International Journal of Surgery Case Reports 94 (2022) 106975

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

Misdiagnosed epidermoid cyst appears Potts Puffy Tumor: A case report and literature review

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ARTICLE INFO

Keywords:
Case report
Potts Puffy Tumor
Epidermoid cyst

ABSTRACT

Introduction: Although nowadays rare, Potts Puffy Tumor (PPT) is a rare but serious diagnosis characterized by osteomyelitis of the frontal bone. The introduction of broad-spectrum antibiotics made PPT an uncommon diagnosis in modern medicine, and it is mostly seen as a complication of frontal sinusitis in children and adolescents.

Case presentation: We report a case of PPT in an elderly man with a sudden and increasing swelling of the forehead, which was initially diagnosed as an epidermoid cyst. Subsequent surgical exploration revealed osteomyelitis of the underlying frontal bone. Medical imaging by computed tomography (CT) and magnetic resonance imaging (MRI) was done, and treatment with Functional Endoscopic Sinus Surgery (FESS) and reconstructive surgery of the frontal bone was conducted.

Discussion: PPT is a rare, yet serious condition which is usually seen as a complication of chronic sinusitis which causes osteomyelitis and a subperiosteal abscess. Despite the atypical presentation of symptoms, the reported case underlines the importance of rapid deployment of medical imaging, referral, and administration of broad-spectrum antibiotics. Furthermore, surgical intervention is indicated in most cases. When left untreated, PPT can result in severe intracranial complications.

Conclusion: The diagnosis PTT is still relevant and may have a nonspecific presentation. A persisting epidermoid cyst after drainage should lead to the suspicion of a PTT. Immediate imaging and adequate treatment are required to prevent serious, potentially lethal intracranial complications.

1. Introduction

A Potts Puffy Tumor (PPT) is a swelling of the forehead described as a subperiosteal abscess due to osteomyelitis of the underlying frontal bone [1]. The English surgeon Percivall Pott (1714–1788) first described PTT in 1760 [2]. Nowadays, this antiquated diagnosis is uncommon due to the extensive use of broad spectrum antibiotics. If present, PPT is most frequently seen in pediatric or young adolescent patients, and PPT in adults is particularly rare [3]. Immunocompromised individuals are also more prone to develop PPT [4]. Usually it develops as a rare complication of chronic sinusitis. Review of PubMed literature has revealed little cases misdiagnosing PTT as an infected sebaceous cyst [5,6]. We present a case of an elderly man with a frontal skin lesion, which was misdiagnosed as an epidermoid cyst in a community hospital, what appeared to be a frontocutaneous fistula.

This case report has been reported in line with the SCARE Criteria [7].

2. Case presentation

An 84-year-old Caucasian male presented with an intermittently painful skin lesion on the forehead. Three months earlier a progressive swelling appeared, growing up to 3 cm elevation. The patient was known with chronic sinusitis, recurrent purulent rhinorrhea, and sporadic headaches. No fever was present, and no history of head injury. The swelling was priorly diagnosed as an epidermoid cyst in another regional hospital. In an attempt to relieve the cyst, a minimal amount of pus was released. After three weeks, a considerable amount of pus flowed spontaneously from the swelling, and antibiotic treatment with clindamycin was started. The wound was left open for drainage and

Abbreviations: CT, Computed tomography; FESS, Functional Endoscopic Sinus Surgery; MRI, Magnetic resonance imaging; PPT, Potts Puffy Tumor.

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https://doi.org/10.1016/j.ijscr.2022.106975
Received 25 January 2022; Received in revised form 14 March 2022; Accepted 24 March 2022
Available online 29 March 2022

secondary healing was intended. After five weeks, the wound persisted, and the patient was therefore referred to our plastic surgery department for surgical intervention. At time of presentation, we saw a small purulent wound central on the forehead with slightly bloody effusion. Palpation was not painful, and no more swelling and minimal erythema was present (Fig. 1).

Under local anesthesia the skin lesion was excised by an experienced plastic surgeon. Underlying chronic inflammatory tissue was exposed, and the frontalis muscle was also affected. Deep underneath the inflammatory tissue, an opening in the frontal cranial bone was palpable. The round opening with smooth bony edges appeared to have a connection to the left frontal sinus (Fig. 2).

In consultation with the department of Otorhinolaryngology, a computed tomography (CT) scan of the sinuses was performed (Fig. 3). The imaging showed an air-fluid level in the left frontal sinus and complete veiling of the left maxillary sinus. Additionally, a ventral defect in the left frontal sinus with a frontocutaneous fistula, as well as a small osseous defect in the posterior wall of the frontal sinus was visible. Antibiotic treatment with amoxicillin/clavulanic acid (625 mg tablets three times daily for two weeks) and nasal drops were started, and magnetic resonance imaging (MRI) of the brain was performed. In addition to the osseous defects, the MRI showed extensive pathological sinusitis in the paranasal sinuses, while no sign of intracranial perforation was observed. The patient was referred to the department of Otorhinolaryngology of a university medical center for further surgical treatment. They performed Functional Endoscopic Sinus Surgery (FESS) with reconstruction of the anterior wall of the frontal bone with a titanium plate. After nine days we saw the patients, and the postoperative course went without complications and the patient was doing well (Fig. 4). The department of Otorhinolaryngology prescribed nasal drops and scheduled multiple follow-up appointments at the outpatient clinic.

3. Discussion

PPT can result from infected open head trauma, but usually it is a rare complication of chronic frontal sinusitis [3,5]. In chronic sinusitis

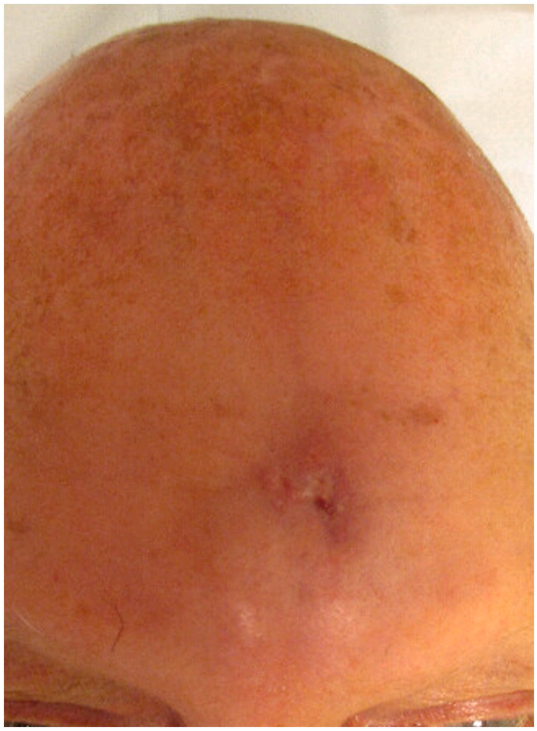

Fig. 1. Frontal skin lesion paramedian left at the time of presentation.

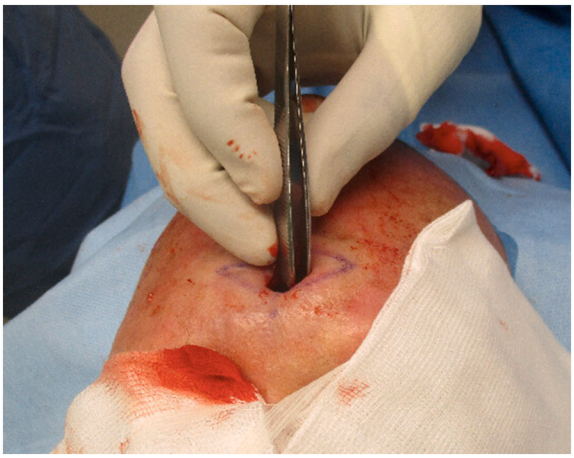

Fig. 2. Perioperative exploration of the defect with forceps, approximately 3 cm deep.

the infection spreads hematogenously, leading to a reduced blood flow, further causing secondary thrombophlebitis in the respective area. This can result in bone necrosis, ultimately leading to the typical manifestation of osteomyelitis [5]. Intracranial complications, such as subdural or epidural empyema, arise form intracranial involvement. These complications can be explained by the fact that mucosa of the frontal sinuses is drained by the diploic veins which in turn are connected to the dural vein plexuses [3,8].

Early diagnosis of PPT is crucial in preventing intracranial complications and therefore reducing morbidity and mortality. PPT is most often characterized by headache, frontal swelling, rhinorrhea and fever. However, the absence of fever is not uncommon, and other symptoms may also vary [4,8]. In the reported case, the patient had no history of fever.

Besides PPT, the differential diagnosis of a frontal swelling includes epidermoid cyst, lipoma, dermoid cyst, infected hematoma, and benign or malignant tumors of the skin, bone or frontal sinuses [9]. At the time of presentation, the frontal swelling of the patient was no longer present, and the clinical differential diagnosis had been reduced to a drained cyst. Referral from another hospital and the atypical presentation could explain the delay in recognition and diagnosis. Also, the condition PPT was unknown to the healthcare professionals involved. Our case report shows an atypical course of symptoms which could raise awareness for the clinical entity and can potentially help prevent future delays in diagnostics.

If there is any suspicion for PPT, imaging should be conducted. A CT-scan is the most important imaging modality for early diagnosis of PTT. Magnetic resonance imaging can provide additional information on intracranial involvement, since MRI gives a better insight into soft tissue injuries [10].

In case of PPT, treatment with broad-spectrum antibiotics should be started, preferably intravenously [6]. It is recommended to take microbiological samples and laboratory tests, which is not done in the presented case at time of presentation. Generally surgical intervention is indicated to prevent further spread of the infection by drainage of the sinuses and removal of osteomyelitic bone and granulation tissue. An external approach is fast and relatively simple but gives an aesthetically less gratifying result. An increased use of intranasal endoscopic sinus surgery has resulted in less morbidity rates and a reduced recovery time [1,6]. If there is intracranial involvement, surgical and antibiotic therapy are required, and neurosurgical intervention might be necessary [11].

Serious intracranial complications associated with PPT include epidural and subdural empyema, meningitis, brain abscess, sinus thrombosis, orbital cellulitis, subarachnoid inflammation, and

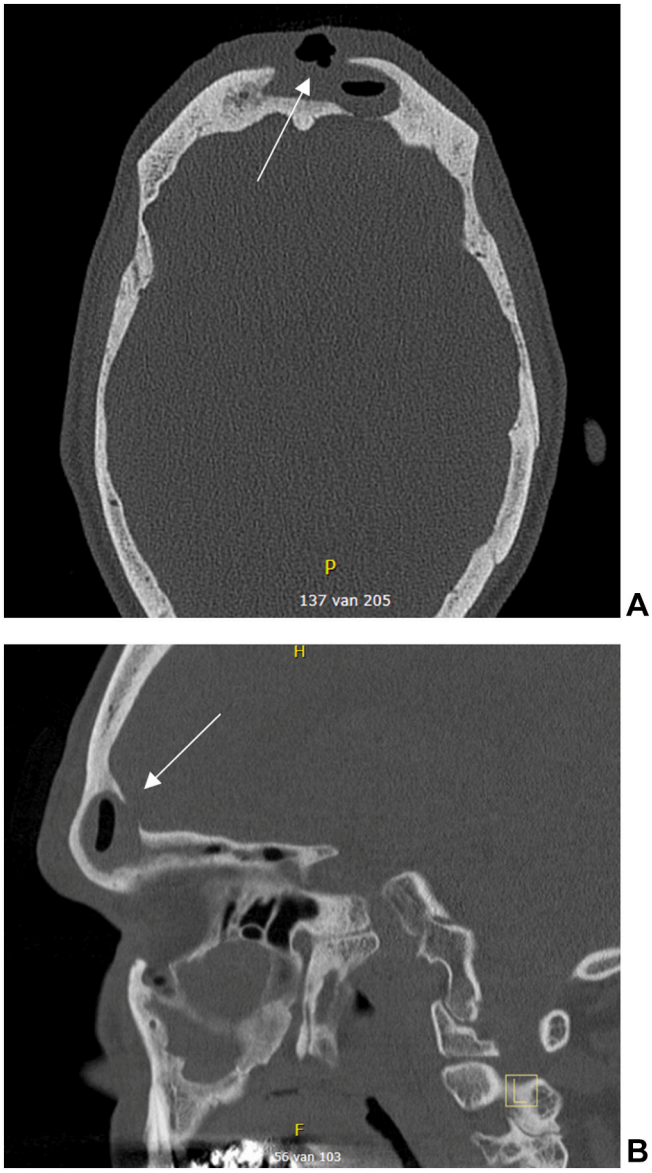

Fig. 3. A) Axial view of the CT of the sinuses indicates a ventral defect with a diameter of approximately 2 × 1.5 cm. B) Sagittal view of the CT showing an osseous defect of approximately 5 mm in de dorsal wall of the left frontal sinus.

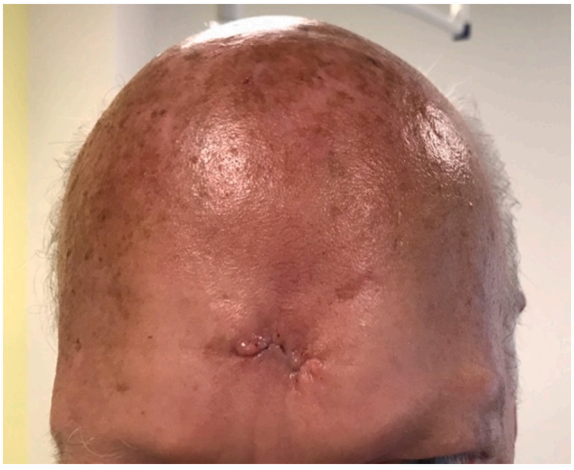

Fig. 4. Frontal scar nine days after FESS.

infraorbital abscess [5]. These intracranial complications can be missed due to an asymptomatic course, especially when patients are on antibiotics, which can mask the symptoms. Signs of intracranial involvement include altered mental status, nausea, vomiting, papilledema, and nuchal rigidity. More advanced findings include focal neurological signs such as seizures and hemiparesis [12].

Most patients suffering from PTT recover without any neurological residual symptoms. Since the broad use of antibiotics, mortality has dropped from 60% to 3.7% [6]. However, due to the risk of serious intracranial complications, early diagnosis and treatment of PPT is vital [9].

4. Conclusion

In conclusion, PPT is presented with nonspecific symptoms and, if untreated, serious complications may occur. Our case shows that in modern medicine a frontocutaneous fistula can still occur, even at an older age. Swelling of the forehead may already have disappeared, but the osteomyelitis may remain present subcutaneously. An atypically presenting, previously drained epidermoid cyst, should put a PPT in the differential diagnosis. In order to prevent serious intracranial complications associated with PPT, acute imaging, antibiotic treatment and adequate surgery is crucial.

Sources of funding

No funding has taken place.

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. The analysis of one case report is intended to develop information to be shared for medical or educational purposes. Therefore, in consultation with the local medical ethical committee, it does not meet the criteria of research and it does not require any kind of ethical review.

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 by the Editor-in-Chief of this journal on request.

Guarantor

Drs. J. Sawor.

Registration of research studies

This is no First in Man case report.

Provenance and peer review

Not commissioned, externally peer-reviewed.

CRediT authorship contribution statement

Roos Salemans	Conceptualization, data extraction, interpretation of data, drafting and revising the manuscript, approval of final manuscript
Ennie Bijkerk	Conceptualization, interpretation of data, drafting and revising the manuscript, approval of manuscript
John Sawor	

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(*continued*)

	Plastic surgeon, conceptualization, supervision, revising and editing the manuscript, approval of final manuscript
Andrzej Piatkowski	Plastic surgeon, revising and editing the manuscript, approval of final manuscript

Declaration of competing interest

There were no conflicts of interest.

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