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Clinical Record

An unusual presentation of inverted papilloma: case report and literature review

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Abstract

Objective: Inverted papilloma is a rare but locally aggressive tumour with the potential for malignant transformation. Intracranial extension or complications secondary to inverted papilloma are extremely rare. We report a case of inverted papilloma with a large frontal sinus mucocele eroding the frontal sinus, which presented with sudden neurological compromise. A literature review on intracranial extension of such tumours is also included.

Methods: A Medline search of articles, using the terms 'inverted papilloma', 'Ringertz tumour', 'intracranial extension' and 'complication'. Suitable references from the collected articles were also reviewed. Articles published in English were selected and reviewed.

Results: A total of 10 cases was identified. Intracranial spread was more commonly seen in recurrent cases, especially if the recurrence involved the cribriform plate, fovea ethmoidalis or orbits. Cases with extradural disease seemed to have a better prognosis than those with intradural spread.

Conclusions: Intracranial involvement of inverted papilloma is extremely rare, and is usually seen in recurrent cases. This case report highlights an unusual but serious case of inverted papilloma presenting with acute neurological deterioration secondary to a large frontal sinus mucocele eroding the frontal sinus. A literature review on intracranial extension of inverted papilloma indicated that common sites of intracranial spread include the cribriform plate, fovea ethmoidalis and orbits. The prognosis for patients with such tumours depends on the type of dural involvement, with intradural extension carrying a poorer prognosis.

Key words: Inverted Papilloma; Tumour Metastasis; Paranasal Sinus Neoplasms; Intracranial Neoplasms

Introduction

Inverted papilloma is a benign but locally aggressive tumour of the head and neck. We report a rare case of inverted papilloma with a large secondary frontal sinus mucocele, which presented with sudden neurological deterioration. To our knowledge, this is the first report of inverted papilloma presenting with neurological compromise. A literature review on intracranial extension of inverted papilloma is also included.

Case report

A 32-year-old man presented to the otolaryngology department with a two-year history of left-sided nasal obstruction. He denied suffering headache, rhinorrhoea, anosmia, diplopia or facial pain.

Nasal examination, including endoscopy, revealed unilateral nasal polyps occluding the left nasal cavity. The rest of the ENT examination was normal.

The patient was scheduled to undergo an examination of the nose and intranasal polypectomy under general anaesthesia.

However, two weeks later the patient was admitted as an emergency with multiple epileptic seizures resulting in acute neurological deterioration. He required intubation and ventilation and was admitted to the intensive care unit.

A computed tomography (CT) head scan revealed a large frontal sinus mucocele (Figure 1) eroding the posterior wall of the frontal sinus and extending into the anterior cranial fossa on the left side. Compression of the lateral ventricles was also noted, with displacement of midline structures to the right.

The patient was extubated two days later. At this stage, he was noted to have expressive dysphasia but was otherwise neurologically stable, with no further seizures. Computed tomography scanning of the paranasal sinuses (Figure 2) showed extensive polyps occluding the nasal cavity and the maxillary, ethmoid and frontal sinuses on the left side.

The patient underwent emergency endoscopic sinus surgery for removal of polypoidal tissue for histological diagnosis, and also for attempted endoscopic drainage of the frontal mucocele. Intra-operatively, polyps were removed from the lateral nasal wall and the anterior and posterior ethmoidal cells. The polyps were grossly abnormal in appearance, with areas of caseous material and hard bony elements. The frontal recess was examined; there were no signs of mucopus. Drainage of the frontal mucocele was not possible endoscopically.

Subsequently, a left frontotemporal craniotomy was performed using a posteriorly based pterional incision extending across the midline. Following burr hole placement, frank pus was drained and the multiloculated mucocele was seen filling the left frontal region, with several large pieces of ectopic bone. The lesion was completely excised, and a pericranial flap was fashioned to cover the frontal sinus defect.

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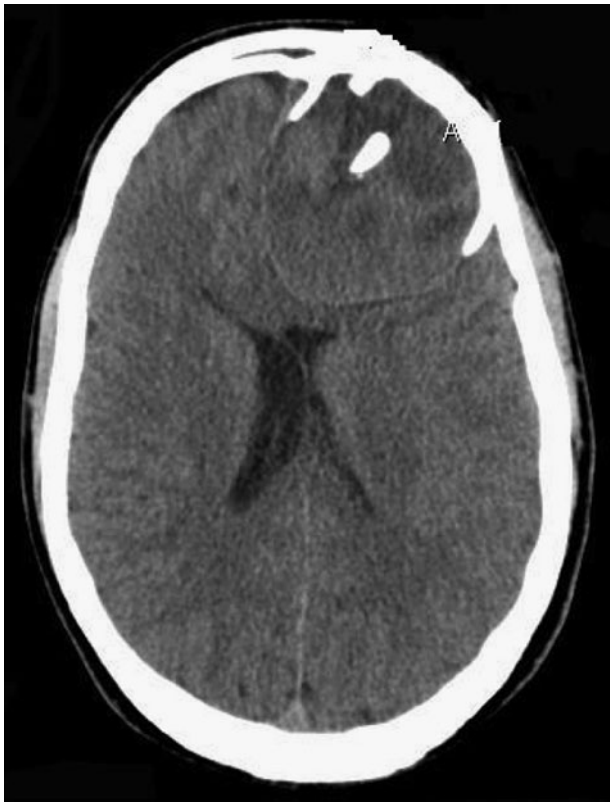

FIG. 1
Axial computed tomography scan showing mucocele in the anterior cranial fossa, extending across the midline.

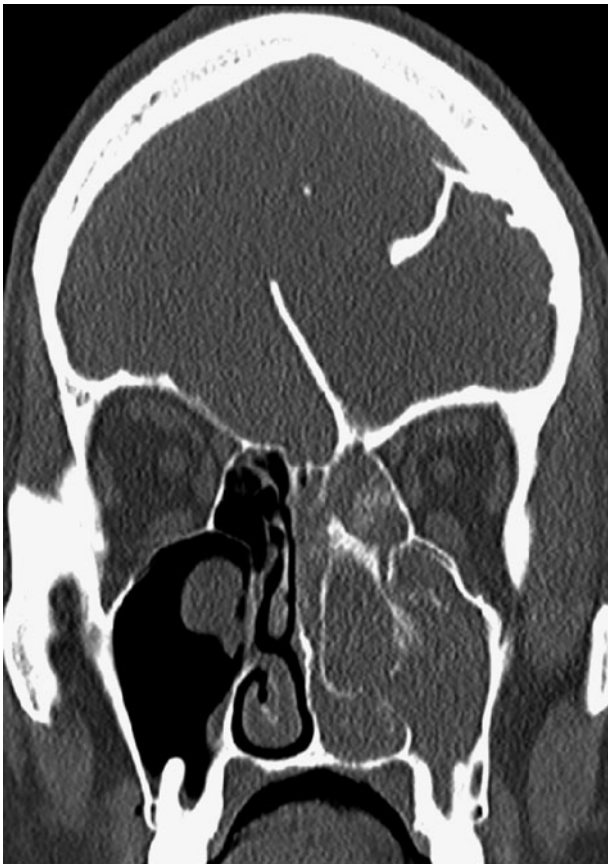

FIG. 2
Coronal computed tomography scan showing complete opacification of the ethmoid, maxillary and frontal sinuses on the left side.

Histologically, the nasal polyps comprised metaplastic squamous epithelium with an inverted growth pattern and areas of low-grade dysplasia; the reported diagnosis was inverted papilloma.

The patient was treated with appropriate antibiotics (ceftazidime, flucloxacillin and metronidazole) and recovered well after surgery, with no neurological sequelae.

Discussion

Inverted papilloma is a benign epithelial tumour which accounts for 0.5 to 4 per cent of all primary nasal tumours.[1,2] It is a locally aggressive lesion with a high risk of recurrence and the potential for malignant transformation into squamous cell carcinoma (occurring in 10 per cent of cases).[3] The vast majority of recurrences reportedly occur at the margins of previous surgical resections.[4] The aetiology of this tumour remains unclear; however, allergy, human papilloma virus, chronic rhinosinusitis, environmental carcinogens and tobacco smoking have been implicated.[5,6] Common sites comprise the lateral nasal wall and ethmoid and maxillary sinuses.[7–9]

Lawson *et al.* studied a series of 100 patients and reported that the most common presenting symptom of inverted papilloma was unilateral nasal obstruction, followed by rhinorrhoea and epistaxis.[4] To our knowledge, the current case represents the first report of an inverted papilloma presenting with sudden neurological deterioration, and the second report of a secondary frontal sinus mucocele.[10] Inverted papilloma usually presents between the fifth and sixth decade of life. However, our patient was younger (32 years), suggesting that inverted papilloma may have a more aggressive nature in younger patients.

In our patient, we believe that obstruction of the frontonasal duct resulted in a large frontal sinus mucocele. Rapidly expanding mucoceles may cause bony erosion and mimic a malignant process. In such circumstances, a CT scan is useful in differentiating the two. In the case of a mucocele, there is usually a fat plane between the tumour margin and the surrounding tissue planes.[11] Erosion of the posterior wall by a frontal sinus mucocele can result in complications such as subdural empyema, meningitis and brain abscess.

Reports on the management of frontal sinus mucocele with intracranial extension are divided into intranasal endoscopic approaches and combined intranasal–transcranial approaches with transcranial excision.[12] In experienced hands, an intranasal endoscopic approach is less invasive and avoids the need for a craniotomy; however, some authors prefer a combined approach to ensure complete removal of the lesion and to reduce the risk of further recurrence.[13]

Table I summarises previous reports of cases of inverted papilloma with intracranial disease and/or complications (accessed via a Medline search, using the search terms 'inverted papilloma', 'Ringertz tumour', 'intracranial extension' and 'complications', excluding non-English language papers). Suitable references from the collected articles were also reviewed. Inverted papilloma with intracranial extension is usually seen in recurrent cases, and this is especially true if the lesion affects the cribriform plate, fovea ethmoidalis or orbits. Intracranial recurrence also depends on intradural spread of disease and the completeness of surgical resection.

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TABLE I

PREVIOUS REPORTS OF INVERTED PAPILLOMA WITH INTRACRANIAL INVOLVEMENT

Study	Pt age (yrs)	Dural involvement	Extension site	Treatment
Guedea *et al.*[16]	41	Intradural	CP, SP + FS	Local excision + RT
Vural *et al.*[17]	51	Intradural	CP	CFR
Peterson & Heim[18]	91	Intradural	FS	None
Van Olphen *et al.*[14]	32	Extradural	CP + FS	CFR
Lewis *et al.*[19]	52	Extradural	Orbit	CFR
Vrabec[20]	67	Extradural	FE	LRMM
Miller *et al.*[15]	42	Extradural	FS	CFR
Myers *et al.*[9]	66	Extradural	Orbits + FS	Local excision
Dolgin *et al.*[3]	23	Extradural	CP + ACF	CFR
Lyons *et al.*[10]	33	Extradural	FS	CFR

Pt = patient; yrs = years; CP = cribriform plate; SP = sphenoid sinus; FS = frontal sinus; FE = fovea ethmoidalis; ACF = anterior cranial fossa; RT = radiotherapy; CFR = craniofacial resection; LRMM = lateral rhinotomy with medial maxillectomy

The 'gold standard' treatment of inverted papilloma is complete surgical excision, as this reduces the risk of recurrence and enables thorough histological examination to detect any coexisting malignant transformation. Local recurrence rates of up to 78 per cent have been reported for techniques such as non-endoscopic transnasal and transantral excision, whereas rates of 0 to 29 per cent have been cited for procedures such as medial maxillectomy with lateral rhinotomy and facial degloving.[17] For lesions affecting the anterior skull base, craniofacial resection may be required.

- **Intracranial involvement of nasal inverted papilloma is rare and is usually associated with recurrent disease**
- **This report highlights an unusual but serious case of inverted papilloma presenting with acute neurological deterioration, secondary to a large frontal sinus mucocele eroding the frontal sinus**
- **The prognosis of patients with these tumours depends on the type of dural involvement, with intradural extension carrying a poorer prognosis**
- **Complete surgical excision remains the treatment of choice, with adjuvant radiotherapy reserved for selected cases**

The prognosis for patients with inverted papilloma with intracranial involvement depends largely on dural spread, with intradural extension of disease having a poorer outcome compared with extradural spread.[14] In the studies reviewed, three out of four cases (75 per cent) with intradural disease extension died an average of nine months post-operatively.[16–19] In contrast, the six patients with extradural disease had an average of four years' disease-free survival.[3,9,10,14,15,20]

Endoscopic resection of inverted papilloma is becoming increasingly popular and offers several advantages, such as adequate illumination, lack of a facial incision, reduced scarring, and decreased facial swelling, nasal crusting and bleeding.[21] A recent meta-analysis of 32 studies comparing endoscopic versus non-endoscopic, contemporary techniques for inverted papilloma excision concluded that endoscopically resected lesions had a lower recurrence rate compared with non-endoscopic resections (15 *vs* 20 per cent, respectively).[22] However, even though endoscopic resection offers these advantages, some authors state that it should be reserved for selected cases.[23]

Surgery remains the mainstay of treatment for inverted papilloma; however, radiotherapy should be considered for cases associated with malignancy, incompletely excised lesions, locally aggressive tumour and multiple recurrences.[24]

Long-term follow up of these patients is imperative as recurrence can occur even after several years.[25] Clinical examination should focus on nasal endoscopy and biopsy of suspicious areas.

Conclusion

Intracranial involvement of inverted papilloma is extremely rare and is usually associated with recurrent disease. This report highlights an unusual but serious case of inverted papilloma presenting with acute neurological deterioration secondary to a large frontal sinus mucocele eroding the frontal sinus. Common sites of intracranial spread of inverted papilloma include the cribriform plate, fovea ethmoidalis and orbits. The prognosis of patients with these tumours depends on the type of dural involvement, with intradural extension carrying a poorer prognosis. Complete surgical excision remains the treatment of choice, with adjuvant radiotherapy being reserved for selected cases. Inverted papillomas are known to recur, and long-term follow up is therefore essential.

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