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Suspected Pott's Puffy Tumour in a Child With Osteogenesis Imperfecta

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Abstract

Pott's puffy tumour is a rare complication of rhinosinusitis, classically characterized by frontal bone osteomyelitis and an associated subperiosteal abscess. We report the case of a boy in his early teens with genetically confirmed COL1A1-related osteogenesis imperfecta who presented with progressive forehead swelling and fever. CT demonstrated left frontal and ethmoid sinus disease with associated frontal soft tissue swelling, without definite evidence of frontal bone osteomyelitis or a bony defect and without orbital or intracranial extension. Following specialist radiological and multidisciplinary review, the overall clinical and imaging findings were considered consistent with suspected PPT. The patient underwent endoscopic sinus surgery and antibiotic treatment, with complete clinical resolution. This case highlights the diagnostic challenges of PPT when classical imaging features are absent. It also raises the question of whether altered bone structure and collagen biology in osteogenesis imperfecta may be relevant to the development or spread of sinonasal infection, although any association remains unestablished.

Categories: Otolaryngology
Keywords: acute rhinosinusitis, endoscopic sinus surgery, frontal swelling, osteogenesis imperfecta, pott's puffy tumour

Introduction

Pott's puffy tumour (PPT) is an uncommon but potentially serious complication of frontal sinusitis, characterized by frontal bone osteomyelitis and an associated subperiosteal abscess [1-3]. It occurs predominantly in children and adolescents and commonly presents with frontal swelling, headache, and fever [1,2,4]. Infection can extend beyond the frontal sinus, resulting in orbital or intracranial complications, including epidural abscess, subdural empyema, and cerebral abscess [1-3]. Reviews of paediatric PPT cases have reported intracranial complications in a substantial proportion of patients, highlighting the potential severity of this condition and the importance of early recognition [1,2,4]. Recent international consensus recommendations also emphasize the importance of multidisciplinary assessment and management in these patients [5].

Osteogenesis imperfecta (OI) is a genetic connective tissue disorder most commonly caused by pathogenic variants affecting type I collagen, particularly those in COL1A1 and COL1A2 [6]. These abnormalities affect not only bone strength but also its microarchitecture, including cortical thickness, trabecular structure, and bone porosity [7]. Although OI is primarily recognized for its skeletal manifestations, its effects on connective tissue and bone structure may have wider clinical implications. Diagnosis can be challenging when classical radiological features, particularly frontal bone erosion or a clearly defined subperiosteal abscess, are absent.

We describe a child with genetically confirmed OI who presented with frontal swelling in the setting of acute rhinosinusitis and was managed for suspected PPT. We also discuss the diagnostic challenges associated with establishing the diagnosis in the absence of definitive radiological findings and the possible relevance of altered bone structure in OI.

Case Presentation

A boy in his early teens presented with progressive forehead swelling and discomfort. The swelling developed over the course of one day while he was abroad and initially improved with simple analgesia. Over the following days, he developed a fever and increasing frontal pain. He reported no visual disturbance, diplopia, rhinorrhoea, or neurological symptoms.

His medical history included genetically confirmed OI due to a de novo pathogenic heterozygous COL1A1 splice-site variant (c.3531+2T>C). He had sustained multiple fractures throughout childhood and received pamidronate infusions every three months. Other medical history included attention-deficit/hyperactivity disorder and joint hypermobility.

Examination revealed a tender midline frontal swelling without erythema. There was no periorbital oedema,

ophthalmoplegia, or neurological deficit. Nasal endoscopy demonstrated mucopus within the left nasal cavity.

Initial blood tests showed a mildly raised white cell count and a CRP of approximately 30 mg/L. No microbiological investigations were performed. CT demonstrated extensive unilateral sinus disease involving the left frontal and ethmoid sinuses, with associated frontal soft tissue swelling. There was no definite evidence of frontal bone osteomyelitis or an obvious bony defect and no orbital or intracranial extension (Figures *1*, *2*). Specialist radiological review raised the possibility of a small associated collection. Although the classical radiological features of PPT were not definitively demonstrated, the combination of fever, tender frontal swelling, and underlying frontal sinus disease, together with the possibility of a small collection on specialist review, supported the multidisciplinary team's working diagnosis of suspected PPT. MRI was not performed.

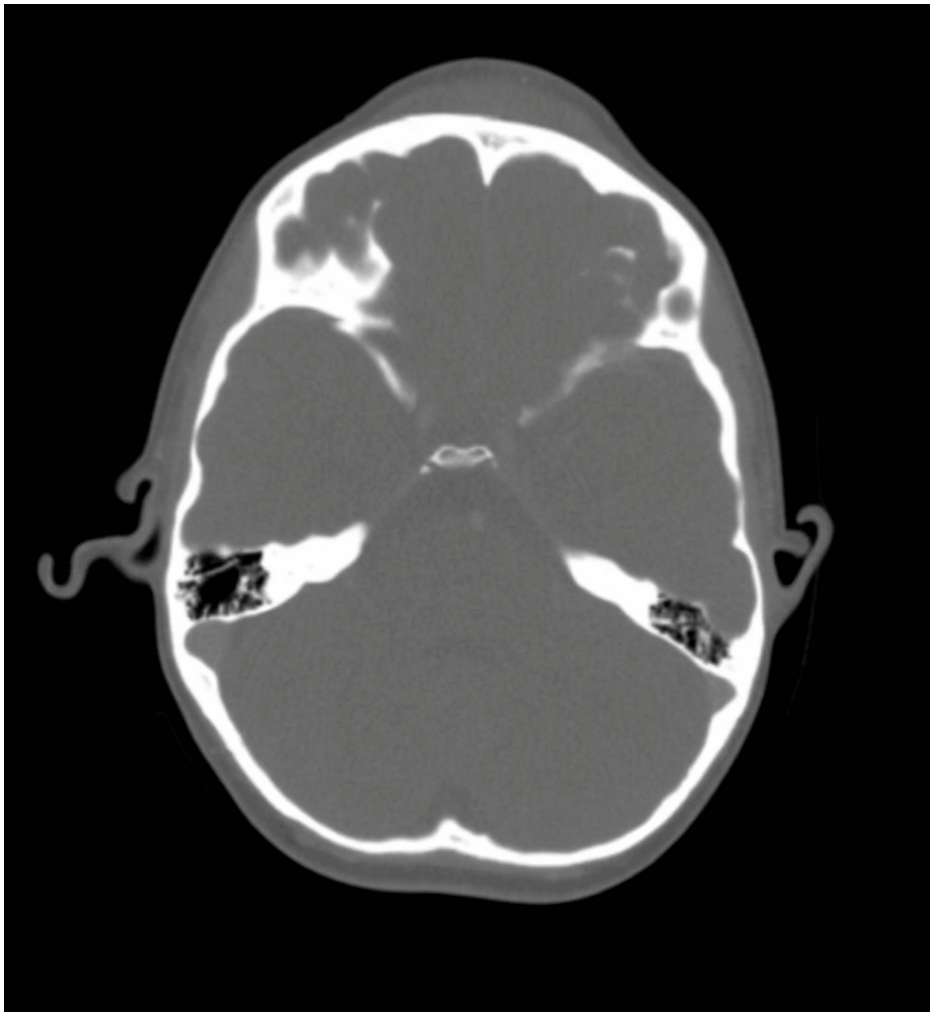


FIGURE 1: Coronal contrast-enhanced CT demonstrating extensive opacification of the left frontal and ethmoid sinuses with associated frontal soft tissue swelling.



FIGURE 2: Sagittal contrast-enhanced CT demonstrating left-sided paranasal sinus opacification with anterior frontal soft tissue swelling. No intracranial extension.

The patient was admitted under otolaryngology and commenced on intravenous co-amoxiclav, saline irrigation, topical decongestants, and intranasal corticosteroids. He subsequently underwent endoscopic septoplasty and left functional endoscopic sinus surgery. Septoplasty was performed to improve surgical access. The procedure included a left uncinectomy, middle meatal antrostomy, left bulla ethmoidalis removal, and frontal recess exploration, with identification of the left frontal sinus. Intraoperatively, the mucosa was markedly vascular, and mucopus was present within the left maxillary sinus. The affected sinuses were irrigated.

He received four days of intravenous co-amoxiclav followed by a 10-day course of oral antibiotics. His fever resolved, and the forehead swelling substantially reduced following surgery. He was discharged the following day. At outpatient review 10 days later, the swelling had resolved completely. He reported no nasal obstruction, rhinorrhoea, visual symptoms, or facial pain. Flexible nasal endoscopy demonstrated patent nasal passages without ongoing inflammation or mucopurulent discharge.

Discussion

This case illustrates the diagnostic challenge of evaluating frontal swelling in a child with acute sinus disease when CT does not definitively demonstrate the classical features of PPT. Although the combination of fever, frontal swelling, and frontal sinus disease raised clinical suspicion, the absence of definite frontal bone osteomyelitis or a confirmed subperiosteal abscess meant that the diagnosis could not be established with certainty. Nevertheless, the patient responded well to antibiotic treatment and endoscopic sinus surgery, with complete clinical resolution.

In PPT, frontal sinus infection can spread beyond the sinus either by direct extension through the sinus walls or via the valveless diploic veins, which communicate with the dural venous system [3,8]. Venous spread has

been proposed as a mechanism of extrasinus dissemination, even in the absence of obvious bony destruction. This was described in a published case of PPT with intracranial extension, where the sinus walls remained intact and venous spread was considered a possible mechanism [8]. However, the mechanism of frontal soft tissue involvement in our patient remains uncertain.

CT is important for assessing sinus disease, bony involvement, and associated collections, while MRI can help identify intracranial or orbital complications [1-3,5,9]. In this case, CT demonstrated frontal and ethmoid sinus disease with overlying soft tissue swelling but no definite osteomyelitis or subperiosteal abscess. Although specialist radiological review raised the possibility of a small collection, the findings remained inconclusive. MRI was not performed, and the diagnosis remained that of suspected PPT rather than a radiologically confirmed case. Endoscopic sinus surgery was undertaken to address the underlying sinonasal infection. Septoplasty was required for access, and the procedure included frontal recess exploration and irrigation of the affected sinuses. The patient subsequently achieved complete clinical resolution.

OI is associated with abnormalities of type I collagen that affect bone structure, strength, and microarchitecture [6,7]. A literature review has also described otological manifestations, particularly hearing impairment, in individuals with OI [10]. The altered bone architecture associated with OI raises the possibility that it could be relevant to the development or spread of sinonasal infection. In particular, changes in cortical thickness and bone porosity provide a theoretical basis for considering whether the local response to infection might differ in patients with OI [7]. However, no established association between OI and PPT has been reported, and the significance of their coexistence in this patient remains uncertain.

A limitation of this case is that the diagnosis of PPT could not be definitively confirmed, as imaging did not demonstrate frontal bone osteomyelitis or a definite subperiosteal abscess. The case therefore demonstrates the clinical and radiological challenges of suspected PPT but cannot establish whether OI influenced the presentation or progression of the infection.

Conclusions

Frontal swelling in a child presenting with acute rhinosinusitis warrants careful assessment for potentially serious extrasinus complications, including PPT. This case highlights the importance of recognizing suspected PPT even when classical radiological features are not definitively demonstrated and of integrating clinical findings, imaging, and multidisciplinary specialist assessment when establishing a working diagnosis. Early antibiotic treatment and appropriate surgical management may achieve favourable outcomes, as demonstrated by the complete clinical resolution in our patient. The coexistence of genetically confirmed osteogenesis imperfecta raises an interesting question about whether abnormalities in collagen biology and bone microarchitecture may influence susceptibility to or progression of sinonasal infection. However, this remains a theoretical possibility, and this case cannot establish a causal relationship. Further clinical reports may help determine whether this represents a relevant association or an incidental finding.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Rebecca Gan Foon Yong

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Drafting of the manuscript: Rebecca Gan Foon Yong

Critical review of the manuscript for important intellectual content: Rebecca Gan Foon Yong, Vishnu S. Sunkaraneni

Supervision: Vishnu S. Sunkaraneni

Disclosures

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