Concurrent Atlantoaxial Septic Arthritis and Septic Thrombosis of the Ophthalmic Vein due to Staphylococcus aureus: A Case Report and Review of the Literature

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Atlantoaxial joint septic arthritis and superior ophthalmic vein thrombosis are uncommon manifestations of Staphylococcus aureus infection. A 68-year-old man presented with acute-onset neck pain and diplopia. Imaging studies revealed atlantoaxial septic arthritis and right superior ophthalmic vein thrombosis. Blood cultures grew methicillin-susceptible S. aureus. We review the literature describing these 2 uncommon manifestations of a common pathogen.

Keywords. atlanto-axial joint; diplopia; ophthalmic vein septic thrombosis; septic arthritis; Staphylococcus aureus bacteremia.

Of bacterial pathogens, Staphylococcus aureus remains among the most virulent, with a 30-day mortality rate for bacteremia around 20% [1, 2]. Even methicillin-susceptible isolates are challenging to treat, especially when they cause infections with atypical manifestations. Here, we describe a patient with 2 concurrent and uncommon presentations of methicillin-susceptible S. aureus (MSSA), atlantoaxial septic arthritis, and septic thrombosis of the ophthalmic vein without cavernous sinus thrombosis.

CASE REPORT

A 68-year-old man with hypertension and a remote history of total prostatectomy and radiotherapy for prostate cancer presented to the hospital with a 4-day history of worsening neck pain, right shoulder pain, and 1 episode of diplopia. He reported left wrist pain and swelling 3 weeks before presentation.

Within hours of admission, his temperature increased to 39.2°C; he was otherwise hemodynamically stable. His physical exam was significant for right eye ptosis, right cranial nerve VI palsy, and severe cervical tenderness and stiffness. His left wrist and right shoulder were swollen, erythematous, and tender, with a decreased range of motion. Laboratory values at admission included a white blood cell count of 14.8 × 10³/μL with 89% neutrophils, an albumin of 2.3 g/dL, a total protein of 8.3 g/dL, an erythrocyte sedimentation rate (ESR) of >120 mm/h, a C-reactive protein of >190 mg/dL (normal range, 1.0–10.0), and negative HIV serology. Blood cultures collected on admission grew MSSA. A transthoracic echocardiogram did not reveal any valvular lesions or vegetations. After initial magnetic resonance imaging (MRI) and a computerized tomography (CT) scan of the neck that were inconclusive, a whole-body ¹⁸fluorodeoxyglucose-positron emission tomography scan detected abnormal hypermetabolic activity in the atlantoaxial joint and in the right shoulder (Figure 1) but not in the left wrist or in or around the heart valves. An MRI of the brain and orbits revealed the presence of right superior ophthalmic vein thrombosis without evidence of cavernous sinus thrombosis. Consulting neurosurgeons recommended conservative management. After starting anticoagulation, the patient developed hematemesis. An upper endoscopy revealed esophagitis and gastric ulcers; because of these findings, the patient did not receive a transesophageal echocardiogram or further anticoagulation.

Figure 1. Images obtained using a ¹⁸fluorodeoxyglucose-positron emission tomography/computed tomography scan showing abnormal hypermetabolic activity in the (A) atlanto-axial joint and (B) right shoulder. Images obtained via magnetic resonance imaging indicated a thrombus, detected as (C) an area of hyperintensity within the right superior ophthalmic vein before gadolinium (arrow) and (D) the absence of contrast within the right superior ophthalmic vein after gadolinium.
### Table 1. Summary of Previously Reported Cases of Atlanto-Axial Septic Arthritis and Superior Ophthalmic Vein Thrombosis Without Cavernous Sinus Thrombosis

| Characteristics                      | AASA (n = 6) | SOVT (n = 14) | AASA and SOVT (n = 1)¹ | References |
|--------------------------------------|--------------|---------------|------------------------|------------|
| Average age (range), y               | 62 (53–76)   | 54 (20–80)    | 68                     |            |
| Maleb                               | 5 of 6       | 9 of 14       | 1                      |            |
| Pathogen                             |              |               |                        |            |
| *Staphylococcus aureus*              | 5            | 3             | 1                      | [3–7, 9, 10, 15] |
| Otherc                              | 1            | 1             |                        | [8, 11]    |
| Unknown or not specified             | 10           |               |                        | [12–14, 16–21, 23] |
| Concomitant sites of infectiond      |              |               |                        |            |
| Bacteremia                          | 2            | 4             | 1                      | [3, 4, 9, 10, 15] |
| Dental                              | 1            | 3             |                        | [2, 12, 17, 18] |
| Orbital or facial cellulitis         | 6            |               |                        | [10–12, 14, 15, 23] |
| Sinusitis                           | 5            |               |                        | [9, 13, 14, 16, 18] |
| Intraorbital abscess                | 2            |               |                        | [11, 13]   |
| Retropharyngeal abscess             | 1            |               |                        | [6]        |
| Indeterminate or unspecified        | 2            | 3             |                        | [5, 8, 19–21] |
| Invasive proceduresb                |              |               |                        |            |
| None                                | 1            | 7             |                        | [3, 12, 14–19, 21] |
| Minor procedurec                    | 1            | 1             |                        | [7, 12]    |
| Major proceduref                    | 4            | 5             | 1                      | [4–6, 8–11, 13, 20] |
| Length of therapyg                  |              |               |                        |            |
| 7–14 d                               | 3            |               |                        | [11, 14, 16, 21] |
| 6–12 wk                             | 6            | 3             | 1                      | [3–10, 12, 18] |
| Outcomes¹                           |              |               |                        |            |
| Recovered                           | 5            | 10            |                        | [4–10, 13, 14, 16–21] |
| Improved with sequelaeh             | 1            | 2             | 1                      | [3, 11, 12] |
| Deceased                            | 1            |               |                        | [15]       |

Abbreviations: AASA, atlanto-axial septic arthritis; SOVT, superior ophthalmic vein thrombosis.

¹This case report.
²Unspecified [23].
³*Streptococcus anginosus* for AASA; *Serratia marcescens* and a coagulase-negative *Staphylococcus* species for SOVT.
⁴Some individuals with SOVT had more than 1 concomitant infection.
⁵Joint aspiration for AASA; dental extraction for SOVT.
⁶Intraoperative abscess drainage, laminectomy, joint stabilization AASA; orbital abscess drainage, sinus drainage, dacryocystorhinostomy, excision of adenoid cystic carcinoma for SOVT.
⁷Several reports did not specify length of therapy; 1 patient died during treatment [15]; some longer courses were intravenous, followed by oral medications.
⁸Some restriction of neck rotations for AASA; visual loss, diplopia, permanent unilateral blindness for SOVT; neck pain, diplopia, and cranial nerve VI palsy for this case report.
⁹Due to retroperitoneal bleed; the patient received anticoagulation as part of her therapy for SOVT.

With the identification of MSSA, his antibiotics were narrowed to cefazolin. The patient continued to have persistent fever that resolved several days after surgical debridement of his right shoulder. Conservative management of his atlantoaxial septic arthritis was unsuccessful. During a prolonged course of intravenous cefazolin, he developed worsening right arm weakness and numbness with pain. A repeat MRI obtained 7 weeks after the previous cervical MRI showed worsening erosion of the odontoid pannus and widening of the transverse atlantal ligament, for which he underwent arthrodesis from C1 to C4 to address atlanto-axial instability. He continued to receive intravenous cefazolin for the next 6 weeks, followed by long-term suppression with oral cephalaxin.

**DISCUSSION**

To the best of our knowledge, this is the first report describing the concurrent presentation of atlantoaxial septic arthritis and septic thrombosis of the ophthalmic vein. For our patient, we suspect that a left wrist infection led to the *S. aureus* bacteremia with subsequent development of 2 rare manifestations of infection. Table 1 summarizes previous reports in the English literature between 1991 and October 2018 describing atlantoaxial septic arthritis (n = 6) and septic ophthalmic vein thrombosis without cavernous sinus thrombosis (n = 14) in adults.

Previous reports of atlantoaxial septic arthritis detailed clinical presentations of subacute to acute onset of neck pain with fever, 5 of which were due to *S. aureus* [3–7]. Two cases presented with bacteremia [3, 4], 1 with a dental infection [7] and 1 with a retropharyngeal abscess [6]. The source of infection was indeterminant in the remaining 2 cases [5, 8]. Four of the cases required a surgical intervention involving the cervical spine [4–6, 8]. All patients received at least 6 weeks of antibiotics and clinically improved.
Septic thrombosis of the superior ophthalmic vein without involvement of cavernous sinus is also rare. The most common feature described in previously reported cases of septic superior ophthalmic vein thrombosis was limited extraocular movements (n = 13) [9–21]. The abducens nerve, or cranial nerve VI, courses through the central part of the superior orbital fissure, running lateral to the superior ophthalmic vein, and thus thrombosis of this vein can lead to direct compression of the abducens nerve, which innervates the lateral rectus muscle [22]. Other presenting symptoms included pain (n = 11), periorbital swelling (n = 10), proptosis (n = 9), and visual deficits (n = 8). Eleven of the 14 cases reported that the SOVT developed in conjunction with another head and neck infection, with cellulitis of the face or orbit (n = 6), sinusitis (n = 5), and dental infection (n = 3) being most common [9–18, 23]. S. aureus was identified as the pathogen in 3 cases [9, 10, 15] and Serratia marcescens and a coagulase-negative Staphylococci spp. were recovered from a fourth case [11]. Six cases required surgery or an interventional procedure for source control [9–13, 20]. Ten cases recovered completely, and 2 cases improved but with permanent loss of vision in the affected eye [11, 12]. One patient died due to a retroperitoneal bleed that likely occurred as a consequence of anticoagulation started to treat her septic thrombosis of the ophthalmic vein [15].

Overall, the prognosis for isolated superior ophthalmic vein thrombosis, including septic and aseptic causes, appears favorable, with mortality around 5% and long-term loss of visual acuity affecting 12% of individuals [17]. In their review, van der Poel et al. concurred with suggestions from other authors that superior ophthalmic vein thrombosis may progress to cavernous sinus thrombosis, which has a mortality rate of around 30%, even in the postantibiotic era [17, 24]. Although anticoagulation may reduce morbidity and mortality for people with cavernous sinus thrombosis, its role for treating superior ophthalmic vein thrombosis is less clear. In our review, 7 cases received anticoagulation, with 1 fatal outcome attributed to a retroperitoneal bleed [15] while 6 other case reports described anticoagulation including with heparin, low-molecular weight heparin, and aspirin without sequelae [11, 12, 14, 16, 17, 20]. The decision to start anticoagulation for individuals with septic superior ophthalmic vein thrombosis is still best considered on a case-by-case basis, taking into account age, comorbid conditions, response to antibiotic therapy, and whether invasive procedures were performed.

CONCLUSIONS

Even with early recognition and prompt initiation of appropriate antibiotic therapy, our patient with atlantoaxial septic arthritis and septic thrombosis of the ophthalmic vein still experienced prolonged illness, a testament to the potential for unusual infectious manifestations of S. aureus with significant morbidity.

Acknowledgments

Consent. Written, informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review from the Editor-in-Chief of this journal.

Disclaimer. The findings and conclusions in this document are those of the authors, who are responsible for its content, and do not necessarily represent the views of the VA or of the US Government.

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