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Invasive fungal sinusitis with abducens nerve palsy

SHORT CASE REPORT

EIRIK MATHISEN

eirik.mathisen@so-hf.no
Department of Otorhinolaryngology
Østfold Hospital Trust
Eirik Mathisen, specialist in otorhinolaryngology, senior consultant and medical adviser
The author has completed the ICMJE form and declares no conflicts of interest.

SAAD ABALLI

Section for Clinical Immunology and Infectious Diseases
Oslo University Hospital, Rikshospitalet
Saad Aballi, specialist in internal medicine and infectious diseases, senior consultant
The author has completed the ICMJE form and declares no conflicts of interest.

TORSTEIN GRØNSETH

Department of Otorhinolaryngology
Oslo University Hospital
Torstein Grønseth PhD, specialist in otorhinolaryngology, senior consultant
The author has completed the ICMJE form and declares no conflicts of interest.

KRISTINE MARIE AARBERG LUND

Department of Infectious Diseases
Østfold Hospital Trust
Kristine Marie Aarberg Lund, specialist in internal medicine and infectious diseases, senior consultant
The author has completed the ICMJE form and declares no conflicts of interest.

ASTRID DEAN ALVIM

Department of Radiology

Østfold Hospital Trust
Astrid Dean Alvim, specialist in radiology and senior consultant
The author has completed the ICMJE form and declares no conflicts of interest.

Background

Though rare, sphenoid sinusitis can cause abducens nerve palsy because of the anatomical proximity of the sphenoid sinus and the abducens nerve.

Case presentation

A male patient in his late seventies presented with double vision and left abducens nerve palsy. Imaging revealed sinus opacifications later identified as due to *Scedosporium apiospermum*, a rare fungal pathogen. Despite being immunocompetent, the patient developed invasive fungal sinusitis involving the sphenoid sinus. Treatment included systemic antifungal therapy with voriconazole and sinus surgery. Follow-up imaging showed sinus recovery and bony healing. Clinical improvement was noted, with resolution of abducens nerve palsy at the four-month follow-up.

Interpretation

This case highlights the importance of assessing relevant imaging findings outside the central nervous system on routine brain imaging, as well as the role of effective antifungal therapy and surgical management in rare cases of invasive fungal infection in immunocompetent patients. Prompt intervention can significantly improve outcomes.

A patient was admitted to hospital with double vision and ocular muscle paralysis. The underlying cause was found to be a fungal infection of the sphenoid sinus. This condition is rare, particularly in immunocompetent and healthy patients.

A man in his seventies was admitted to the neurology department with headache and double vision. He had previously been healthy and was not taking any regular medications. His symptoms started with a low-grade, pressing headache in the right side of the parieto-occipital region. The pain subsided during the night. The next day, the patient developed a more intense headache along with double vision and was admitted to the neurology department. He was afebrile, CRP 2 mg/L (reference value < 6), leukocytes 9.2×10^9/L (3.7–9.8), blood pressure 156/72 and pulse 61. His general condition was good.

Upon admission, a left-sided abducens paresis was observed (Figures 1a and 1b). A CT scan of the head was immediately performed to determine whether this was due to a cerebral event. Head CT showed no intracranial pathology. Opacities were observed in several of the sinuses, including the left sphenoidal sinus, where there were sclerotic walls and calcium deposits.

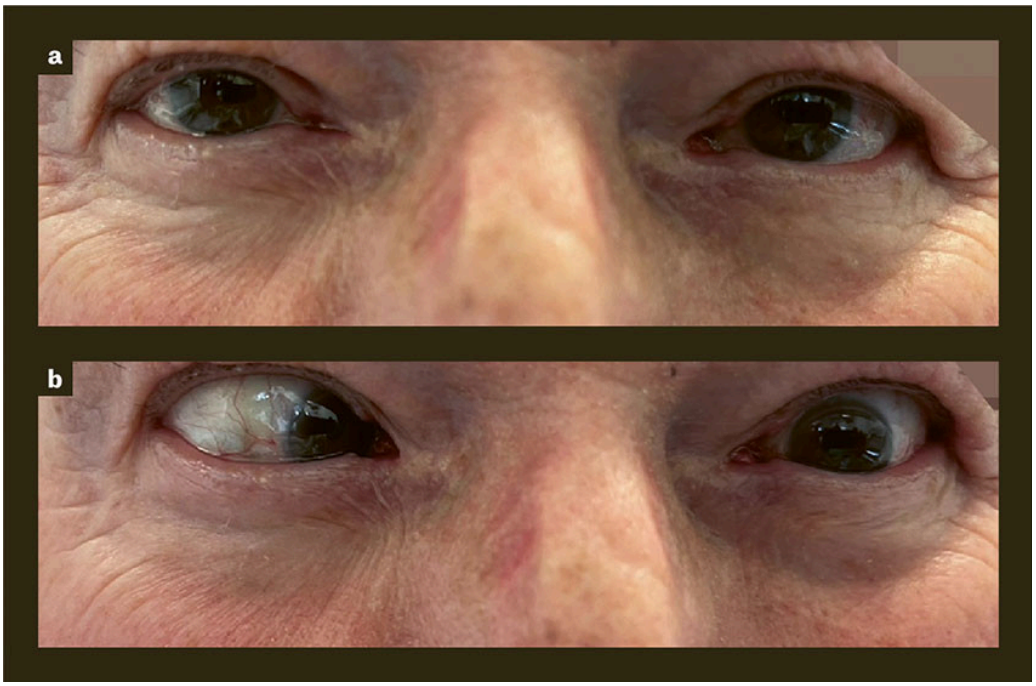

Figure 1 The images show the patient with left-sided abducens paresis and esotropic strabismus. When the patient looks straight ahead, the left eye is turned inward/medially (a). The left eye lacks temporal movement and does not track when the patient looks to the left (b).

Based on the patient's medical history and clinical findings, a stroke could not be ruled out, and treatment was therefore initiated with oral acetylsalicylic acid 75 mg once daily. Eight hours after admission, MRI of the head was performed, which showed no intracranial pathology but did reveal sinus opacification with a low T2 signal (Figure 2). In combination with the clinical presentation and CT findings, this suggested chronic fungal sinusitis. Involvement of the left sphenoid sinus indicated possible involvement of the adjacent left abducens nerve.

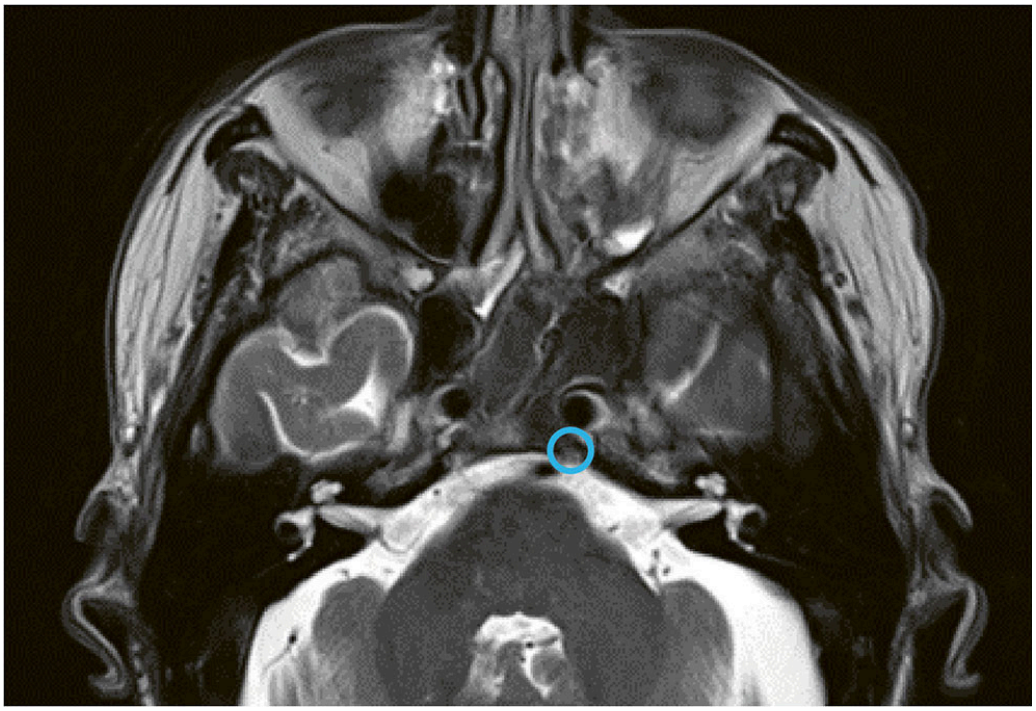

Figure 2 Axial T2-weighted MRI of the head showed no cerebral or intracranial pathology but demonstrated low signal intensity in the left sphenoidal sinus. In combination with the clinical picture and CT findings, this raised suspicion of fungal sinusitis. The circle around the entrance

to Dorello's canal shows the course of the left abducens nerve in close proximity to the sphenoid sinus.

An ENT consultation was requested, during which the examination revealed significant medialisation of the medial wall of the maxillary sinus and purulent discharge in the middle meatus. Two days after admission, an antrostomy was performed under local anaesthesia to allow visual access to the maxillary sinus. Dark green, clay-like material was observed, which is a typical finding in fungal sinusitis. The material was sent for culture, but no growth was detected on blood or chocolate agar. Based on radiological and clinical findings consistent with fungal infection, systemic antifungal treatment was initiated with intravenous micafungin 100 mg once daily, along with oral prednisolone 30 mg once daily.

On day three after admission, the patient was referred for a CT scan of the sinuses, which revealed bone destruction of the clivus at Dorello's canal (Figure 3). This reinforced the suspicion of invasive fungal sinusitis, and the radiological findings potentially explained the left abducens nerve palsy (1). Six days after admission, the patient was transferred to the ENT department, and acetylsalicylic acid was discontinued.

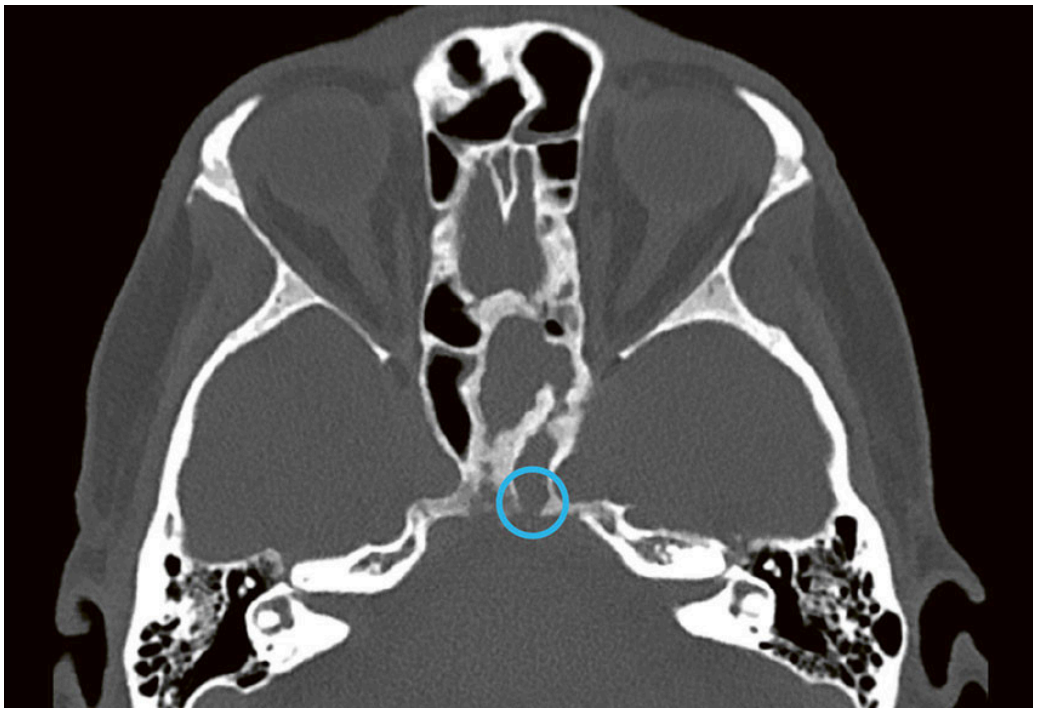

Figure 3 CT of the sinuses showed an opacified, sclerotic left sphenoid sinus consistent with chronic inflammation, accompanied by destruction of the clivus (marked)

A repeat sinus procedure was performed under general anaesthesia seven days after admission. The ethmoid cells were opened, the maxillary sinus was re-cleaned and a wide opening into the sphenoid sinus was formed. Fungal changes similar to those observed during the initial procedure were found in both the maxillary and sphenoid sinuses. New tissue and culture samples were obtained, and visibly infected tissue was removed. Fungal culture identified *Scedosporium apiospermum*, with a low minimum inhibitory concentration for voriconazole of 0.032. Following consultation with an infectious disease specialist, micafungin was discontinued and treatment with intravenous voriconazole 200 mg twice daily was initiated.

After just over one week of treatment with intravenous voriconazole, the patient was discharged in good general condition with a further six-week course of oral voriconazole 300 mg twice daily. The patient still had abducens nerve palsy and used a patch over his left eye to avoid diplopia. Seven weeks after symptom onset, the abducens paresis persisted, and a follow-up CT of the sinuses was ordered. This showed an almost normal, aerated sphenoidal sinus and signs of bony healing. At a telephone follow-up one month later, the patient reported significant improvement, experiencing diplopia only late in the day when fatigued. At the final check-up, nearly four months after symptom onset, the patient was symptom-free, with no objective signs of abducens paresis. Rhinoscopy showed completely normal findings in the nasal cavity and paranasal sinuses.

Due to the invasive fungal sinusitis caused by a rare fungal species, the patient was evaluated by infectious disease specialists during the recovery phase for potential underlying immunodeficiency or malignancy. Apart from his age, no factors were identified that would increase the risk of opportunistic infections.

Discussion

The abducens nerve, cranial nerve VI, is a purely motor nerve responsible for innervating the eye's lateral rectus muscle. From the brainstem, the nerve crosses the prepontine cistern and proceeds intradurally in close proximity to the dorsal clivus within Dorello's canal (Figure 4). It then passes through the cavernous sinus before entering the orbit via the superior orbital fissure.

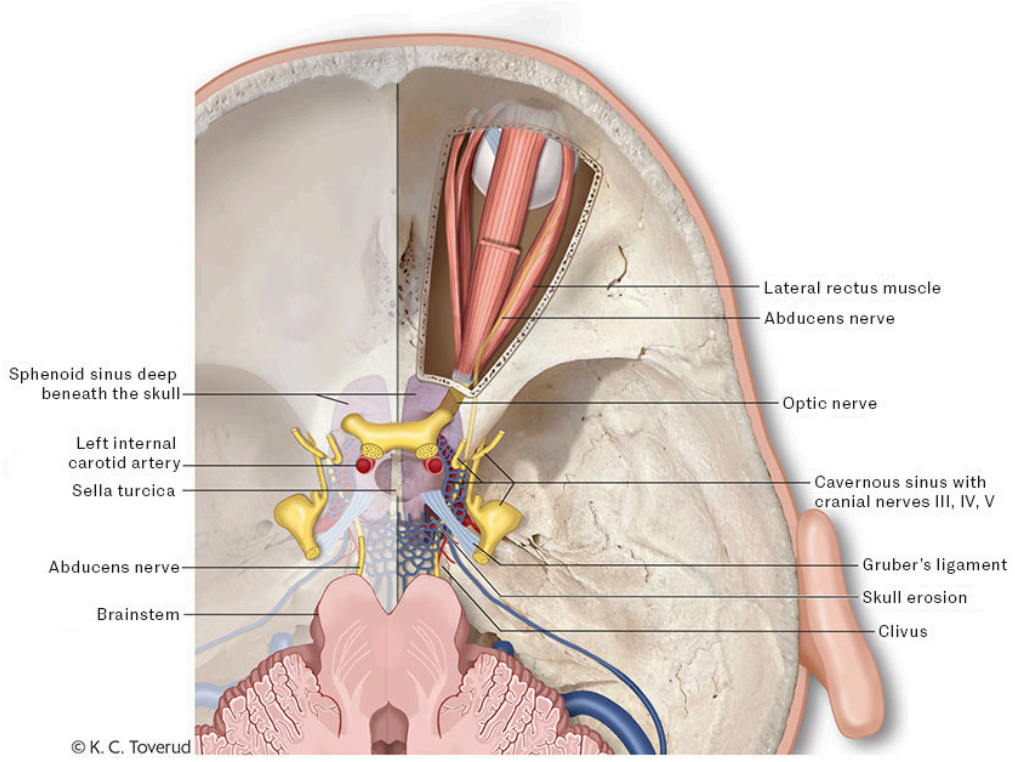

Figure 4 Anatomical relationships of the abducens nerve. On the left side of the illustration, the skull base is shown covered by dura. Dorello's canal begins where the abducens nerve penetrates the dura and ends at the anterior edge of Gruber's ligament. On the right side, the

dura has been removed to reveal how the abducens nerve in the canal lies against the erosion in the clivus. Illustration: Kari C. Toverud

The abducens nerve is the most commonly affected nerve in isolated extraocular muscle palsy (2). When an underlying cause is identified, the palsy is usually due to microvascular ischaemia or trauma. Skull base tumours, complicated otomastoiditis and COVID-19 are among the less common causes. Although sphenoidal sinusitis is a rare cause of abducens nerve palsy, its close anatomical proximity makes this a possible aetiology (1). The nerve can be affected either within the cavernous sinus or in Dorello's canal, as in this patient.

Species of *Scedosporium* can cause opportunistic fungal infections that are often difficult to treat due to the fungus' inherent resistance to many antifungal agents. Voriconazole is the preferred treatment, where it is tolerated (3). Micafungin, which was initially administered to this patient, is an appropriate empirical choice for yeast infections but is ineffective against mould infections. No fungal growth was observed from the material collected during the first procedure. The specimen was not cultured on Sabouraud dextrose agar – a medium specifically designed for the growth and detection of mould and yeast fungi. Nevertheless, growth of *Scedosporium* could be expected with prolonged incubation on blood and chocolate agar, which was performed. The lack of fungal growth could potentially be due to suboptimal specimen quality.

There are several species within the *Scedosporium* genus, at least five of which are known to cause disease in humans. These are filamentous fungi found in soil and contaminated water. Surgery is recommended as part of the treatment, alongside systemic antifungal therapy. The fungus can cause both localised and invasive disease, typically affecting the skin or soft tissues. Immunocompetent individuals can also be infected, but invasive disease is primarily seen in patients with underlying conditions (4). Further investigation, particularly in relation to haematological malignancy and immunodeficiency, must be considered on a case-by-case basis. To date, only a few case reports have described invasive infection caused by *Scedosporium apiospermum* in immunocompetent patients (5, 6).

Voriconazole has variable metabolism, and dosage therefore requires monitoring of serum concentrations and individual adjustment. For invasive fungal infections, the recommended serum concentration is 2.0–5.0 mg/L. The drug is associated with a range of potential side effects, and patients should undergo weekly monitoring of electrolytes, liver function and renal function during the first month of treatment as a minimum (7).

The patient has consented to publication of the article.

The article has been peer-reviewed.

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