CASE REPORT

Skull osteomyelitis as a rare complication of cat scratch disease

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ABSTRACT

Bartonella henselae, the causative agent of cat scratch disease (CSD), is one of the most common causes of regional lymphadenitis in children. Other less common manifestations of *B. henselae* infection including fever of unknown origin, neuroretinitis, and osteomyelitis are being increasingly recognized. We describe a 3-year-old female with a recent history of typical CSD involving lymph nodes who developed osteomyelitis of the skull, a very rarely recognized complication of this infection.

Key words: *Bartonella henselae*, cat scratch disease, osteomyelitis

INTRODUCTION

Bartonella henselae, the causative agent of cat scratch disease (CSD), is one of the most common causes of regional lymphadenitis in children, usually resulting in mild, self-limited infection. However, cat scratch disease has a broad spectrum of presentations in healthy children, including fever of unknown origin (with multiple hepatosplenic abscesses), neuroretinitis, osteomyelitis, and encephalitis.[1,2] Serologic testing usually confirms the diagnosis of *Bartonella* infections, but both false positive and false negative results have been reported. New diagnostics, including nucleic acid amplification tests such as polymerase chain reaction (PCR), have allowed confirmation of *B. henselae* infection in patients with atypical manifestations.[3] In this report, we describe a 3-year-old female with a recent history of typical CSD lymphadenitis who subsequently developed osteomyelitis of the skull, a very rarely recognized complication of this infection.

CASE REPORT

A 3-year-old Caucasian female who was recently diagnosed and treated for CSD presented to the emergency department for a 2-week history of worsening scalp lump with redness. The lesion was in the left frontal region. She did not have a fever or any other systemic symptoms. Two weeks earlier, she finished a 5-day course of azithromycin for suspected CSD lymphadenitis. At that time, she had presented with right side inguinal and preauricular lumps, measuring 2 cm and 1 cm, respectively. Both lumps were tender and erythematous. The patient had no significant medical history. Her growth and development were appropriate for her age. Her immunizations were complete for her age. She lived with her parents in Flint and had never traveled outside Michigan. The family had one dog and two kittens at home. Her mother reported that the kittens scratched the patient frequently.

On admission, the patient was alert, interactive, and in no distress. She was afebrile, and her vital signs were normal. Her examination was significant for a 4 cm × 4 cm lump located in the left frontal area. There was mild erythema overlying the lesion, but it was not tender. There was a small area of fluctuance at the center. A 1 cm × 1 cm preauricular lymph

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node was palpable on the right. A 2 cm area of induration and hyperpigmentation was noted in the right groin with no drainage. Skin examinations showed no other lesions. Her abdominal examination revealed no organomegaly and the remainder of the physical examination was normal.

The patient's white blood cell count was 5600/mm^3, with 48% neutrophils, 44% lymphocytes, 5% monocytes, and 3% eosinophils. The hemoglobin was 9.6 g/dL, and the platelet count was 389,000/mm^3. Aspartate aminotransferase and alanine aminotransferase were 12 and 24 units/L, respectively. The C reactive protein was 3.5 mg/dl and the erythrocyte sedimentation rate was 44 mm/h. Rapid plasma reagin and human immunodeficiency virus antibody by enzyme-linked immunosorbent assay were negative. A tuberculin skin test had 0 mm of induration. *B. henselae* immunoglobulin M and immunoglobulin G titers, by indirect florescent antibodies, were elevated at 1:32 and 1:512, respectively.

Skull radiography revealed that a focal irregular lucency involving the left frontal bone [Figure 1] and computed tomography of the head showed a focal soft-tissue lesion and a destructive process affecting the adjacent bone in the left frontal region. Magnetic resonance imaging of the head revealed septated scalp swelling with a destructive process involving adjacent calvaria with intracranial extension [Figure 2]. Pediatric neurosurgery performed a left frontal craniotomy and described purulent material on opening the skin, along with the erosion of the left frontal bone. Tissue microscopic examination showed chronic granulomatous tissue. The examination of the aspirate with a Warthin–Starry stain showed multiple clusters of organisms [Figure 3]. Aerobic, anaerobic, fungal, and mycobacterial culture were performed; a few colonies of *Staphylococcus epidermidis* grew in aerobic culture (considered a contaminant), and other cultures were negative. Finally, PCR from the skull abscess aspirate was positive for *Bartonella* species. No further speciation was performed, and culture for *Bartonella* culture was not attempted. The patient received 6-week treatment course with azithromycin and rifampin therapy with complete healing at the site of the infection.

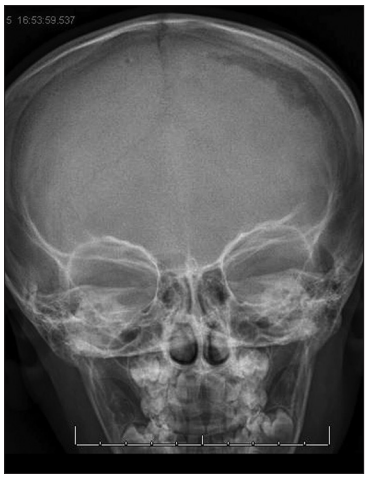

Figure 1: Skull X-ray showing lytic lesion in the left frontal bone (original)

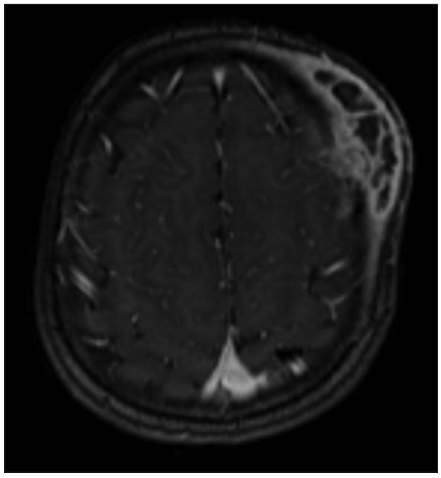

Figure 2: Septated scalp swelling measuring 5.5 cm × 2.4 cm × 1.7 cm within the left frontal region with a destructive process involving adjacent calvarium (original)

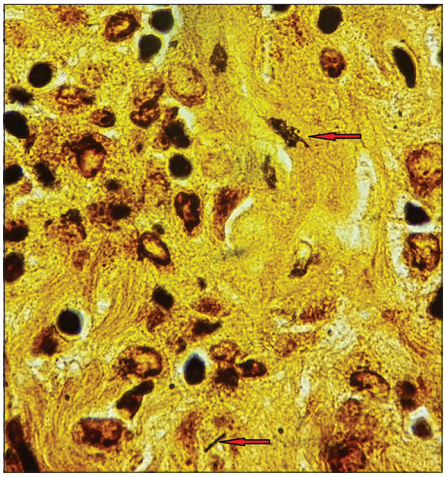

Figure 3: Warthin stain showing clumps of rod-shaped organisms (original)

DISCUSSION

Although rare, osteomyelitis is a known complication of CSD, with the vertebral column and pelvic girdle being the most commonly reported sites.[4] Skull osteomyelitis has been reported previously as a complication of *Bartonella* infection, but is exceedingly rare. A literature review by Hajjaji *et al.* before 2007 found 47 reported cases of osteomyelitis due to *B. henselae*, with 4 of those involving the skull.[4] In this review, patients with *Bartonella* osteomyelitis usually had a subacute presentation with mild constitutional symptoms. Fever was present in 78% as part of their initial presentation, and most cases involved the axial skeleton.[4,5]

Since 2007, skull involvement was noted in just 1 of the 14 cases reported by Puri *et al*.[5] The frontal bone, parietal bone, and mastoid bone have all been reported to be involved.[4-6] In addition, one other child presenting with lytic skull lesion involving the forehead after having cervical nodal disease has been reported.[7] Overall, the clinical presentation of skull *Bartonella* osteomyelitis resembled that of *Bartonella* infection involving other bones. The skull infection had subacute or chronic course, associated lymph node disease, and need for debridement.[7] Nevertheless, the reported long-term prognosis has been excellent, with complete recovery.[4,7] Similarly, our patient had complete healing and recovery, consistent with the good outcome reported in other patients with *Bartonella* osteomyelitis.

The pathogenesis of *Bartonella* osteomyelitis is poorly understood, but both direct extension from infected lymph nodes (contiguous infection) and distant infection (presumably from hematogenous or lymphohematogenous spread) have been reported.[4,5,7] Eight of the 14 cases reported by Puri *et al*. tested positive for *Bartonella* by PCR at the site of the infection.[5] The hematogenous spread seems likely in our patient, who presented with inguinal and R-sided preauricular adenopathy and later developed osteomyelitis of the left frontal bone.

Spontaneous clinical improvement is expected in most cases of classical CSD. Thus, it is not surprising that the review by Hajjaji *et al*. concluded that some cases of *Bartonella* osteomyelitis have resolved despite treatment with antibiotics lacking activity against this organism.[4] On the other hand, the poor response to antibiotic treatment and the progression despite treatment in other reported cases suggests that therapy must be individualized.[7] The role of antibiotic therapy of usually self-limited CSD in normal hosts is not well defined, while the evidence for benefit in immunocompromised patients with *Bartonella* infection is much stronger. Antibiotics that are considered effective against CSD include azithromycin, rifampin, ciprofloxacin, trimethoprim-sulfamethoxazole, and parenteral gentamicin. In patients with a hepatosplenic CSD with prolonged fever, rifampin therapy reportedly has been effective.[8] Gentamicin in combination with doxycycline is recommended as a treatment regimen for endocarditis. For musculoskeletal disease, most cases are managed with a combination of multiple courses of antibiotics and surgery. The optimal approach is yet to be determined.[4,7]

CONCLUSION

Our case highlights the importance of considering CSD in the differential of lytic skull lesions. Although rare, CSD may cause lytic skull lesions, which poses a diagnostic challenge, as immunologic, neoplastic, and other infectious etiologies must be ruled out.

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Conflicts of interest

There are no conflicts of interest.

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