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Case Report

A rare case report: Pott's Puffy tumor and Lemierre's syndrome with intracranial complications in an adult male ☆

Ines Azouz[a,*], *Marwa Moussaoui*[a], *Dhia Dahech*[a], *Rihab Jdidi*[b], *Amene Aissa*[a]

[a] *Department of Radiology, Ibn Jazzar Regional Hospital (Kairouan), Faculty of Medicine of Sousse, Tunisia*
[b] *Department of Neurosurgery, Ibn Jazzar Regional Hospital (Kairouan), Faculty of Medecine of Sousse, Tunisia*

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ABSTRACT

Pott Puffy Tumor (PPT) is extremely rare, yet potentially severe condition characterized by osteomyelitis of the frontal bone associated with one or multiple subperiosteal abscesses, primarily from nasosinusitis. It is characterized by localized frontal swelling accompanied by a subperiosteal abscess. Clinicians and radiologists do not widely recognize this complication of frontal sinusitis and, hence it is likely to be overlooked in clinical practice.

We describe, through this article, a unique case of frontal osteomyelitis in an immunocompromised patient, complicated by a subperiosteal orbital abscess, subdural empyema, intracerebral abscesses, superior sagittal sinus thrombosis, and pulmonary septic emboli, which also resulted in septic pulmonary infarcts, aligning with Lemierre's syndrome. Treatment included a combination of antibiotics and surgery, with careful monitoring for orbital and intracranial complications.

Introduction

PPT was initially described by English neurosurgeon Percivall Pott in the 18th century following a case of brain trauma [1]. Since then, more than 300 cases have been reported mainly attributed to sinusitis [2]. Clinical presentation can be deceptive, particularly in children and immunocompromised individuals. This condition can lead to significant neurological complications that may be life-threatening. Intracranial complications are the most feared and occur in approximately 60 to 85% of patients [3]. A thorough understanding of this complication allows for early diagnosis and adequate treatment for each specific complication, which is the only way to ensure a better prognosis. Both broad-spectrum antibiotic therapy and surgical intervention are required for effective management.

We report a case of Pott's puffy tumor as a complication of acute on chronic odontogenic sinusitis, with associated periorbital cellulitis, multiple intracranial and pulmonary complications, and Lemierre's syndrome.

Abbreviations: CRP, C reactive protein; CT, Computed tomography; MRI, Magnetic resonance imaging; PPF, Pott Puffy Tumor.

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* Corresponding author.
E-mail address: ines.azouz@etudiant-fmt.utm.tn (I. Azouz).

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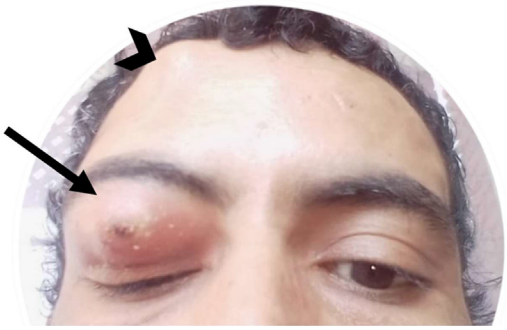

Fig. 1 – A photo of the patient on the day of admission shows an orbital swelling with pustules within it (black arrow), associated with a homolateral forehead swelling (black arrow head).

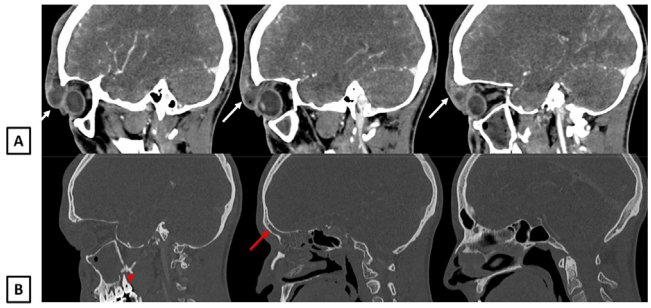

Fig. 2 – In (A) sagittal CT scan in parenchymal window revealing a preseptal orbital collection, with extension into extra-conical fat beneath the orbital roof communicating with the right frontal subgaleal collection (white arrow). In (B): Sagittal CT scan in bone window showing complete pansinus filling with thickening of the maxillary sinus walls, contrasting with thinning of the frontal sinus wall (red arrow). The buccosinus communication is indicated by the red arrowhead adjacent to tooth 18.

Observation

A 39-year-old man with a history of chronic sinusitis was admitted to treatment of right orbital swelling associated with headaches. The patient is reported to be a heavy smoker and drinker, as well as, a drug and cannabis user. No history of nasal drug use, was reported. In the emergency room, the patient reported multiple consultations for severe headaches, which had worsened 10 days before his admission. Additionally, he had blurred vision and fever. The patient revealed that he has already been treated for chronic sinusitis but was lost to follow-up due to poor socioeconomic conditions. He had consulted a private physician for the worsening of his headaches, who prescribed oral corticosteroids. Since his symptoms didn't improve, he sought care at the emergency room, resulting in his admission.

On physical examination, the patient had a right superior orbital swelling, reddish with pustules containing serous fluid and pus, as well as a homolateral frontal base swelling both being tender on palpation (Fig. 1).

Clinical examination revealed tachycardia to 120, a blood pressure of 10/8 mmHg, a respiratory rate of 20 breaths per minute, oxygen saturation of 97% on room air, and a temperature of 40°.

Neurological exam showed: patient was conscious with a Glasgow coma scale score of 15, had sensitivity to light reflex, and a soft neck. Otherwise, the neurological exam was normal.

The lab results showed hyperleukocytosis at 13,000/mm^3, predominantly with neutrophils, and a CRP level of 30 mg/L.

The diagnosis of orbital cellulitis was established, and an emergency cerebrofacial CT scan was performed to identify a subperiosteal abscess. The initial CT scan revealed acute pansinusitis associated with a preseptal collection extending into the extra-conical orbital fat, forming a subperiosteal abscess located on the orbital roof. The preseptal collection communicated with another frontal subgaleal collection on the right side (Fig. 2). The CT scan also revealed a bilateral frontal subdural collection measuring 15 mm in thickness, with meningeal enhancement and hypodensity of the right frontal lobe without a well-defined collection, in addition to a superior sagittal sinus thrombus (Fig. 3). Bone window analysis showed signs of chronic right maxillary sinusitis with thickened sinus walls, in contrast to very thin walls of the right frontal sinus compared to the opposite side and the status of chronic sinusitis. Multiple dental granulomas were identified, along with a right buccosinus communication related to the tooth 18 (Fig. 2). The diagnosis of acute-on-chronic anterior pansinusitis of dental origin was confirmed, with a subgaleal collection and a subdural empyema developing on both sides of the frontal sinus. This represents a classic, yet rare complication of frontal sinusitis known as "Pott's puffy tumor", along with orbital complications and intracranial extension.

The patient was admitted to the neurosurgery department for drainage of the subdural empyema. Empirical triple antibiotic therapy was established, including vancomycin, cefotaxime, and metronidazole. The patient underwent two surgical interventions: the first involved evacuating a subgaleal collection, creating a right frontal bone flap with the release of pus from the bone. There was an infection with the presence of false membranes, leading to the opening of the dura mater, which released pus, followed by bacteriological sampling and irrigation. During the second procedure, the dura mater was found to be destroyed, and the intraoperative samples came back negative. The follow-up CT scan a few days after the second surgical intervention showed the appearance of a right frontal abscess, indicated by a well-defined and enhanced frontal collection with a central liquid component. A significant perilesional edema was noted, causing a deviation of the midline and subfalcine herniation. Meanwhile, there was a notable reduction in the orbital subperiosteal abscess and the empyema. A subsequent brain MRI confirmed the presence of three intracranial collections, the largest measuring 46×42 mm, accompanied by significant perilesional edema and signs of herniation. Partial recanalization of the superior sagittal sinus was also noted (Fig. 4).

A contrast-enhanced chest CT scan was performed a week later, due to the sudden onset of dyspnea during hospital-

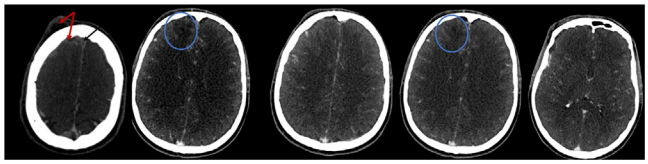

Fig. 3 – Axial CT slices with contrast revealing a collection extending on both sides of the right frontal bone, involving a subgaleal collection on one side and an empyema on the other (red arrows). There is thrombosis of the superior sagittal sinus (black arrow) and a right frontal hypodensity without a defined wall, consistent with a presuppurative focus (blue circle).

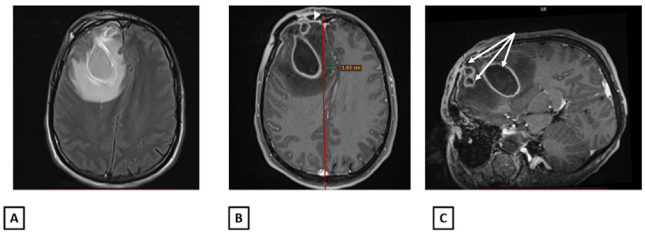

Fig. 4 – Contrast-enhanched Axial T2 FLAIR sequence in (A), contrast-enhanced axial and sagittal T1 sequence in (B), and (C) respectively showing the presence of 3 well-defined right frontal intraparenchymal collections with enhanced walls (white arrows), accompanied with significant perilesional edema, causing a 16mm leftward midline shift. Note the presence of the craniotomy flap with associated meningeal enhancement (white arrowhead).

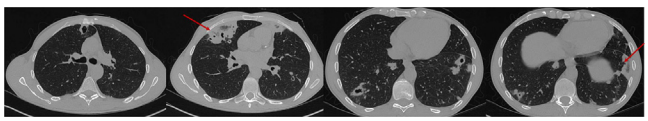

Fig. 5 – Axial CT slices in lung windows revealing triangular peripheral-based pulmonary consolidations with hilar apices and clear centers, displaying a "bubbly" pattern surrounded by a halo consistent with the inverted halo sign indicative of pulmonary infarcts (red arrows). Additionally, scattered cavitated nodules are noted, corresponding to septic emboli, which are more numerous in the lung bases (which are more vascularized).

ization, to rule out a potential pulmonary embolism. The scan revealed diffuse cavitary nodules, mainly located in the lower lung bases, consistent with abscesses. Additionally, peripheral triangular consolidations were observed, with bases oriented toward the pleura and apices toward the hilae. These consolidations showed central lucency, described as "bubbly consolidation'', and an inverted halo sign, suggesting pulmonary infarctions. The combination of an infection in the head and neck region, thrombosis of an intracranial vein, and the presence of septic emboli led to a diagnosis of Lemierre's syndrome (Fig. 5). However, all bacteriological samples were negative, probably due to prior antibiotic therapy. The patient's condition improved both clinically and biologically after surgical treatment, with a favorable outcome under antibiotic therapy.

Discussion

The exact frequency of the Pott Puffy Tumor is hitherto unknown due to the rarity of the condition. The available data in the literature is primarily based on case reports. However, recent studies have highlighted an increase in the number of cases reported in the literature, attributing this rise to advancements in imaging techniques, improved awareness of this condition and its complications, as well as the possible development of antibiotic resistance [4].

PPT usually results from untreated rhinosinusitis, but can also be caused by direct head trauma, substance use, and odontogenic disease, as observed in our case. Infections are usually polymicrobial, with a microbiome primarily consisting of anaerobic bacteria. It is commonly observed in children and adolescents which can be explained by the increased flow in the diploic vein during this period [5–7]. This heightened flow leads to looser connections between the frontal sinus and the bone marrow space [3]. Persistent bacterial overgrowth in the frontal sinus cavity and adjacent soft tissues leads to small vessel thrombosis and venous congestion. This disrupts the frontal periosteal blood supply, initiating an inflammatory reaction characterized by increased intraosseous pressure and extensive necrosis of the trabecular bone matrix. The resulting avascular and ischemic conditions shift the environment from aerobic to anaerobic, causing the growth of opportunistic microorganisms and leading to the formation of abscesses and cortical sinus tracts [2,5,8].

Clinical presentation of PPT is diverse, with the most frequent symptoms being forehead swelling, frontal headache, fever, periorbital edema or erythema, and rhinorrhea. The presence of neurological signs such as headaches, seizures, meningeal syndrome, vision changes, or localized neurological signs should raise concerns about intracranial complications. The rate of intracranial complications in pediatric and adolescent patients with PPT is high, occurring in nearly 72% of cases [4]. Meanwhile, intracranial complications can include epidural abscess, subdural empyema, brain abscess, and sinus thrombosis. In 21% of cases, multiple intracranial complications may coexist [2], highlighting the crucial need for early diagnosis and prompt management to effectively prevent and address these complications. The diagnosis of PPT and its potential complications relies on both clinical evaluation and radiological findings.

Extracranial complications, such as orbital infections, not only frequently co-occur with intracranial disease but also often dominate the clinical presentation in pediatric patients. When intracranial PPT presents without neurological symptoms, these extracranial issues can lead to misdiagnosis and delays in both diagnosis and treatment. For instance, orbital involvement - as seen in our case - is common and has been reported in 29% of cases in the literature [2]. The use of

corticosteroids and our patient's immune status could explain the spread of the infection and the delay in management.

Diagnosis relies on clinical presentation, typically a history of rhinosinusitis, and emergency imaging, as the prognosis of PPT hinges on timely intervention. Contrast-enhanced cerebral CT is the investigation of choice to confirm the diagnosis, showing features such as frontal sinusitis, osteomyelitis with bone erosion, and subperiosteal abscesses. Intracranial complications include subdural or epidural empyema, most notably frontal brain abscesses, as well as sinus thrombosis and acute meningitis. All these complications can be diagnosed with contrast-enhanced cerebral CT. If available in an emergency setting, contrast-enhanced brain MRI may be the preferred imaging modality, as it offers superior visualization of soft tissues and is considered the gold standard for diagnosing intracranial complications [8,9].

Lemierre syndrome, on the other hand, is a rare complication with up to 5.5 cases per million reported in 2017 [10]. It primarily results from oropharyngeal infections and involves septic thrombosis of the jugular vein accompanied by septic emboli, usually in the context of sepsis. Isolation of Fusobacterium necrophorum or jugular vein thrombosis is not essential for diagnosis. More inclusive definitions and variants in the literature encompass a broader range of head and neck infectious foci beyond the tonsils, as well as a wide spectrum of thromboembolic complications, from cerebral sinus veins to the carotid artery, which may occur alone or in conjunction with internal jugular vein thrombosis [11]. To our knowledge, this is the only reported case of Pott's puffy tumor complicated by Lemierre's syndrome, which makes it particularly unique, given that both conditions are rare complications individually. The multitude of complications in this patient could be attributed to immunosuppression, substance abuse, poor socioeconomic and oral health, and prior corticosteroid use, which contributed to a significant delay in diagnosis.

Managing PPT frequently demands an interdisciplinary approach involving specialists in infectious diseases, otorhinolaryngology, neurosurgery, and ophthalmology, tailored to the complications encountered during hospitalization and follow-up. The treatment involves antibiotic therapy with effective central nervous system penetration and management of both intra- and extracranial collections [12]. Collections can be drained using endoscopic techniques (endoscopic sinus surgery), an open approach, or a combination of both methods [13]. In this patient, surgical drainage was achieved through a craniotomy. Surgical drainage and prolonged antibiotic therapy lead to complete recovery in 62.5% of cases, with a mortality rate estimated at 12% and neurological complications occurring in 12%-40% of cases [9]. The treatment also requires addressing the primary source of infection, specifically chronic sinusitis and, in the case of our patient, odontogenic infection which prevents a vicious cycle and reduces the risk of recurrence.

Conclusion

Pott's Puffy Tumor is indeed an uncommon condition, but it should be systematically considered when encountering frontal swelling, especially in adolescents and young adults, as it can be caused by various factors. Prompt diagnosis and management with a multidisciplinary approach are crucial. Imaging plays a key role in the diagnostic process, with CT scans often being the first-line tool to assess bony involvement and the extent of osteomyelitis. MRI, on the other hand, provides superior visualization of soft tissue, detecting abscess formation and potential intracranial complications such as subdural empyema or brain abscess. Identifying these complications is essential, as they may modify therapeutic measures, including the urgency of surgical intervention. Treatment typically involves a combination of intravenous broad-spectrum antibiotics and surgical drainage or debridement. Early intervention is key to achieving a favorable outcome and minimizing the risk of severe complications.

Patient consent

I, author of this article can confirm that we have obtained written, informed consent from the patient for the publication of his case, including the disclosure of his photo, results from biological and imaging tests, while ensuring that no further personal information will be disclosed. This consent is in compliance with the necessary ethical and privacy considerations for the preparation and submission of our case report article.

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