

Pott's puffy tumour and severe intracranial complications in a patient with schizophrenia: a case report*

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Rhinology Online, Vol 3: 123 - 127, 2020

<http://doi.org/10.4193/RHINOL/20.014>

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***Received for publication:**

March 5, 2020

Accepted: August 12, 2020

Published: August 22, 2020

Abstract

Background: Pott's puffy tumour is a subperiosteal abscess with cranial osteomyelitis. In compliant patients, antibiotic and surgical treatment is effective. However, patients with schizophrenia can present disease atypically, affecting the outcome and leading to life-threatening complications.

Case presentation: We present a case where a 47-year-old man with schizophrenia declined surgery and follow-up of chronic frontal rhinosinusitis with a Pott's puffy tumour. Nine months later he was found unconscious with osteomyelitis in his forehead, cerebritis and left-sided subdural empyema originating from the Pott's puffy tumour. A multidisciplinary team of otolaryngologists and neurosurgeons performed acute endoscopic sinus surgery and craniotomy. Despite an intensive antibiotic treatment regimen and revision craniotomies, the patient suffered severe sequelae.

Conclusion: Altered disease presentation and potential compliance issues must be kept in mind when treating psychiatric patients with severe and complicated sinonasal infection. This case illustrates the challenging interplay between rhinology and psychiatry.

Key words: Pott puffy tumour, frontal sinusitis, subdural empyema, osteomyelitis, schizophrenia

Introduction

Pott's puffy tumour (PPT) comprises a subperiosteal abscess located superficially to the anterior wall of the frontal sinus, where osteomyelitis has eroded the bone. The condition mainly affects males and is caused by head trauma or acute or chronic frontal sinusitis⁽¹⁾.

Due to improved diagnosis and treatment, complication rates for suppurative frontal sinusitis with PPT have decreased. If untreated, severe, life-threatening complications can arise and timely treatment is essential as the prognosis is correlated to the time of diagnosis⁽²⁾. One complication is concurrent spread of infection to neighbouring structures like the brain and eyes⁽²⁾. Patients with mental disorders are not always able to perceive or report symptoms of somatic disease. This can result in a misleading disease presentation without key symptoms and lead to

substantial morbidity—particularly in patients with schizophrenia⁽³⁾. Accordingly, schizophrenia increases risks and complicates treatment of somatic disease due to reduced pain perception and reporting, as well as a reduced or absent impulse to seek medical attention and perform self-care^(4–7).

Here, we present a challenging case of a patient with schizophrenia suffering from PPT with subsequent severe irreversible complications due to delayed intervention.

Case presentation

A 47-year-old Caucasian male, diagnosed with schizophrenia in his teens, was referred to our otorhinolaryngology (ORL) department on suspicion of cancer due to a slow growing tumour on his forehead (Figure 1). He reported a 22-year history of nasal obstruction and 1–2 years of daily epistaxis. Prior to admission,

he had never consulted a physician with his sinonasal symptoms and his diagnosis of chronic rhinosinusitis with nasal polyps was therefore unknown. He reported no physical complaints such as pain, fatigue or visual symptoms. Other than schizophrenia, the patient's medical history was non-contributory. He had no allergies and his only medication was the antipsychotic drug pimozide. Rhinoscopy revealed polypoidal masses on the left side in relation to the middle turbinate. A soft tumour above the left eye displaced the eyeball 2 mm inferiorly. Vision and ocular movement was intact. A few days before his visit, a sinocutaneous fistula had eroded medially to his left eye (Figure 2). Biopsies were taken at first visit. A computed tomography (CT) scan of the sinuses showed opacification in the ethmoidal and frontal sinuses extending to the left orbit. Furthermore, defects in the frontal and posterior walls of the frontal sinuses were visualised (Figure 3). Magnetic resonance imaging revealed frontal sinus ostial obstruction and bilateral mucocoele but no intracranial involvement. Biopsies from the nasal cavity showed acute and chronic inflammation with no malignancy. Multidisciplinary surgery by neuro- and ORL-surgeons was scheduled to restore frontal sinus drainage and cranialise the posterior frontal sinus wall. Nevertheless, after two weeks of consideration, the patient declined surgery. According to Danish law compulsory treatment was not an option. A letter was sent to the patient inviting him for a follow-up visit, but he did not reply.

Nine months later he was found unconscious in his home. He had not been in contact with his relatives for four days. The clinical examination revealed a Glasgow Coma Scale 3 (no eye opening, no motor- or verbal response), neck stiffness, anisocoria and pus flowing from a sinocutaneous fistula in his forehead. Further, he had widespread pressure ulcers. After intubation on-site, ceftriaxone, penicillin and methylprednisolone were administered intravenously on suspicion of meningitis, and he was referred to our hospital. During transport he developed generalised tonic-clonic seizures, which were treated with diazepam. Upon admission, a CT scan showed pansinusitis, left frontal lobe cerebritis and a left-sided subdural empyema causing midline shift (Figure 4). Intravenous metronidazole was added to the antibiotic treatment. The patient was operated on vital indication by neurosurgeons who removed the subdural empyema via craniotomy. On post-operative day (POD) one, ORL operated and found mainly left-sided disease; via computer-assisted functional endoscopic sinus surgery, openings for ethmoid, maxillary and sphenoid sinuses were restored. The frontal recess was completely blocked and drainage was re-established using drills, accompanied by external access. The frontal intersinus septum was taken down, and a Draf IIa[®] was performed, in which the frontal sinus floor was resected from the lamina papyracea to the middle turbinate, creating a 4x4 mm opening to the left frontal sinus. A surgical drain was fixed through the subcutaneous fistula.

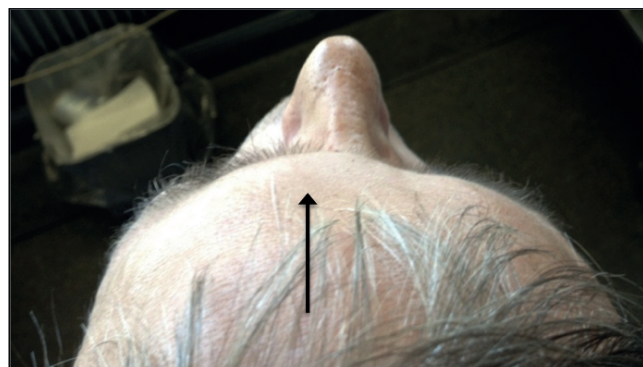


Figure 1. A visible tumour superior to the left eye (at first visit) is shown from above (arrow).



Figure 2. Bloody secretion from a sinocutaneous fistula (at first visit) is shown.

On POD two, a control CT showed residual empyema and severe cerebritis. On POD three, microscopy from the empyema revealed *Streptococcus anginosus* and the antibiotic regimen was changed to moxifloxacin and meropenem. On POD four, infection parameters significantly dropped. The patient developed generalised tonic-clonic seizures during attempts at reducing sedation, and remained heavily sedated until POD five, where he regained consciousness. After extubation on POD seven, he was able to maintain eye contact, but did not speak or move his extremities in the following days. These findings were initially attributed to his mental disorder, however, 32 hours post-extubation (POD eight) a new CT-scan revealed progression of the subdural empyema, increased mass effect and widespread cerebritis of the frontal and temporal lobes. Methylprednisolone was administered, and antibiotic treatment intensified, resulting in clinical improvement over the next few days; the patient began to speak a few words and started moving. As his movement and speech increased, expressive aphasia and a right-sided hemiparesis became evident on POD ten. Non-verbal communication was normalised by POD 20. Despite intravenous antibiotics, no clinical improvement followed for two weeks and blood infecti-

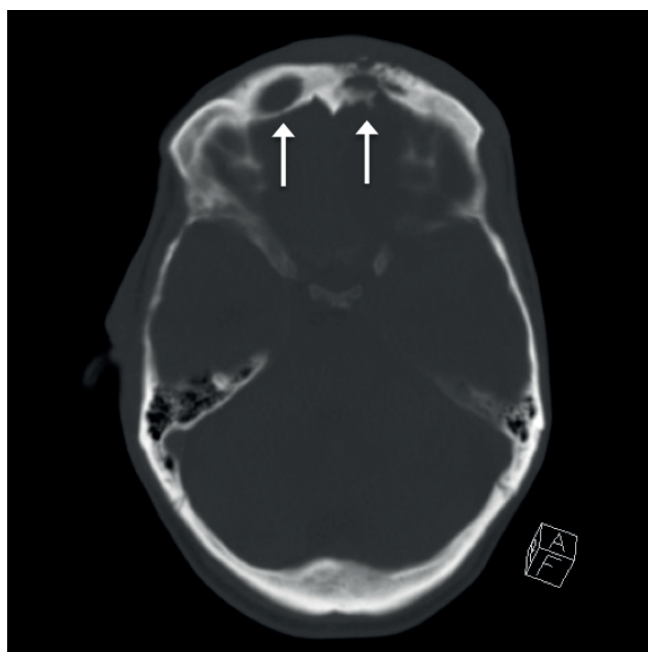


Figure 3. Axial CT scan (at first visit) showing osseous destruction of the posterior wall of the right frontal sinus and osseous destruction of both the anterior and posterior wall of the left frontal sinus (arrows).

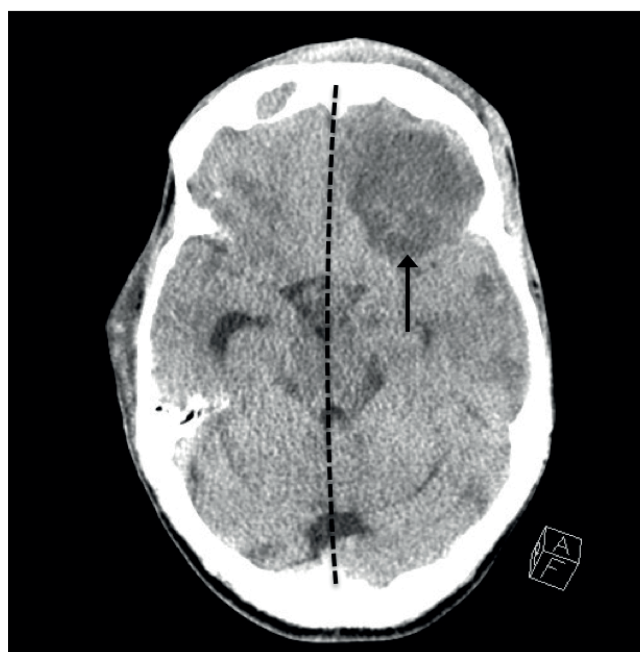


Figure 4. Axial CT scan (at acute and second admission) illustrating a left-sided subdural empyema 45x42mm (arrow) and midline shift (dotted line).

on parameters rose. On POD 35 and 44, neurosurgeons removed the frontal bone affected by osteomyelitis, gaining control of the infection. Antibiotics were administered for four more weeks. The patient was discharged to rehabilitation 67 days after the initial surgery with significant sequelae.

At the last follow-up, 38 months after discharge, sequelae were unchanged and included cognitive deficits, partial expressive dysphasia, a right-sided hemiparesis and epilepsy with monthly seizures. Consequently, his brother now functions as his legal guardian.

Discussion

Here we present—to our knowledge—the first case of a patient suffering from schizophrenia and PPT associated with severe complications.

The case is important for several reasons. First, the case illustrates a non-optimal interplay between a patient with schizophrenia and doctors treating physical disease, and also elucidates how the combination of a severe sinonasal infection with concurrent mental disease can lead to major irreversible impairment. The case adds new knowledge by highlighting the need for special attention and follow-up of a vulnerable and undertreated patient group in otorhinolaryngology. No direct aetiological association between sinonasal infection and schizophrenia has been reported, but several studies have described adverse disease behaviour in patients with schizophrenia^(4,5,9,10). Disease presentation is often atypical, which can com-

plicate or delay the diagnostic process. The patient was initially referred with a fistula from the frontal sinus without subjective symptoms or complaints of sinonasal infection, probably leading to the delayed diagnosis of a suppurative frontal sinusitis with PPT. Following the diagnosis, the patient did not accept the recommended treatment, and he also chose not to follow the invitation for follow-up. He was first seen at an advanced stage of disease with an intracranial spread of the infection.

Pain is defined as an unpleasant sensory and emotional experience associated with actual or potential tissue damage⁽¹¹⁾. Patients with schizophrenia have been hypothesised to have an altered ability to perceive pain, which has been confirmed in experimental and epidemiological studies^(6,7,12–14). Additionally, poor social skills in conjunction with reduced reporting of pain and reduced help-seeking abilities contribute to altered pain-related behaviour^(7,12,15). Finally, fear of hospitalisation causes some patients with schizophrenia to refuse or avoid medical attention⁽³⁾ and misattribution of physical symptoms to concurrent mental disorders by health professionals can lead to under-diagnosis of somatic disease⁽³⁾.

As a result, patients with schizophrenia often receive delayed treatment for somatic disease^(4,5,7,13,15). This emphasises the need for special attention to be paid to somatic disease when treating these patients. Correspondingly, lack of help-seeking behaviour or reporting of symptoms in the presented case, made it possible for the infection to spread intracranially.

Several studies have described similar diagnostic delays of acute somatic disease in patients with schizophrenia. In one such case, a diagnostic delay of meningitis occurred due to absent cardinal symptoms⁽¹⁰⁾. Likewise, in a study of 55 patients with appendicitis, those with schizophrenia were diagnosed later than psychiatrically healthy patients, resulting in higher rates of complications and death⁽⁹⁾. Finally, misleading patient behaviour and distorted pain perception have been considered to contribute to diagnostic delays in acute abdomen and myocardial infarctions^(16,17).

In primary care, patients with schizophrenia frequently receive suboptimal treatment, and when admitted to hospitals they often receive fewer tests and poorer treatment^(18,19). For acute infections that require hospitalisation, patients with schizophrenia are particularly vulnerable and infections in the central nervous system have the worst prognosis⁽¹⁹⁾.

Despite increased focus on comorbidity in schizophrenia, not only acute but also chronic physical disease such as lifestyle diseases is underdiagnosed and undertreated, and life expectancy is reduced by 10–20 years⁽¹⁸⁾.

Danish legislation enables doctors to perform involuntary treatment only if the patient is psychotic or in a similar state and if the disease is immediately life-threatening or substantially damaging to the patient's health⁽²⁰⁾. In the present case, no forced treatment was performed, as the patient was neither in a life-threatening nor psychotic state. However, we hypothesise that a mandatory follow-up regime, including contact with relatives and a general practitioner, could have led to a less severe disease course, as the lack of follow-up likely contributed to the poor outcome.

Conclusions

In challenging cases like the one presented, we suggest taking

a multidisciplinary approach comprising somatic, psychiatric and social interventions in both the primary and hospital sectors in collaboration with the patient and relatives, as this could improve follow-up and treatment.

Acknowledgments

None

Authorship contribution

All authors contributed significantly to the preparation, drafting and writing of the manuscript, and approved the submission.

Conflict of interest

The authors declare that there is no conflict of interest.

Ethics approval

Not applicable.

Consent for publication

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient/parent/guardian/relative of the patient. A copy of the consent form is available for review by the Editor of this journal.

Availability of data and materials

The datasets used during the current study are available from the corresponding author on reasonable request.

Funding

No funding.

Abbreviations

PTT: Pott's puffy tumour; ORL: Otorhinolaryngology; CT: computed tomography; POD: post-operative day.

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