

Isolated invasive fungal sphenoid sinusitis – a case study

Izolowane inwazyjne zapalenie zatoki klinowej – opis przypadku

Authors' Contribution:

A – Study Design
B – Data Collection
C – Statistical Analysis
D – Manuscript Preparation
E – Literature Search
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ABSTRACT:

Invasive fungal sphenoid sinusitis is a rare and potentially life threatening disease. The incidence rate of fungal sphenoiditis among patients with isolated sphenoid disease varies from 4.5% to 26.8%. Generally, the symptoms of sphenoid sinusitis are non-specific and include headaches, visual disorder, post-nasal drip, nasal obstruction and paralysis or paresis of single or multiple cranial nerves. This study describes a case of isolated invasive fungal left-sided sphenoiditis in an immunocompetent 56-year-old woman. The first sinus computed tomography (CT) examination revealed complete opacification of the left sphenoid sinus, the second CT scan showed also erosions of the bony walls of sphenoid sinus. Endoscopic sinus surgery was performed and intraoperatively fixed and dilated left pupil was observed; thus, the operating team decided to finish the operation immediately. Left-sided oculomotor nerve paresis was diagnosed by the ophthalmologist. Pathologic analysis demonstrated fungal hyphae morphologically identical to *Aspergillus spp.* Due to a suspicion of cerebrospinal fluid (CSF) leak, increasing pneumocephalus and manifestation of mental changes in our subject, endoscopic reoperation with CFS fistula closure was done nine days after the first operation. The patient was treated with intravenous voriconazole and ceftriaxone for one month, and after discharge with itraconazole orally for 3 months. Full recovery of the left oculomotor nerve function ensued 4 months after surgery and no signs of recurrence of the fungal disease were present up to date.

KEYWORDS:

case study, isolated sinusitis, sphenoid fungal sinusitis

STRESZCZENIE:

Inwazyjne grzybicze zapalenie zatoki klinowej jest rzadką, ale potencjalnie zagrażającą życiu chorobą. Częstość występowania grzybiczego zapalenia zatok wśród pacjentów z izolowanym zapaleniem zatoki klinowej wynosi od 4.5% do 26.8%. Najczęściej objawy zapalenia zatok klinowych są niespecyficzne i obejmują: bóle głowy, zaburzenia widzenia, sptywanie po tylnej ścianie gardła, niedrożność nosa oraz pojedyncze lub mnogie porażenie bądź niedowład nerwów czaszkowych. W niniejszej pracy opisany został przypadek izolowanego, inwazyjnego zapalenia lewej zatoki klinowej u 56-letniej pacjentki bez zaburzeń odporności. Pierwsze badanie tomografii komputerowej zatok (TK) ujawniło całkowite zaciemnienie lewej zatoki klinowej, a drugie badanie TK wykazało również erozję ścian zatoki klinowej. Przeprowadzono endoskopowy zabieg zatok i śródoperacyjnie zaobserwowano nieruchomą i poszerzoną lewą źrenicę. Z tego powodu zespół chirurgiczny zdecydował o natychmiastowym przerwaniu zabiegu. Okulista rozpoznał niedowład lewego nerwu okoruchowego. Badanie histopatologiczne wykazało obecność strzępków grzybni, podobnych do *Aspergillus spp.* Ze względu na podejrzenie wycieku płynu mózgowo-rdzeniowego (PMR) oraz nasilenie odmy śródczaszkowej i pojawienie się zmian psychicznych u naszej pacjentki, endoskopowa reoperacja z zamknięciem przetoki PMR została wykonana dziewięć dni po pierwszej operacji. Kobieta była leczona dożylnie worykonazolem i ceftriaksonem przez miesiąc, a po wypisie do domu doustnie itraconazolem przez 3 miesiące. Pełny powrót funkcji lewego nerwu okoruchowego nastąpił po 4 miesiącach od operacji i do tej pory pacjentka nie ma objawów nawrotu choroby grzybiczej.

SŁOWA KLUCZOWE: analiza przypadku, grzybicze zapalenie zatok, izolowane zapalenie zatok

ABBREVIATIONS

CSF – cerebrospinal fluid
CT – computed tomography
ESS – endoscopic sinus surgery
MRI – magnetic resonance imaging

INTRODUCTION

Isolated sphenoid sinusitis is a rare condition and the incidence rate of fungal sphenoiditis among patients with isolated sphenoid disease varies from 4.5% to 26.8% [1, 2, 3, 4]. In general, fungal sinusitis is classified into non-invasive and invasive. The first

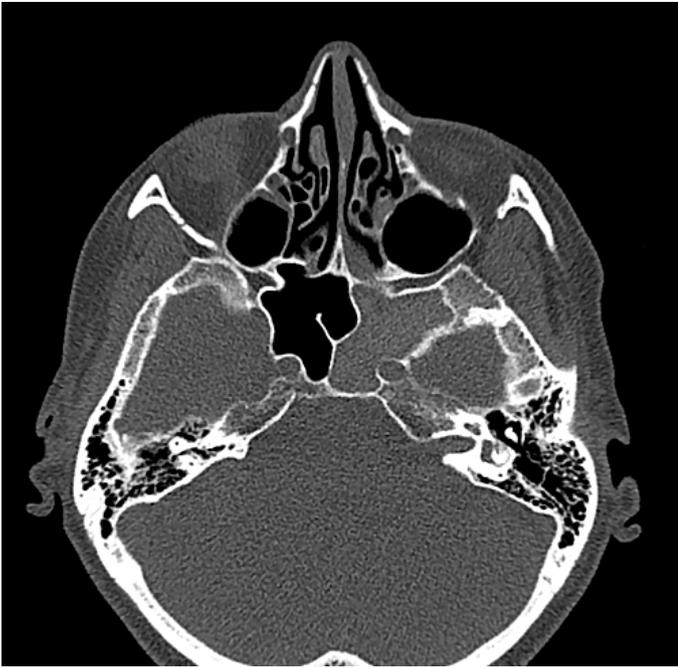


Fig. 1. Unenhanced computed tomography scan, axial view, performed one year before surgery. Complete opacification of the left sphenoid sinus without bone erosions of sphenoid sinus walls is apparent.

group contains saprophytic fungal infestation, fungal ball and allergic fungal rhinosinusitis. Acute invasive fungal rhinosinusitis, chronic invasive fungal rhinosinusitis, chronic granulomatous invasive fungal rhinosinusitis were classified to the second group [5]. Non-invasive forms of fungal sinusitis have a good prognosis, in contrast to invasive disease which has high mortality rates ranging from 50–80% [6, 7, 8].

Invasive fungal sphenoiditis is more dangerous than invasive fungal infection of the other paranasal sinuses because of the fact that sphenoid sinus is surrounded by other vital structures such as internal carotid artery, orbital apex, cavernous sinus, optic nerve, pituitary gland and cranial nerves (III, IV, V1, V2, VI). The most common symptoms of sphenoid sinusitis are non-specific and include headaches, visual disorder, postnasal drip, nasal congestion and paralysis/paresis of cranial nerve [2].

In this study we present the case of a 56-year-old immunocompetent woman who developed oculomotor nerve palsy [third cranial nerve (CN III)] and cerebrospinal fluid (CSF) leak as a consequence of endoscopic sinus surgery (ESS) due to invasive isolated fungal infection of the left sphenoid sinus.

CASE REPORT

A 56-year-old woman with a past medical history of allergic rhinitis presented with persistent strong headache lasting from 18 months. She complained also of nasal obstruction and post-nasal drip syndrome. The patient was initially treated at a local otolaryngology outpatient clinic where empirical oral antibiotic therapy and nasal steroids were used and sinus CT was performed. Only isolated opacification of the left sphenoid sinus with proper bone



Fig. 2. Unenhanced computed tomography scan, axial view, performed few days before surgery shows total opacification of the left sphenoid sinus with defect of the sinus wall.

boundaries were found in that study (Fig. 1.). Conservative therapy was unsuccessful so about one year after the first CT scans, a new no-contrast-enhancement CT examination was recommended, in which opacification of the left and right sphenoid sinus (high density of about 100 Hounsfield units), intralesional hyperdensity or calcification and suspicion of intracranial invasion through massive erosion of bone borders were described (Fig. 2.). In addition to this, MRI confirmed complete involvement of the left sphenoid sinus. The fluid filling the left sphenoid sinus was described. There was a suspicion of post-inflammatory changes and no contrast enhancement areas in the brain were seen. After admission to the Department of Otolaryngology, Head and Neck Surgery, the patient's vital signs were correct (a temperature of 36.6 degree Celsius, a pulse of 99/minute and blood pressure of 150/90 mm Hg). Laboratory parameters were also within normal limits. Physical examination revealed a deviated septum with hypertrophy of the inferior turbinate and partial nasal obstruction.

ESS was performed under general anaesthesia using endoscopic magnified view in 4K technology. Since a non-specific lesion of sphenoid sinus was suspected, an intraoperative histopathological examination was planned. The surgeon performed endoscopic left-sided septoplasty followed by middle mucotomy. In the right sphenoid recess, the natural sphenoid ostium was opened widely. A lot of consistent necrotic tissue was evacuated from the right sphenoid sinus using suction and irrigation. The mucosa around the sphenoid ostium were polypoid altered, thus the polyps around the sphenoid ostium were removed. On the left side sphenoidectomy was performed. There was much more greyish mass on the left than the right side and consequently it was evacuated with Blakesley nasal forceps. Low-pressure irrigation of the sinus cavity with saline was also done. The bony walls of the sinus were thin. The collected material was sent for histopathological and

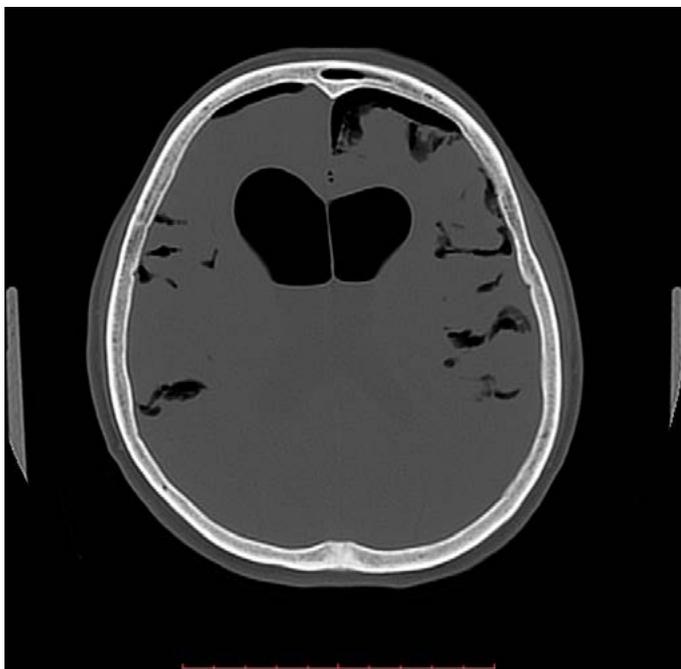


Fig. 3. Computed tomography scan, axial view, performed 7 days after surgery. Pneumocephalus with compression of frontal lobes and presence of air in anterior horns of lateral ventricles are seen.

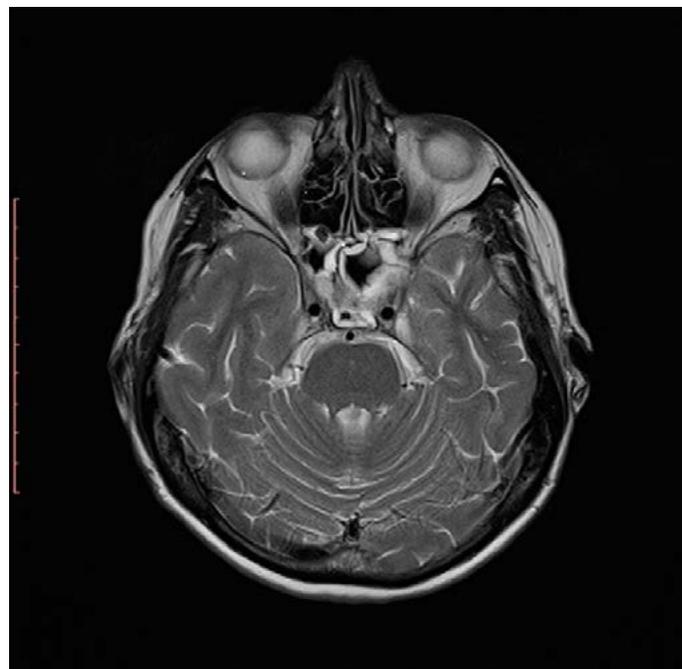


Fig. 4. MRI scan with contrast enhancement, axial T2W image, performed 2 months after discharge. MRI shows mucosal thickening in both sphenoid sinuses, greater in the left than the right sinus and the presence of a fatty patch in the posterior part of sphenoid sinus.

mycological examination. Histopathological intraoperative examination revealed hyphae morphologically identical to *Aspergillus* intermingled with mucus-filled mass, inflammatory infiltration and bacterial colonies. Examination was negative for neoplastic cells. Moreover, postoperative histopathological outcomes were similar. Extensive bleeding from the left sphenopalatine recess were stopped with bipolar coagulation. After haemostasis spongostan was positioned in both sphenoid recesses. During operation the anaesthetist revealed short-term asystole. Next, 1 mg of atropine was given which resulted in return to correct normal sinus rhythm. Since our patient presented fixed and dilated left pupil, the surgeon decided to finish the operation immediately without taking much more tissues to histopathological examination and without further cleaning of the sinus cavity from necrotic masses. In addition to this, the doctor requested direct urgent ophthalmologist and neurosurgeon consultations and CT scans of the brain. Consensual light response could not be examined due to the influence of anaesthetic drugs on pupils. The ophthalmologist diagnosed left oculomotor nerve paresis with ptosis and limited adduction, supraduction and infraduction. Moreover, CT scan revealed irregular, hyperdense structures situated upper to the dorsum sellae on the left side which spread medially to the carotid artery, posteriorly to the anterior communicating artery, anteriorly to the basilar artery and enfolded the left oculomotor nerve (which were similar to haemorrhagic lesions). Sphenoid sinus wall destructions, greater on the left than the right side were seen, involving posterior and lateral walls. Optic nerves were intact.

There were no other neurological deficits except oculomotor nerve paresis, so the neurological recommendations included placing the patient in Fowler position of 30°, intravenous osmotherapy using mannitol and furosemide, monitoring of vital signs and another CT scan in the next day.

The patient was treated with intravenous voriconazole 200 mg every 12 hours and ceftriaxone 1.0 g every 12 hours. Then laboratory tests for the level of electrolytes and liver enzymes along with kidney function were regularly monitored.

The next day, even though analgesic medications were given, the patient complained of headache, systolic blood pressure was increased occasionally above 200 mmHg, close supervision was continued. There were no specific signs of CSF leak beyond the typical postoperative post-nasal drip. Endoscopic examination of sphenoid recesses did not confirm leakage with certainty. Control CT scan of the head demonstrated pneumocephalus, with compression of frontal lobes and presence of air in anterior horns of lateral ventricles (Fig. 3.). Also, small heterogeneous opacification lateral to the sella turcica on the left side was described. The sphenoid sinus was filled with air and fluid. The neurosurgeon recommended maintaining lying position. In addition to this, personality changes, psychomotor retardation and confusion in our patient were observed.

In the second day after surgery the endoscopic examination was done on operating theatre by ENT-neurosurgeon team. CSF leak has not been confirmed thus conservative management was continued. The next day after reoperation herpes labialis of the upper lip occurred, so acyclovir was administered intravenously 250 mg every 8 hours. Another postoperative control CT scan of the head showed a decrease in blood volume in the previously described reservoirs around skull base. However, mycological workup did not reveal fungal culture.

In the following days, the patient's condition did not improve. Nine days post-surgery enlargement reoperation was planned due to endoscopic suspicion of CSF leak, pneumocephalus confirmed in CT

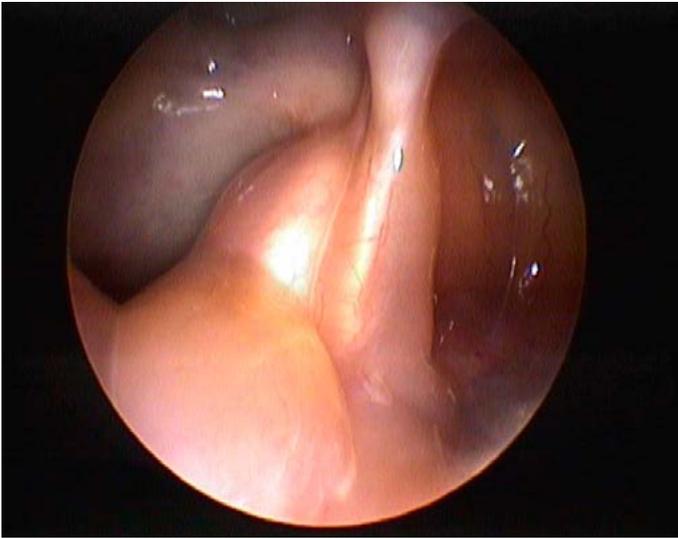


Fig. 5. Endoscopic view of the left sphenoid sinus, 3 months after surgery. There are no signs of fungal disease recurrence. The presence of a fatty patch in the posterior part of sphenoid sinus is noted.

scans and manifestation of mental changes in our subject. Under general anaesthesia, endoscopic exploration of sphenoid sinus and sellar region showed a small fistula and CSF leak localized in the posterior wall of left sphenoid sinus. The surgical team decided to not remove the sphenoidal mucosa in the region of fistula (clivus) completely due to the very thin bone and high risk of bleeding, which could be fatal. Subsequently, the fistula region was coagulated and closed by using abdominal fat, tissue glue and nasal packing sponge. We decided not to use the most common material for reconstruction of skull base known as Hadad-Bassagasteguy flap because of septal mucosal damage in that region which had made during septoplasty in the first surgery. The surgical procedure was performed without complications and the patient did not develop any new neurological symptoms.

24 hours after surgery the patient was transferred to Department of Neurosurgery in a good general condition and with left-sided oculomotor nerve paresis.

During further observation in the Department of Neurosurgery, treatment with voriconazole, ceftriaxone and analgesic medications was continued, the patient's condition improved, also CSF leak was not observed. Control CT scans performed 3 and 10 days after admission to the Department of Neurosurgery demonstrated a reduction of intracranial air. The patient was discharged with a recommendation of continuing orally antifungal therapy with itraconazole 100 mg every day for 3 months with monitoring of kidney and liver function, eye muscle exercises and laryngological, ophthalmic and neurosurgery control visits. A postoperative visit was carried out every 1 month. A follow-up MRI scan of the head with contrast enhancement performed 2 months after discharge revealed mucosal thickening in both sphenoid sinuses, greater in the left than the right sinus and the presence of a fatty patch in the posterior part of sphenoid sinus (Fig. 4.). Endoscopic examination of the operated region did not show any signs of fungal disease recurrence (Fig. 5.). About 12 weeks after discharge there was full recovery of oculomotor nerve function and no signs of re-

currence of the disease were present up to date. The patient does not report any worrying symptoms from the nose and paranasal sinuses. Moreover, persistent headache has completely relieved.

DISCUSSION

Invasive fungal sphenoiditis is a rare condition occurring most commonly in immunocompromised patients, especially elderly subjects with poorly controlled diabetes mellitus, malignancies or using prolonged systemic corticosteroid therapy [9]. Due to nonspecific symptoms diagnosis of fungal sphenoiditis is probably underestimated. CT scans play an important role in preliminary diagnosis of fungal sinusitis, also MRI should be considered in case of doubt [4]. Both CT scans and MRI enable to plan a surgery in order to complete fungal tissue debridement which is essential for achieving therapeutic success. Our patients' second CT scan presented a pathological mass in the left sphenoid sinus in addition to erosions of the bony walls which may mimic a malignancy of the sphenoid sinus. Although several reports showed possible radiological findings in fungal sinusitis [2,9], they are non-specific, so differentiation between chronic invasive fungal sinusitis and malignancy, especially when bone erosions are present is tough and may not be based on imaging findings [10]. Bone erosions which developed as a result of pressure necrosis due to inflammatory process were reported in 4–17% of fungus ball, non-invasive form of fungal sinusitis [11].

Histological examination is always required to establish a differentiation between invasive or non-invasive form of fungal sinusitis. In our patient intraoperative histopathological examination was done and was consistent with the final histopathological outcome. In addition to this, mycological workup was negative for fungus, but it is known from literature that positive cultures are reported only in 10.5% to 72.7% of cases [12].

Oculomotor nerve palsy in isolated sphenoid sinusitis is an uncommon but significant complication. Mograbi and Soundry in their study reviewed 17 cases with ocular cranial nerve (III, IV and VI) palsies secondary to sphenoid sinusitis and among them 18% ($n = 3$) had isolated oculomotor nerve palsy [13]. Overall, major complications after ESS defined as orbital haemorrhage, blindness, diplopia, haemorrhage, and skull base defects (cerebrospinal fluid leak, meningitis, pneumocephalus, intracranial abscess, intraparenchymal damage) occurred in 1–2% all ESS [14]. Data regarding cranial neuropathy after ESS performed due to fungal sinusitis are scarce [13]. We speculated that the mechanism of oculomotor nerve paresis in our patient was focal injury of the oculomotor nerve by increased/decreased pressure generated in the cavernous sinus region by irrigation and removal of inflammatory tissue, even so it was performed using endoscopic magnifying view in 4K technology by an experienced surgical team. On the one hand, such a procedure is required for complete removal of necrotic fungal tissues, on the other hand destructions of the sphenoid bone are a potential risk factor for complications during surgery [9].

The patient was referred to our clinic about 18 months after the onset of symptoms. In our patient, the headache was

strong, persistent and did not response to nonsteroidal anti-inflammatory drugs. It is possible that an earlier performance of the procedure could have allowed to avoid such complications, because the first CT scan did not reveal features of lysis of the sphenoid sinus walls.

However, the question is how early surgery should be considered for patients with isolated opacification of the sphenoid sinus and how often should one perform CT scans to exclude bone erosion confirming the severity of the process and being an indication for surgery without unnecessary delay?

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