient was apyrexial; on examination, the toe showed mild swelling and erythema, and palpation caused discomfort. The girl could freely bear weight on her foot with no limitations.

Laboratory examinations

The patient's white blood cell count was 7600 cells/mm^3, C-reactive protein was less than 0.3 mg/L, and her erythrocyte sedimentation rate was 7 mm/h.

Imaging examinations

An initial plain radiograph revealed a constitutional fusion of the toe's distal interphalangeal joint but without any

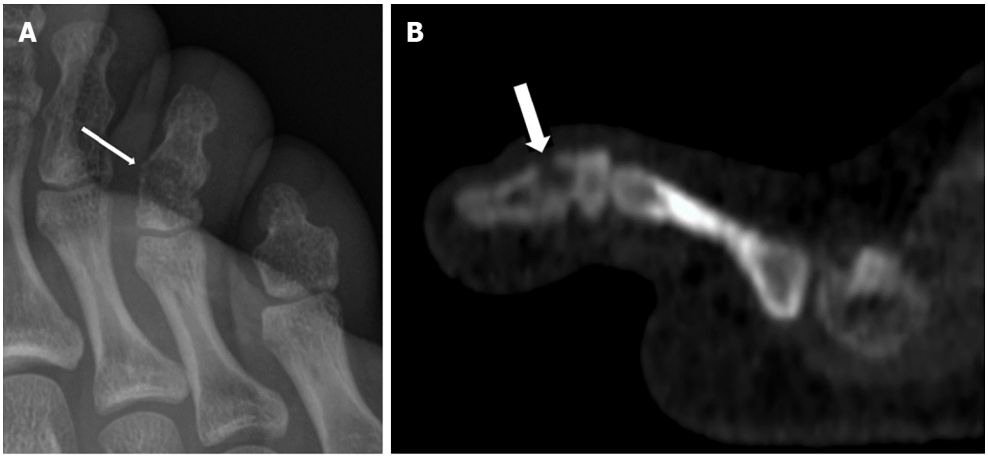

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Figure 1 Imaging. A: A plain radiograph performed after 2 mo of follow-up revealed an ill-defined lytic lesion of the fourth toe's fused distal phalanx (white arrow); B: A computed tomography scan revealed an eccentrically-located lytic focus of the phalangeal metaphysis that was eroding the dorsal cortex (white arrow).

relevant pathology.

FINAL DIAGNOSIS

Toe phalanx lytic lesion.

TREATMENT

The patient was administered oral antibiotics (co-amoxicillin), and this treatment continued for 15 d, although a full resolution of symptoms was achieved after only a few days of treatment, and inflammatory markers remained normal. Three months after surgery, the toe phalanx lytic lesion was completely resolved.

OUTCOME AND FOLLOW-UP

After 2 mo, a new plain radiograph revealed a subtle, ill-defined lytic lesion of the toe phalanx (Figure 1A). A complementary computed tomography (CT) scan of the foot confirmed a lytic lesion with cortical erosion of the metaphysis of the fused toe phalanx (Figure 1B). We performed a direct open biopsy of the toe lytic lesion, involving the debridement and curettage of the pathologic tissue, which resulted in a limited bone defect (3-4 mm). Microbiological cultures of the material removed were made on a solid medium, and matrix-assisted laser desorption/ionization time-of-flight mass spectrometry was used to identify the bacteria, revealing *S. caprae*.

DISCUSSION

S. caprae is a commensal coagulase-negative *Staphylococcus* that may become a human pathogen in community-acquired or nosocomial infections. Most osteoarticular *S. caprae* infections result from infected orthopedic devices, especially joint prostheses[3,8,17,18,21-27] and internal osteosynthesis devices[17,19,27]. Genome analysis has demonstrated that *S. caprae* is closely related to *S. epidermidis* and *S. capitis* at the species level, especially in its ability to form biofilms, which may explain the virulence of *S. caprae* infections[31]. The formation of a biofilm is considered an essential step in the pathogenesis of CoNS. Another important step in the induction of an infection is the adhesion of bacterial cells to host tissues and their ability to grow into a biofilm[32]. The genetic determinants of biofilm formation include the *icaADBC* operon, which codes for the biosynthetic enzymes involved in producing polysaccharide intercellular adhesin[33,34]. *S. caprae* expresses the *ica* operon providing the pathogen with the ability to form a biofilm on orthopedic osteosynthesis devices, thus conferring the bacterium with resistance to the immune system and antibiotics[17,35].

Despite this, *S. caprae* has never been reported to cause subacute osteomyelitis. The present case is thus the first to show that *S. caprae* can be responsible for subacute osteomyelitis even when no orthopedic device is present. Subacute osteomyelitis is an osseous infection with a duration of more than 3 wk without acute symptoms. Subacute osteomyelitis may result from the inadequate treatment of acute osteomyelitis or may occur in settings displaying strong host resistance to infection, an illness due to less virulent organisms, prior exposure to antibiotics, or a combination of all these factors[36,

37]. We hypothesize that *S. caprae* was one of the few virulent pathogens that could have become an opportunistic pathogen in this case, but the subject managed to maintain it relatively well-controlled. Indeed, *S. caprae* is recognized as being less virulent than *S. aureus* and other CoNS[27]. Nevertheless, *S. caprae* has the bacterial characteristics required for the development of subacute osteomyelitis.

CONCLUSION

Subacute *S. caprae* osteomyelitis is a little-known and probably underestimated community-acquired infectious disease. This microorganism's pathogenicity should be seen as more than a classic nosocomial orthopedic device infection. *S. caprae* is closely related to *S. epidermidis* and *S. capitis* at the species level, especially in its ability to form biofilms, which may explain the virulence of these pathogens. The difficulty in detecting *S. caprae* is attributable to the fact that conventional phenotypic identification systems still misidentify it. *S. caprae* should therefore be included in the list of organisms that can cause subacute osteomyelitis, such as *S. aureus*, *Kingella kingae*, *Salmonella* and *Streptococcus species*, and *Mycobacterium tuberculosis*.

FOOTNOTES

Author contributions: Vazquez O analyzed the data and wrote the manuscript; and all authors have read and approved the final manuscript.

Informed consent statement: We, the authors, consent to the publication of identifiable details, which can include photographs and/or videos and/or case history and/or details within the text ("Material") to be published in the above Journal and Article.

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