INTRODUCTION

This case is of a 34-year-old immunocompromised male who presented with Pott's puffy tumour as a complication of frontal sinusitis. Surgical incision and drainage was performed with endoscopic debridement of sinuses, in addition to treatment with antibiotics and nasal decongestants. This case highlights immunosuppression, particularly with tumour necrosis factor (TNF) inhibitors, as a potential risk factor for development of Pott's puffy tumour as a complication of frontal sinusitis.

CASE REPORT

A 34-year-old gentleman presented to the emergency department of a regional hospital with 2 days of left-sided periorbital swelling superior to the eye socket with associated erythema and tenderness. He was assessed to have normal visual acuity and extraocular movements. He had a history of severe ankylosing spondylitis for which he was taking a TNF inhibitor, adalimumab. His blood tests showed a slightly raised C-reactive protein of 82 mg/l but otherwise no abnormality. Magnetic resonance imaging (MRI) was performed on the day of admission and noted significant left frontal sinusitis complicated by a defect of the frontal cortical bone as well as oedema and enhancement consistent with a Pott's puffy tumour (Fig. 1). There was also an abscess of the periorbital soft tissue. Immediate treatment was given in the form of phenylephrine nasal decongestants and intravenous antibiotics (amoxicillin with clavulanic acid), and the patient was transferred to a tertiary centre with ear, nose and throat surgery capacity.

Within 24 hours of arrival at the tertiary centre, an incision and drainage of the soft tissue abscess and periorbital abscess was performed. At the same time the sinuses were debrided...
via endoscopic approach and a wide maxillary antrostomy and sphenoidectomy was performed. It was discovered that the frontal recess was completely occluded by a bony wall (Fig. 2). A cannula was placed into the frontal sinus for regular irrigation during the post-operative period with the other sinuses irrigated via nasal douche. Intravenous antibiotics and regular irrigation were continued for a further 5 days, and the TNF inhibitor was withheld on the advice of a rheumatologist. After this time the frontal sinus cannula was removed and the patient was discharged home. He was reviewed as an outpatient 6 days later and had a palpable soft tissue collection in the same area, which was promptly drained surgically and appeared to be a mucocele intraoperatively. None of the cultures identified a specific causative organism, although a Gram stain from the original admission showed gram negative bacilli. The patient was planned to have definitive surgery to open the bony occlusion of the frontal sinus and allow proper drainage. The patient was noted not to have any recurrence of symptoms on follow-up 1 year post-operatively.

DISCUSSION

Pott's puffy tumour is a rare clinical entity and is most commonly a complication of frontal sinusitis in children [1]. This case is unique in that it presents in an adult with no traumatic history and immunosuppression as the only risk factor. TNF inhibitors do not appear to have a significant association with sinusitis but due to being immunosuppressants may slightly increase the risk of upper respiratory tract infections [2]. A literature review was conducted to investigate the role of immunosuppression in Pott's puffy tumour patients.

A search was conducted on 10 March 2020 of CINAHL, Cochrane Embase, Informit, Google Scholar (first 50 results only), Medline and Scopus of ‘Pott’s Puffy’ and variations of this phrase. Initial 804 records were reviewed by title, abstract and full text (as detailed in Fig. 3) and narrowed down to 13 papers of adult Pott’s puffy tumour where immunosuppression was noted in the case.

Of the unique papers identified, 155 only included paediatric patients, whereas 128 included adult patients. This would indicate that Pott's puffy tumour in adult patients is not quite as rare as previously thought. The paediatric cases were reviewed by abstract only for immunosuppression and only four were found. Two girls also on the same immunosuppressant medication as this patient (TNF inhibitor adalimumab) developed Pott's puffy tumour [3]. Another case of a 14 year old on an immunosuppressive study drug for juvenile idiopathic arthritis was published in the USA [4]. Finally one child with Type 2 diabetes mellitus in a case series was diagnosed with Pott's puffy tumour [1].
Table I. Summary of all cases Pott’s puffy tumour cases found with immunosuppression of patients

| Author/et al. | Age | Sex | Presenting complaint | Immunocompromised | Intracranial spread | Pathogen | Surgical management | Complications |
|---------------|-----|-----|----------------------|-------------------|--------------------|----------|--------------------|---------------|
| Adams et al.  | 55  | M   | Pain, swelling, watery discharge | Diabetes mellitus | Left meningeal abscess | Pseudomonas aeruginosa | Bilateral fronto-basal catheter sinusotomy, bilateral complete ethmoidectomy, bilateral middle mental antrostomy, right sphenoidectomy | Chronic ethmoid sinusitis and left orbital abscess requiring surgery |
| Akiyama et al.| 21–83 | M+F | Diabetes, chronic renal failure, aplastic anaemia, breast cancer with bone metastasis | — | — | — | External (58.1%) or endoscopic (32.9%) frontal sinus surgery, frontal bone debridement, craniotomy, simple percutaneous drainage (9.7%) | 2 cases of postoperative recurrence |
| Chow and Szeto | 60  | F   | Headache, swelling, fever, paresthesias | End stage renal failure | — | Mycobacterium tuberculosis | — | — |
| Donville-Lewis et al. | 21 | F   | Headache, swelling, chemosis | Pregnancy | Right epidural empyema posterior to frontal sinus | Streptococcus milleri | Percutaneous drainage, unilateral craniotomy, middle mental antrostomy, anterior ethmoidectomy, frontal sinus access dissection | Caesarean section |
| Effat et al.  | 62  | F   | Headache, swelling, fever, purulent, discharge | Insulin-dependent diabetes, chronic renal failure on twice weekly hemodialysis | Erosion of lamina papyracea of left anterior and middle ethmoid cells and subperiosteal abscess | Moraxella | Cruciate incision and drainage, daily debridement | Fistula |
| Eriyazigu et al. | 27 | M   | Pain headache, swelling, fever | Type 1 diabetes mellitus | Subperiosteal abscess in vertex region associated with dural thickening and irregularities of epidural space | — | Booronal skin incision under local, drainage of subperiosteal abscess, curette | — |
| Gil-Carcedo et al. | 48 | M   | Left hemiparesis, neck stiffness, right mydriasis, confusion, seizure | Chronic alcoholism | Epidural abscess, subdural empyema | Unclassified aerobic streptococcus | Right frontal-pterional craniotomy, intracranial antrostomy and bilateral transantral ethmoidectomy, reconstruction with acrylic grafts | — |
| Goldfarb et al. | 58 | M   | Pain, swelling | Heavy smoking, diabetes mellitus | — | — | Incision and drainage | Fistula between frontal sinus and forehead skin |
| Husta and Reichner | 43 | M   | Altered mental status, proptosis | HIV, cocaine abuse | Subdural empyema, intracranial abscesses resulting in midline shift | Streptococcus mitis | Drainage of the right frontal subperiosteal abscess, left temporal craniotomy and evacuation of subdural empyema | Hypoxic respiratory failure requiring intubation, multiple bilateral cavitary lung abscesses in bases |
| Lamoreau and Fanciullo | 55 | M   | Left eye abscess, fever | Alcoholic cirrhosis, hepatitis C, nicotine, marijuana/other drug use | Left frontal bony erosion | Viridans like streptococci | Incision and drainage | Increased petit mal symptoms of known epilepsy |
| Miller | 60 | M   | Headache, swelling | Diabetes mellitus, prednisolone (weaning dose for allergy) | Erosion of anterior frontal sinus with anterior extension to meninges | Streptococcus anginosus | Spontaneously ruptured in hospital | — |
| Pain et al.  | 81  | M   | Rainless swelling, weight loss, profound asthenia | Diabetes mellitus | Erosion of left frontal bone and orbital roof | Streptococcus constellatus | Endoscopic sinus surgery | — |
| Sekine et al. | 56 | M   | Headache, recurrent frontal swelling, eyelid erythema | Rheumatoid arthritis, treatment (prednisolone, ivermectin, bupivacaine) | — | Streptococcus milleri | Biocoronal incision and endoscopic approach for drainage. Anterolateral thigh flap reconstruction. Revision of forehead cavity with titanium plate later | — |
Of the 13 adult patients’ papers comprising of both case reports and case series, 12 patients were immunosuppressed and 1 was pregnant (Table 1). The most commonly noted was diabetes in seven patients, and there was one patient with rheumatoid arthritis being treated with oral prednisolone, iguratimod and bucillamine.

Most Pott’s puffy tumour cases appear to be in previously fit and well patients, so immunosuppression may play less of a role than neglect of symptoms and delayed clinical review. In this patient the location being several hours drive from the nearest hospital may be more relevant than their immunosuppression. Nonetheless, a higher index of suspicion for severe complications of sinusitis should still be considered for immunosuppressive patients.

ETHICS APPROVAL
LNR/QTHS/67480.

CONFLICT OF INTEREST STATEMENT
None declared.

REFERENCES
1. Bambakidis NC, Cohen AR. Intracranial complications of frontal sinusitis in children: Pott’s puffy tumor revisited. Pediatr Neurosurg 2001;35:82–9.
2. MIMS Online. MIMS Online: Etanercept Full Product Information. Sydney: MIMS Australia; 2020. Available from: https://www.mimsonline.com.au.
3. Christensen AE, Toftedal P. Sinogenic subdural empyema in two girls treated with adalimumab for chronic recurrent multifocal osteomyelitis (CRMO). Pediatr Rheumatol 2017; 15:28–9.
4. Chandwani V. Pott’s puffy tumour & epidural empyemas as complications of sinusitis in a teenager with jia. J Investig Med 2018;66:480.
5. Adams J, Foster D, Stephenson L, Bourguillon P. Pott puffy tumor. Consultant 2017;57:251–3.
6. Akiyama K, Karaki M, Mori N. Evaluation of adult Pott’s puffy tumor: our five cases and 27 literature cases. Laryngoscope 2012;122:2382–8.
7. Chow KM, Szeto CC. Headache caused by Pott’s puffy tumor. Headache 2003;43:916.
8. Domville-Lewis C, Friedland PL, Santa Maria PL. Pott’s puffy tumour and intracranial complications of frontal sinusitis in pregnancy. J Laryngol Otol 2013;127:S35–S8.
9. Effat KG, Karam M, El-Kabani A. Pott’s puffy tumour caused by mucormycosis. J Laryngol Otol 2005;119:643–5.
10. Evliyaoglu C, Bademci G, Yucel E, Keskil S. Pott’s puffy tumor of the vertex years after trauma in a diabetic patient: case report. Neurocirugia 2005;16:54–7.
11. Gil-Carcedo LM, Izquierdo JM, Gonzalez M. Intracranial complications of frontal sinusitis. A report of two cases. J Laryngol Otol 1984;98:941–5.
12. Goldfarb A, Hocwald E, Gross M, Eliashar R. Frontal sinus cutaneous fistula: a complication of Pott's puffy tumor. Otolaryngol Head Neck Surg 2004;130:490–1.

13. Husta B, Reichner C. Pott's puffy tumor: a case with intracranial and pulmonary manifestation. Chest 2012;142:167A.

14. Lamoreau KP, Fanciullo LM. Pott's puffy tumour mimicking preseptal cellulitis. Clin Exp Optom 2008;91:400–2.

15. Miller B. A 60-year-old man with forehead swelling. Cleve Clin J Med 2016;83:95–6.

16. Pasin F, Bonardi S, Modoni A. When the diagnosis is written on the forehead. Eur J Internal Med 2018;47:e1–2.

17. Sekine R, Omura K, Ishida K, Tanaka Y. Recurrent Pott's puffy tumor treated with anterior skull base resection with reconstruction of the anterolateral thigh flap. J Craniofac Surg 2019;30:e94–e6.