

A case of orbital apex syndrome due to *Pseudomonas aeruginosa* infection

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Abstract

Orbital apex syndrome is commonly been thought to have a poor prognosis. Many cases of this syndrome have been reported to be caused by paranasal sinus mycosis. We encountered a very rare case (60-year-old woman) of sinusitis with orbital apex syndrome due to *Pseudomonas aeruginosa* infection. She had received insulin and dialysis for diabetes and diabetic nephropathy, moreover anticoagulants after heart by-pass surgery. She underwent endoscopic sinus operation and was treated with antibiotics, but her loss of left vision did not improve. Recently, sinusitis cases due to *Pseudomonas aeruginosa* were reported to be increasing. Therefore, we should consider the possibility of *Pseudomonas aeruginosa* as well as mycosis as infections of the sinus, especially inpatients who are immunocompromised body.

Introduction

Orbital apex syndrome involves the optic nerve and structures in the apex of the orbita within the superior orbital fissures, including the nerves to the extraocular muscles and the first division of the trigeminal nerve.¹ This syndrome has been considered to have a poor prognosis.² Many studies have reported that this syndrome is caused by paranasal sinus mycosis.¹⁻⁶ Additionally, case reports of orbital apex syndrome due to *Pseudomonas aeruginosa* infection have been very rare. In Japan, such a case has not been reported. We present a case of orbital apex syndrome with left pansinusitis caused by *Pseudomonas aeruginosa* infection based on bacteria culture and postoperative pathologic examination, and treated by endoscopic sinus surgery.

Case Report

A 60-year-old Japanese woman presented at our hospital with left frontal pain of one

month duration and 12-day history of left visual disturbance as her chief complaints. Her first examination showed loss of the left eye light, paralysis of the left ocular muscles and anesthesia in the left first branch of the trigeminal nerve. She was alert, conscious and her vital signs were stable (temperature, 36.5°C; Blood pressure, 101/58 mmHg; Heart, 68/min). In her past history, she had received insulin and dialysis for diabetes and diabetic nephropathy, and anticoagulants after heart by-pass surgery. Blood examinations showed high levels of inflammation (WBC 6300/ μ L, CRP 2.8 mg) The other abnormal findings were glucose 494 mg/dL, HbA1c 9.7% (poor control), β -Dglucan 20.1 pg/mL (normal range :0-20 pg/mL) BUN 47 and creatinine 7.49, PT 15% (normal range: 70-100), and PT(INR) 4.18 (normal range: 0.90-1.10) (under dialysis). Computed tomography (CT) and magnetic resonance imaging (MRI) showed the findings of bilateral pansinusitis. The region of the left orbital apex showed low intensity by T1weight and high intensity by T2 weight (Figure 1). Bacteria culture of left nasal discharge showed *Pseudomonas aeruginosa* but not mycosis. From the above examination data, our patient was diagnosed as orbital apex syndrome due to sinusitis with *Pseudomonas aeruginosa* infection.

Preoperatively, the patient was treated with anticoagulants after heart by-pass operation, control together with insulin and dialysis three times/a week for diabetes and diabetic nephropathy, and received antibiotics (TAZ/PIPC) 2.25 g \times 3/a day for 4 days. The orbital apex syndrome was confirmed to be due to sinusitis with *Pseudomonas aeruginosa* infection by both bacteria culture and histopathologic examination in the operative wound. Postoperatively, the patient received TAZ/PIPC 2.25 g \times 3/a day for 4 days based on the results of a sensitivity test for *Pseudomonas aeruginosa*, and continued to receive anticoagulants, together with insulin and dialysis three times/a week. She underwent bilateral sphenoid-maxillaryectomy by endoscopic sinus operation under local anesthesia. Operative findings included pus pooling in the bilateral sphenoid-maxillary sinus, but not fungus or tumor (Figure 2). The pus of the left ethmoid sinus wound showed *Pseudomonas aeruginosa* (Mucoïd type) by bacteria culture. Histopathologic examination of the surgically removed left - ethmoid mucosa tissues showed mucinous substances surrounding bacteria (Figure 3), but not fungus, mycosis or tumors. The patient's postoperative general condition improved without surgical complications, but the visual loss remained. In our follow up by clinical findings, bacteria culture, MRI or MRA, she was free of *Pseudomonas aeruginosa* infection and brain complications for 1 year after treatment.

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Key words: sinusitis, *Pseudomonas aeruginosa*, orbital apex syndrome, visual disturbance, endoscopic sinus operation.

Received for publication: 19 September 2011.
Revision received: 3 November 2011.
Accepted for publication: 16 November 2011.

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Clinics and Practice 2011; 1:e127
doi:10.4081/cp.2011.e127

Discussion

Some cases of orbital apex syndrome were reported to be caused by paranasal sinus mycosis and showed poor prognoses. The mortality rate of this syndrome was 15-34%.^{1,6} However, there have been very few case reports of orbital apex syndrome due to *Pseudomonas aeruginosa* infection and none in Japan. Our search of PubMed and other systems revealed only 3 case as follows. Scully⁷ presented a patient in the US infected with *coagulase-negative staphylococci*, *propionibacter*, and *Pseudomonas aeruginosa*. This patient required treatment for primary hypothyroidism, insulin-dependent diabetes mellitus, hypertension, chronic atria fibrillation, and chronic obstructive pulmonary disease, and had a history of Hodgkin's disease and chronic lymphocytic leukemia. Colson and Daily⁸ reported a patient in the US with orbital apex syndrome and cavernous sinus thrombosis due to infection with *Staphylococcus aureus* and *Pseudomonas aeruginosa*. Chuna and Cullen⁹ in Singapore described a case with orbital apex syndrome due to *Aspergillus galactomannann* and *Pseudomonas aeruginosa* in uncontrolled diabetes. Their cases were mixed forms. However, our case showed orbital apex syndrome due to only *Pseudomonas aeruginosa*. Suzuki¹⁰ reported that sinusitis due to *Pseudomonas aeruginosa* recently showed a tendency to increase in Japan. *Pseudomonas aeruginosa* is classified into non-mucoïd and mucoïd types. Most cases of *Pseudomonas aeruginosa* infection belong to the mucoïd type, which can induce biofilms.¹¹ Bacterial biofilms can cause infections because they increase the resistance to antibiotics and phagocytosis.¹²

Therefore, attention should be paid to biofilms produced by *Pseudomonas aeruginosa*.^{10,13} Presently, the antibiotics recommended for *Pseudomonas aeruginosa* in Japan are tazobactam/piperacillin (TAZ/PIPC), meropenem (MEPM) or prulifloxacin (PUFX).¹⁰⁻¹¹ Our cases showed the mucoid type that is sensitive to TAZ/PIPC. The incidence of multidrug-resistant *Pseudomonas aeruginosa* (MDRPA) and metallo- β -lactamase-producing *Pseudomonas aeruginosa* has increased worldwide.¹⁴ In Japan, sinusitis with MDRPA had been not found until 2007 in a nationwide survey of clinical isolates from patients with otolaryngological field infection.¹⁵ However, in 2009, Saito¹⁴ reported sinusitis, orbital cellulites, and pneumonia caused by MDRPA in a patient with acute myeloid leukemia. In this report, A 74-year-old man with acute myeloid leukemia received induction chemotherapy. One day after chemotherapy, the patient developed febrile neutropenia. *Pseudomonas aeruginosa* as well as mycosis is sometimes observed as an opportunistic infection.¹¹ In the present case, the patient received insulin and dialysis for diabetes and diabetic nephropathy, and anticoagulants after heart by-pass operation. The three patients presented by the above (Scully,⁷ Colson and Daily,⁸ Cullen⁹) as well as our patient were

immunocompromised. Therefore in immunocompromised, sinusitis patients, we should keep the possibility of *Pseudomonas aeruginosa* as well as mycosis in mind and protect against severe complications such as orbital apex syndrome.

Previously, aggressive surgical debridement was a widely recognized method for managing orbital apex syndrome, though, serious problems requiring extensive plastic surgery could result with residual defects necessitating psychotherapy for the emotional difficulties associated with the treatment.⁴ It is especially important to note that, in the most serious complications of orbital apex syndrome, cavernous thrombophlebitis can occur.¹⁶ Even radical operations including ophthalmectomy cannot improve cavernous thrombophlebitis. Recently, the development of both antimicrobial treatments and endoscopic sinus operation has contributed to the successful management of orbital apex syndrome by the combined therapy of antimicrobial drug and drainage by endoscopic sinus operation.¹⁷⁻¹⁸ Zuma¹⁹ described that ozone aeration could disinfect *Pseudomonas aeruginosa*. Similarly, drainage by endoscopic sinus operation combined with antibiotics was effective for our case. Because orbital apex syndrome due to

mycosis has the possibility of brain aneurysm and infarction by thrombophlebitis after treatment, we should follow up by MRI and MRA.

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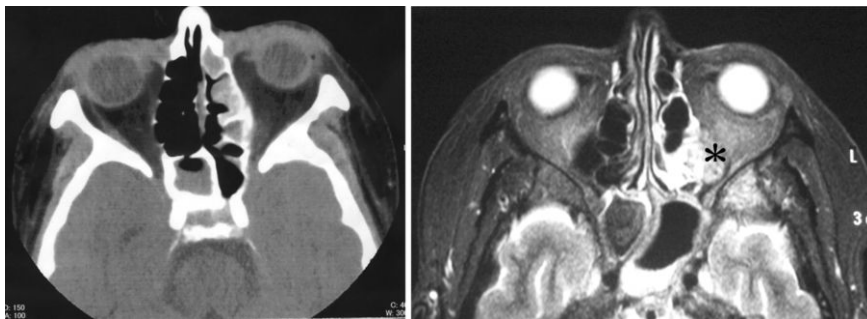


Figure 1. Preoperative magnetic resonance imaging and computed tomography . The disturbed area of the left orbital apex (asterisk) showed high intensity by T2 weight (right), but could not be identified by computed tomography (left).

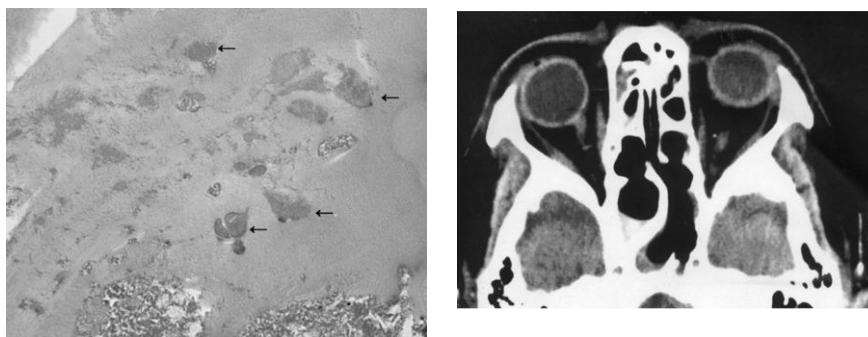


Figure 2. A plain computed tomography (CT) at 4 weeks after operation revealed the disappearing soft tissue density of the left ethmoid sinus wound in comparison with the preoperative CT (see Figure 1).

Figure 3. Histopathologic examination (H&E stain : X400). In the left ethmoid mucosa tissues, mucinous substances surrounding the bacteria (arrows) could be observed with the inflammatory changes but there was no mycosis or tumor.

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