Case Report

Dropped Head Syndrome in a Patient with *Aeromonas hydrophila*-induced Septic Arthritis of the Shoulder

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Abstract

Septic arthritis of the shoulder is very rarely caused by *Aeromonas hydrophila*. We present the case of a 45-year-old woman who presented with symptoms of painful shoulder for 2 weeks and dropped head for 1 week prior to admission. *A. hydrophila* was isolated from a culture of purulent synovial fluid. Magnetic resonance imaging revealed profuse abscess collection between the right infraspinatus muscle and trapezius muscle and swelling of the right and left paraspinatus muscles, which suggested myositis-related dropped head syndrome. After surgery with arthrotomy, function of the shoulder and neck extensors was significantly improved.

Keywords: *Aeromonas hydrophila*, dropped head syndrome, septic arthritis

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Introduction

Aeromonas hydrophila is a rare cause of septic arthritis. Shoulder is the third most common joint affected (approximately 3%–12% of cases) after knee and hip, with the dominant pathogen being *Staphylococcus aureus* (40%–50%).[1] To the best of our knowledge, there has been no previously reported case of *A. hydrophila*-induced septic arthritis of the shoulder. Dropped head syndrome (DHS) is characterized by severe weakness of neck extensor muscles, which results in chin-on-chest deformity. Numerous diseases are associated with DHS. It is a novelty to identify DHS caused by *A. hydrophila*-induced septic arthritis of the shoulder.

Case report

A 45-year-old woman presented to the emergency department with a 2-week history of swelling, warmth, and pain in the right shoulder accompanied by intermittent mild fever. One week before admission, she started presenting progressive weakness of the neck and eventually could not look straight ahead. Her surgical history was significant for arthroscopic repair of rotator cuff tear approximately one year ago. The initial surgery and postoperative course were uneventful. She had sustained multiple abrasion wounds on her limbs during a recreational trip to the river one month before admission.

On arrival, she had a temperature of 38.5°C, heart rate of 101 beats per minute, respiratory rate of 18 breaths per minute, and a blood pressure of 128/73 mmHg. On examination, the shoulder was swollen, tender and warm to the touch, with painful restricted range of movements. There was no discharge or open wound on the shoulder. She had profound neck flexion with limitation of active extension, but could passively raise her head with her hands (Figure 1a). Radiography of the right shoulder displayed retention of an anchor in the right greater tuberosity with adjacent soft tissue swelling (Figure 2). A clinical diagnosis of right shoulder septic arthritis was made, after which 20 mL of cloudy and purulent fluid was aspirated from the joint. Synovial fluid analysis showed a leukocyte count of 313,900/μL; 98% were neutrophils. Synovial fluid gram-stain examination revealed many leukocytes and gram-negative bacilli. The patient's serum white blood cell count was 29,040/μL (normal range, 3200–11,000/μL) with neutrophils predominance (95.2%), and her C-reactive protein level was 7.63 mg/dL (normal reference, 0.8 mg/dL).

Empirical treatment for septic arthritis was started on admission (vancomycin one gram intravenously for 12 hours/day and ceftazidime two grams intravenously for 8 hours/day). However, pyrexia and neck weakness with dropped head persisted. The synovial fluid culture yielded *A. hydrophila* susceptible to all antibiotics tested except ampicillin. Contrast-enhanced magnetic resonance imaging of the cervical spine disclosed a massive fluid collection between the right infraspinatus muscle and trapezius muscle, with swelling and edematous changes in the right and left paraspinatus muscles indicating myositis (Figure 3). Subsequently, the patient underwent open arthrotomy with debridement of the right shoulder. Approximately 500 mL of pus was extracted. A chest tube was inserted into the right shoulder for continuous drainage and irrigation with normal saline (approximately 3000 mL/day).

Three days postoperatively, her pyrexia was resolved and the ability to actively raise her head was noticeable (Figure 1b). Based on the Medical Research Council grading system for muscle strength, the muscle power of the neck extensor muscles improved from 1 (on admission) to 4 (after surgery), and swelling of the right shoulder alleviated significantly. The serum white blood cell count and C-reactive protein level was 10,430/μL and 0.73

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Figure 1. (a) The patient presented to our hospital with a dropped head and could not raise her head actively. **(b)** The function of the neck extensor significantly improved 2 days after arthrotomy with debridement of the right shoulder.

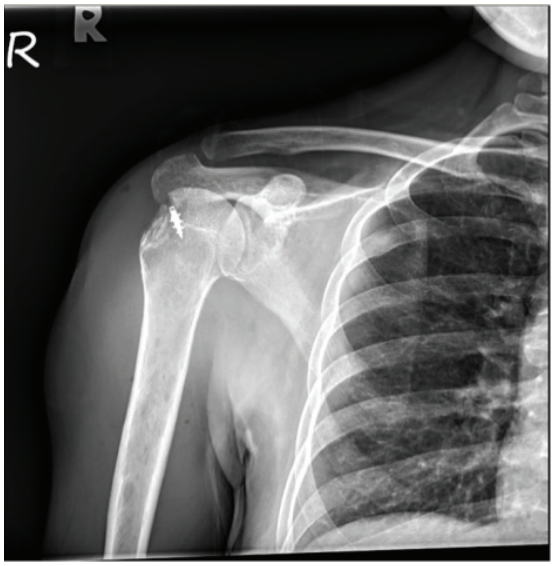

Figure 2. Radiography of right shoulder showed retention of an anchor in the greater tuberosity with adjacent soft tissue swelling.

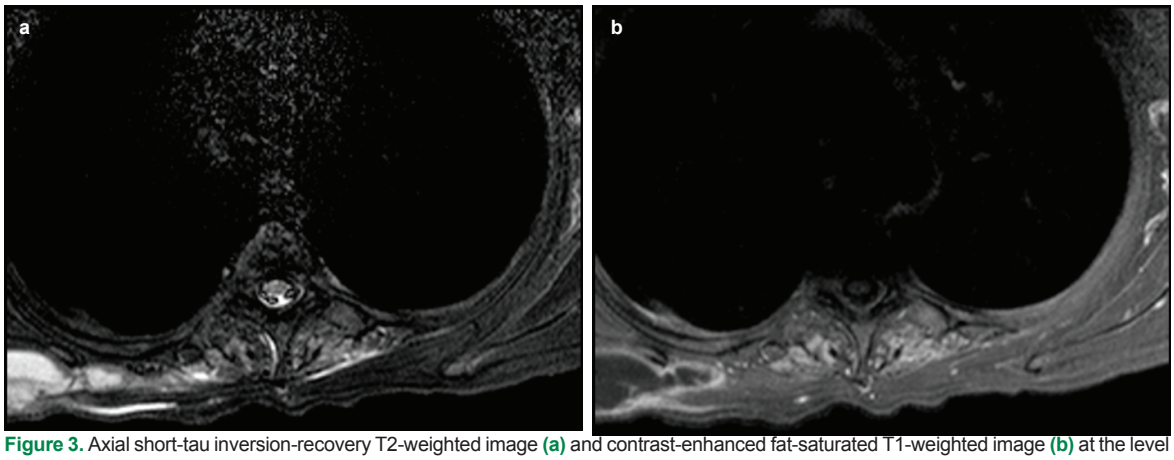

Figure 3. Axial short-tau inversion-recovery T2-weighted image **(a)** and contrast-enhanced fat-saturated T1-weighted image **(b)** at the level of upper chest show hyperintense lobulated fluid collection (arrows) and rim enhancement (arrowheads) involving the right infraspinatus muscle and trapezius muscle together with edematous changes and enhancement of the bilateral paraspinatus muscles.

mg/dL, respectively before discharge. The total hospitalization time was 11 days and she was discharged with oral ciprofloxacin 750 mg every 12 hours for 4 weeks. After 9 months, there was no relapse.

The patient provided informed consent for inclusion in this case report.

Discussion

A. hydrophila are ubiquitous environmental gram-negative bacilli living in fresh and brackish water and can lead to human infections following water-related injuries.[2] These bacteria are known to cause gastroenteritis, septicemia, and soft-tissue infection, ranging from mild pustular lesions to necrotizing fasciitis.[3] Joint infection caused by *A. hydrophila* has unfavorable prognosis. Elwitigala, et al., reviewed 7 cases of septic arthritis due to *A. hydrophila*, among which 3 patients expired, one developed necrotizing fasciitis with above-knee amputation, and another developed osteomyelitis of the hallux.[4] In our case, the septic arthritis was infected by *A. hydrophila* via fresh water injury with hematogeneous seeding to the shoulder. In addition, the presence of an implant in the patient's shoulder provided a shelter where microorganisms could grow in biofilms, rendering the infection difficult to eradicate.[5] It could be one precipitating factor for the development of dropped head due to disease progression.

DHS is characterized by severe weakness of the neck extensor muscles that belong to the paraspinal muscles. In DHS, there is flexibility in neck movement, unlike ankylosing spondylitis, in which the neck becomes rigid.[6] The prevalence of DHS in patients with motor neuron disease is approximately 1%.[6] The common causes of DHS are classified according to the site of impairment: (1) motor neuron (e.g., amyotrophic lateral sclerosis); (2) peripheral nerve (e.g., chronic inflammatory polyneuropathy); (3) neuromuscular junction (e.g., myasthenia gravis); and (4) muscle (e.g., inclusion body myositis, mitochondrial myopathy, and isolated neck extensor myopathy).[7] Other less common causes of DHS include hypothyroidism and scleromyositis.[7,8]

In 2012, a review on DHS by Sharan, et al., suggested that the semispinalis cervicis and capitis muscles may be primarily responsible for neck extension.[6] The magnetic resonance imaging of our patient exhibited myositis of the bilateral paraspinatus muscles, especially the semispinalis capitis and splenius capitis muscles. The patient's recovery after drainage of the massive pus accompanied by adequate antibiotic treatment and these image findings indicated that the DHS was caused by myositis of the paraspinatus muscles that had been infected by dissemination of the septic arthritis of the shoulder.

In conclusion, we report a rare case of *A. hydrophila*-related septic arthritis of the shoulder, in which the infection extended to the paraspinal muscles and caused DHS. Early diagnosis and treatment of septic arthritis are important to avoid disease progression, especially in patients with joint implant or prosthesis. DHS could be induced by infection contiguously spread to the neck extensor muscles.

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