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## **Case report**

A 30-year-old male with a remote history of a right shoulder superior labrum anterior to posterior lesion, treated operatively at an outside hospital, presented with a 3-month history of right shoulder pain. Initial shoulder radiographs were performed and appeared to show no abnormality (Fig. 1). His pain continued for another month, and he also developed painful lymphadenopathy. At that point, a noncontrast magnetic resonance imaging (MRI) was obtained (Fig. 2). This showed a destructive lesion in the scapular neck with communication through the articular surface of the glenoid. There was concern for a malignant process, and a positron emission tomography scan and repeat contrast-enhanced MRI were obtained (Figs. 3 and 4). This showed increased uptake in several lymph nodes. An inguinal lymph node biopsy was performed, tissue was sent for histology, and a referral was made to the orthopedic oncology service due to concern for skeletal malignancy. Our orthopedic oncologists did not feel the imaging was consistent with a metastatic process or a primary bone tumor. Due to the patient's history of previous labral pathology, it was felt to be more consistent with a paralabral cyst or postsurgical changes, and a referral was made to the sports medicine service. Around this time, the initial biopsy histology revealed granulomatous inflammation. Bartonella henselae titers were drawn and found to be significantly elevated, at 1:1280 on July 15, 2021. On the same day that the titers came back positive, he was started on oral rifampin and azithromycin for 4 weeks, until he was taken to the operating room. He remained on these antibiotics for 4 more weeks after surgery, for a total of 8 weeks. Given the combination of lymphadenopathy, elevated B henselae titers, and abnormal appearance of the glenoid

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lesion on MRI, this was felt to be a case of *B* henselae osteomyelitis. A computed tomography scan was then ordered for preoperative planning and the decision was made to perform an arthroscopic irrigation and débridement of the glenoid lesion with possible bone grafting (Fig. 5).

The patient was taken to the operating room and placed in the lateral position with traction on the arm. After sterile prep, the arthroscope was inserted through a posterior portal and cloudy fluid was noted (Fig. 6). This was sent for laboratory analysis but did not return positive for Bartonella titers. Once the camera was inserted, the joint was noted to have extensive synovitis and fibrinous material was noted throughout the joint (Fig. 7). An anterior portal was established, and the capsule was elevated off the anterior glenoid neck, leaving the previous labral repair intact. A curved microfracture awl was used to probe into the cystic area, and purulent material was noted coming from the area of the cyst. This was also collected and sent for laboratory analysis. A drill and curette were then used to débride the cyst (Fig. 8). This material was collected and sent for Warthin-Starry stain, bacterial/fungal culture, and Bartonella polymerase chain reaction (PCR) testing. Bartonella PCR testing from surgical sample (August 24, 2021) was sent out for testing but the test was inconclusive due to the presence of competing inhibitors to the PCR in this specimen. Stains were negative.

He was then seen in clinic postoperative day 1 and was having no issues. He was given a prescription to begin physical therapy for gentle range of motion. At his second postoperative appointment, 2 weeks later, he noted improving pain and motion. He was continued on oral antibiotics from the infectious disease team. He was seen again at 6 weeks and reported 75% improvement in his symptoms. He did note some persistent pain, and a repeat MRI was ordered to look for recurrence of the lesion. The MRI showed a decrease in the size of the cyst and reduction of surrounding inflammation. He was seen once again at 3 months and reported near-complete resolution of his symptoms. By this time, the patient





Institutional review board approval was not required for this case report.

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Figure 1 Initial X-ray taken on March 17, 2021.



Figure 2 Noncontrast MRI showing lytic lesion in the glenoid neck taken on April 22, 2021. *MRI*, magnetic resonance imaging.

had finished his course of antibiotics and C-reactive protein/ erythrocyte sedimentation rate had normalized. At final follow-up, roughly 6 months later he had no complaints and was back to doing all his previous activities without any discomfort.

## Discussion

*B henselae* is a gram-negative bacteria responsible for cat scratch disease (CSD). An estimated 12,000-22,000 cases per year are diagnosed in the United States, with over 50% of cases involving children under 18 years of age.<sup>3,9</sup> As the name would imply, the main carrier of *B henselae* is feral and domesticated cats. The bacteria is transmitted between cats via fleas and ticks.<sup>8</sup> Infection in humans typically occurs through cat bites, scratches, or saliva coming in contact with open wounds. Contact with cats, most often kittens, is reported in 99% of cases, and a site of inoculation is reported 92% of the time.<sup>1</sup> On chart review there was no prior history of exposure to cats, the well-recognized host. Yet, approximately 5



Figure 3 Precontrast MRI taken on July 8, 2021. MRI, magnetic resonance imaging.



Figure 4 Contrast-enhanced MRI taken on July 8, 2021. MRI, magnetic resonance imaging.

months prior to his presentation with shoulder pain, the patient sustained open fractures to his 2nd-5th toes from a horse stepping on his bare foot. Although not the typical host, horses have been shown to be asymptomatic carriers of *B henselae* and this could have seeded an infection.<sup>4</sup> The most common symptoms include fever, lymphadenopathy, and a pustule at the site of a scratch.<sup>5</sup> In 80%-90% of cases CSD is self-limiting and resolves without treatment in 2-4 weeks; however, in rare cases disseminated disease occurs and may warrant treatment with antibiotics or surgical intervention.<sup>5</sup>

Diagnosis of CSD remains difficult. Culture of the organism is unreliable and may take up to 40 days to show growth.<sup>11</sup> Therefore, if taking cultures, holding them for up to 6 weeks is recommended. Alternatively, if a lymph node or other tissue is removed, it can be examined histologically using a Warthin-Starry silver impregnated stain.<sup>10</sup> In this case, the intraoperative cultures did not grow *B henselae*. The patient had been on antibiotics for several weeks before operative intervention, and even in the most ideal situations, the organism is difficult to culture. However, the most effective and widely used way to diagnose CSD is with serologic testing for antibodies against *B henselae* using enzyme immunoassay or indirect fluorescence method.<sup>6</sup> This patient did have positive titers, as well as the classic lymphadenopathy associated with CSD.

Although uncomplicated CSD does not require any treatment other than supportive measures, azithromycin has been shown in a

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Figure 5 Presurgical CT showing glenoid articular involvement taken on August 16, 2021. *CT*, computed tomography.



Figure 7 Fibrinous material seen at the entrance of the intraosseous abscess.



Figure 6 Murky fluid and fibrinous material encountered upon entry of the arthroscope.

prospective double-blinded trial to hasten the resolution of lymphadenopathy, without significant long-term differences between placebo. Significantly less data exist on the best treatment for disseminated disease; however, rifampin, ciprofloxacin, trimethoprim-sulfamethoxazole, and gentamicin are all effective in treating disseminated disease.<sup>8</sup>

*B* henselae osteomyelitis has been reported in the literature, although it is exceedingly rare. One series of 1200 patients reported just 2 (0.17%) with osteolytic lesions.<sup>1</sup> Another series of 1852 patients reported just 5 (0.27%) with osseous involvement.<sup>7</sup> The vertebral column and pelvic girdle make up nearly 70% of reported osseous lesions.<sup>2</sup> To our knowledge, there has never been a case report of *B* henselae osteomyelitis in the scapula. Although *B* henselae associated osteomyelitis can resolve with time and antibiotics, there are a few reports of operatively treated *B* henselae osteomyelitis.<sup>12,13</sup> These were most often in the spine, and due to concern for cord compression from an associated abscess. We elected to operatively treat this patient with arthroscopic irrigation and débridement due to the concern for continued articular cartilage damage from the osseous lesion, which had already begun to penetrate through the glenoid into the shoulder joint. We were



Figure 8 Decompressed intraosseous abscess after usage of microfracture awl and drill.

concerned that antibiotic treatment would not work quickly enough and being a young, active male, we did not want the patient's articular cartilage to be damaged.

#### Conclusion

*B henselae* osteomyelitis is a rare presentation of an otherwise self-limiting disease. It should be included in the differential diagnosis of a patient with tender lymphadenopathy and history of close exposure with cats. Biopsy of the inflamed lymph nodes may show granulomatous inflammation, and the diagnosis can be confirmed with serological testing. Uncomplicated cases can be treated with supportive measures, whereas disseminated cases may need antibiotics or operative treatment for resolution of symptoms. In this case report, we present the only known case of *B henselae* scapular osteomyelitis. More research needs to be performed to determine the best treatment options for osseous involvement of *B henselae* osteomyelitis.

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