CASE REPORT

A rare case of Pott's puffy tumor by *Curvularia* in a young immunocompetent patient: uncommon and diagnostically challenging

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Abstract

Pott's Puffy Tumor (PPT) denotes frontal bone osteomyelitis with the formation of a subperiosteal abscess; it can lead to grave complications if left untreated. It is most commonly caused by bacteria. There are very sparse cases of fungal PPT, and no case is reported with *Curvularia* in an immunocompetent patient. A 20-year-old male patient complained of progressive forehead swelling and occasional forehead pain with mucoid, non-foul-smelling, non-bloody intermittent nasal discharge for the last 6 months. CT showed focally eroded frontal bone. The patient was managed surgically; histopathology and microbiological examinations showed a fungal pathogen. Matrix-Assisted Laser Desorption/Ionization Time-of-Flight Mass Spectrometry identified the fungus *Curvularia*. The patient received antifungal treatment and responded very well. Finding out the underlying etiology and pathogen is of utmost importance to provide curative treatment and prevent future relapses. Though a fungus is a rare cause of PPT, a high level of suspicion and histopathology and microbiological examinations are critical to provide curative treatment.

Keywords: Pott's Puffy Tumor, *Curvularia*, Osteomyelitis, Fungus, Immunocompetent, Sinusitis

Introduction

Pott's Puffy Tumor (PPT) is defined as the frontal bone osteomyelitis and subsequent formation of a subperiosteal abscess. The etiology includes frontal and ethmoidal sinus inflammation / infection or trauma. The common pathogens responsible for PPT are mostly bacterial, with very few cases fungal. The commonly reported pathogens are *Streptococcus species*, *Staphylococcus species*, *Pseudomonas aeruginosa*, and mucormycosis; less common pathogens are *Haemophilus influenzae*, *Bacteroides* and *Aspergillus flavus* [1].

Very few cases of PPT associated with fungal etiology are reported, and most of the patients had an immunocompromised state. The documented common fungus associated with PPT in immuno-compromised patients is *Mucor* [2]. In the literature, very few cases of fungal PPT in immunocompetent patients have been reported and are associated with *Aspergillus* [3]. So far, there are no reports of PPT associated with *Curvularia* species. We are reporting this unique, diagnostically challenging, and possibly the first case of PPT associated with the unusual fungal pathogen *Curvularia*.

Case Report

A 20-year-old male patient with a history of forehead swelling and nasal discharge for the last 6 months presented to otorhinolaryngology out-patient department. The swelling was more on the left than the right side, which was insidious in onset and progressive. It was associated with occasional dull aching pain in the forehead region. The nasal discharge was insidious in onset and was inter-mittent. The nasal discharge was more from the left

side than the right side. The discharge was mucoid in consistency and relieved by over the counter medications. The discharge was never foul smelling or purulent. The patient also complained of occasional left nasal blockage, which was not bothering him. There was no fever, anosmia, hyposmia, or bleeding from the nose. He had no history of any visual disturbances or neurological symptoms. The patient denied any history of trauma or previous sinus surgery. There were no significant comorbid conditions, and personal and family histories were insignificant. While examining, there was a single, oval, diffuse swelling over the region of the frontal sinus, about 5 × 4 cm, more on the left side as shown in Figure 1.

It was firm and doughy in consistency and warm and tender on palpation. Anterior rhinoscopy examination revealed a pale polypoidal mass in the left middle meatus, mucoid discharge, and a deviated septum on the right side. No signs of orbital involvement were present. Diagnostic nasal endoscopy confirmed the finding of a polypoidal mass in the left osteomeatal complex with mucoid discharge and a deviated septum to the right. The blood reports were found within normal limits. A Non-Contrast Computed-Tomography (NCCT) scan of the paranasal sinuses revealed bilateral frontal sinus opacification, more marked on the left, with hyperdense material within. There were focal areas of dehiscence of the anterior wall of the left frontal sinus, with extension of the disease into the subcutaneous plane, as seen in Figure 2.

There was also involvement of the ethmoidal sinuses, with rarefaction and demineralization of the left lamina papyracea. Bilateral mild maxillary sinusitis was also present. There was no intracranial extension of the disease found in the CT scan of the brain. These findings pointed towards an aggressive chronic sinus infection, possibly fungal in etiology, with erosion of surrounding bony structures. The surgery was planned under general anesthesia.

A combined external and endoscopic approach was performed to remove the disease. The disease was approached externally by a brow line incision and elevation of the subperiosteal flaps. The anterior table of the frontal sinus was thinned out and showed erosions intraoperatively, as shown in Figure 3.

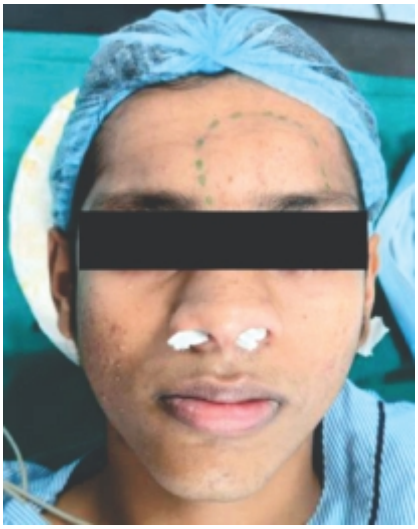

Figure 1: Left forehead swelling

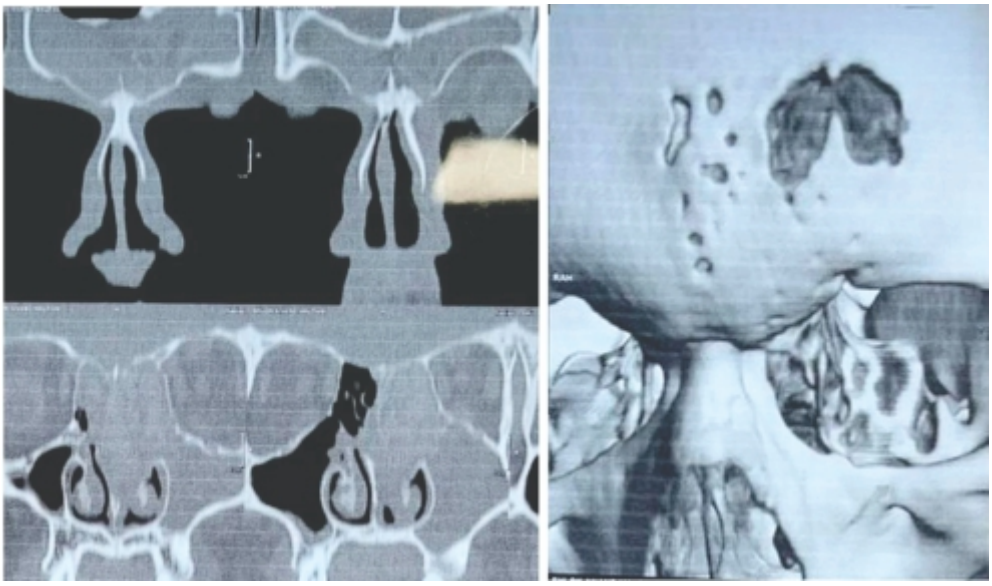

Figure 2: CT images showing frontal, ethmoidal and maxillary sinusitis with hyperdense material within and 3D reconstruction showing eroded frontal outer table

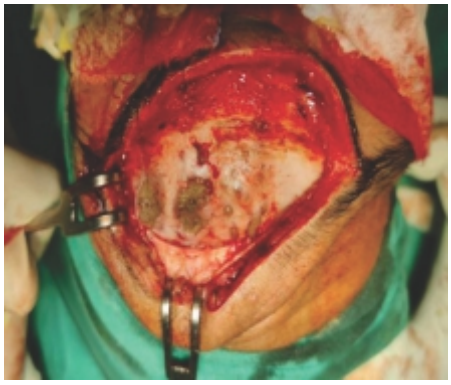

Figure 3: Intraoperative picture showing grayish material coming out through eroded outer table of frontal bone after elevating subperiosteal flap

Cheesy, necrotic material and inflamed mucosa were evacuated and sent for histopathological and microbiological examinations. The frontal sinus was thoroughly debrided, irrigated, and marsupialized into the nasal cavity. Functional endoscopic sinus surgery was performed to remove the disease from all other involved sinuses. The frontal recess was cleared, and wide drainage of the frontal sinus was achieved. After thorough irrigation, the wound was closed in layers. Standard postoperative care was taken. The histopathology report revealed allergic mucin, exhibiting a layered appearance of mucin admixed with cellular debris, sloughed epithelium, and Charcot-Leyden crystals. A few fragments of sinonasal mucosa with chronic inflammation, composed of lymphocytes, plasma

cells, and eosinophils, were observed. There was no granuloma or malignancy. Grocott's methenamine silver and Periodic acid–Schiff special stains revealed fungal hyphae: there were slender, septate, branching hyphae, as well as broad aseptate hyphae, seen. Microbiological examination and potassium hydroxide mount showed the presence of filamentous fungal hyphae. No growth was found on the initial culture for 7 days, and it was subjected to further incubation for 14 days. The culture became positive for fungal growth after 14 days, but the species was difficult to determine. Matrix-Assisted Laser Desorption/Ionization Time-of-Flight Mass Spectrometry (MALDI-TOF MS) was used to identify the fungus, and it was *Curvularia*. The patient had an uneventful postoperative recovery. He was started on empirical antifungal therapy pending final culture results. Once *Curvularia* was confirmed, voriconazole was continued under the guidance of an infectious disease specialist. At 6 months of follow-up, he remained asymptomatic with full resolution of forehead swelling and nasal symptoms. Endoscopic examination showed a well-healed frontal sinusotomy with no residual disease.

Discussion

PPT is a potentially life-threatening complication of frontal sinusitis. It represents the frontal bone osteomyelitis and the formation of an abscess in the subperiosteal plane. If neglected, untreated disease can spread into the cranium either by the venous route or via osteomyelitis of the frontal bone posterior table. In intracranial involvement, a patient may present with subdural empyema, meningitis, epidural, subdural, or intraparenchymal abscess, or cortical vein thrombosis [4]. Therefore, prompt surgical and medical treatment is warranted.

In most cases, PPT is attributed to bacterial infection of the frontal-ethmoidal sinuses, with fungal infections being responsible for very few cases. There are very sparse cases of PPT due to fungal etiology in the literature. Reported cases of fungal osteomyelitis of the frontal bone were seen in immunocompromised patients, and the common pathogens were *Mucor* and *Aspergillus* [1]. *Aspergillus* causing a fulminant sino-orbital disease in a diabetic patient following facial trauma is also documented in literature [5].

There is a case report of maxillary sinus osteomyelitis due to *Curvularia* in a patient with type 1 diabetes mellitus [6]. Another case of maxillary osteomyelitis was reported due to *Mucor* in coronavirus infection with prediabetic status [7]. Cases of allergic fungal rhinosinusitis in immunocompetent patients due to *Curvularia* have been documented [8,9]. No case of PPT in an immunocompetent patient caused by *Curvularia* has been described in the literature. A high level of suspicion, clinical history, and radiology help in diagnosing fungal etiology. At times, it is hard to cultivate a positive fungal culture, and newer modalities like MALDI-TOF MS could help in identifying the pathogen. By identifying the species, specific antifungal agents could help treat the disease and prevent future relapses. In cases of PPT, aggressive surgical debridement and medical management are required.

Conclusion

PPT itself is a rare disease, and due to an unusual fungus like *Curvularia* in an immunocompetent patient, it is a truly odd one. A high level of suspicion and intraoperative findings can provide a clue to the fungal etiology. All surgical specimens should be thoroughly examined, and microbiological cultures

are desirable to identify the right pathogen. Newer modalities of species identification may help in odd clinical scenarios. This will guide us to definitive postoperative management of the etiological organisms and help to cure the condition in patients and also to prevent future relapses that may require revision surgeries.

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