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Original Article

Bilateral inverted papilloma: A report of two cases and review of the current literature

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Abstract Inverted papilloma is locally aggressive tumour which typically presents as a unilateral nasal polyp. Whilst it has a malignant potential it rarely transforms and in its benign form the main concern originates from its locally aggressive nature and substantial propensity to recur. Bilateral disease can also be due to inverted papilloma, sometimes due to direct extension of the tumour, but it can also occur as two distinct lesions. Here we report two cases of bilateral involvement, review the current literature and highlight some important issues on the management and follow-up of this well known neoplasm.

Keywords Inverted papilloma · Paranasal sinus neoplasm · Bilateral

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Introduction

Inverted papilloma is a benign sinonasal tumor which is locally aggressive and has a well recognized malignant potential with a transformation rate often quoted in the literature in the region of 2.1–7% [1–3]. It has been documented across all ages and constitutes 0.5–4% of all sinonasal tumors [4]. The lateral nasal wall and middle meatus are the most common sites for inverting papillomata to arise but they can develop from any part of the Schneiderian membrane in the nose and paranasal sinuses. They can extend to involve all of the ipsilateral sinuses and cases of intracranial extension and dural involvement, whilst rare, have been well documented [5].

An inverting papilloma usually presents as a unilateral nasal polyp and therefore bilateral involvement of the nose and paranasal sinuses is a rare entity. A recent series published from the UK had a reported rate of just under 7% for bilateral involvement [1] whilst other series report rates typically between 2% and 4% [6, 7]. In this article we describe two cases of bilateral inverted papilloma which presented to our department, discuss the possible treatment options available and highlight some important management issues.

Case reports

Patient 1

A 43-year-old female patient presented to the department of ORL-HNS at Bradford Teaching Hospitals with a 1 year history of right sided nasal obstruction, purulent nasal discharge and chronic headache. She was otherwise fit and well with no significant past medical history and examination revealed bilateral intranasal polyposis with complete obstruction of the right nostril.

A CT scan showed obliteration of the right nasal cavity by polypoidal mucosal thickening and opacification of the right paranasal sinuses (Fig. 1). No abnormality was detected

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Fig. 1 Coronal CT scans showing opacification of the right paranasal sinuses caused by an inverted papilloma

on the left side. The patient underwent examination under anesthesia (EUA) which showed polypoidal tissue from both sides of the middle turbinate filling the right nostril. Biopsy confirmed this to be transitional cell papilloma.

The polyp recurred on the right side within a month with obvious left sided middle turbinate papillomatosis. An endoscopic right sided medial maxillectomy with septal stripping was performed. The left middle turbinate was excised and histology confirmed bilateral inverting papilloma. The patient had a further EUA and excision of obvious recurrence on the nasal septum 4 months later, which was widely excised leaving behind a septal perforation. A further interim endoscopic examination and biopsies showed no recurrence. Fourteen months from the right medial maxillectomy a further recurrence was noted on the floor and lateral wall of the right nasal cavity. This was widely excised once again utilizing an endoscopic approach. To date, 2 years following the last endoscopic procedure, the patient remains free of any recurrent inverting papilloma.

Patient 2

A 75-year-old male patient presented with unilateral nasal obstruction and an obvious polyp in his right nasal cavity. He subsequently underwent a right sided polypectomy and was found to have inverted papilloma on the resected specimen. He was only followed up for a 6 months period and was discharged by a trainee without an endoscopic assessment of his nasal cavity being peformed. One year later he represented with a 6 months history of epiphora and right periorbital swelling. On examination with a zero degree rigid endoscope he was found to have a polypoidal lesion obstructing the right nasal cavity. He also had mild proptosis. A CT scan of the sinuses was requested and showed an extensive lesion which had infiltrated the orbit and invaded the floor of the frontal sinus (Fig. 2). A right external frontal ethmoidectomy and hemimaxillectomy was therefore performed and the histology was confirmed to be inverted papilloma with no evidence of malignancy.

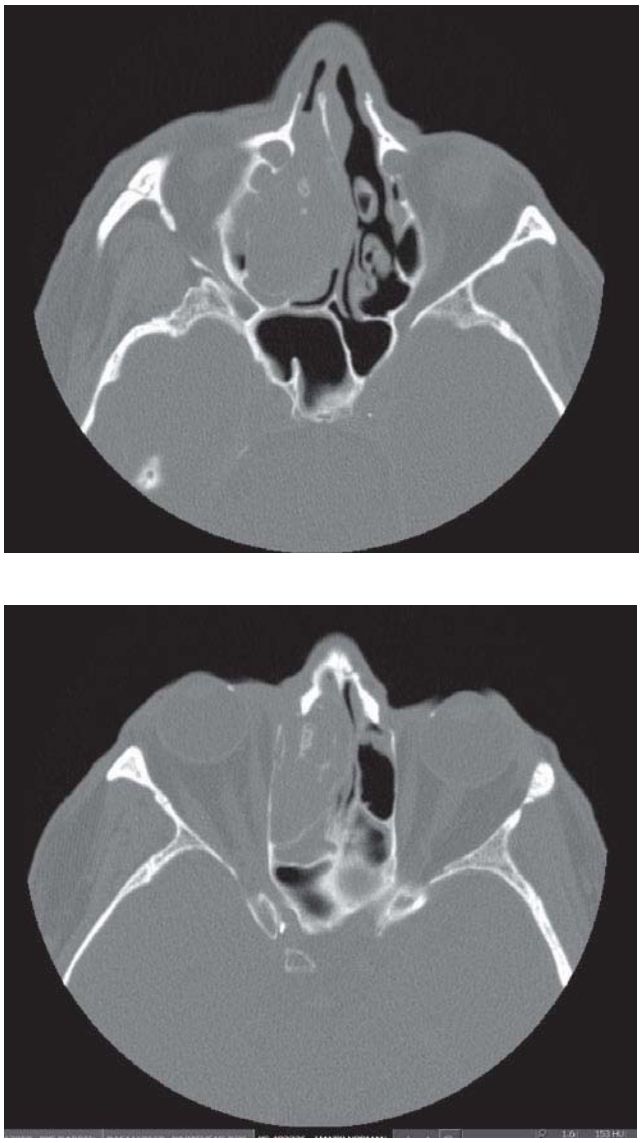

Fig. 2 Axial CT scans showing an inverted papilloma filling the right nasal cavity and eroding the medial wall of the orbit

He was followed up in the outpatient clinic with regular endoscopic examinations for a period of 2 years. It was then noted that he had polypoidal disease forming in both nasal cavities around the region of the frontal recess. A CT scan confirmed this and a combined open-endoscopic procedure was performed to clear the polypoidal tissue. The histology confirmed inverting papilloma arising from the right nasal cavity and the left frontal recess. The patient has now been under review for 3 years from his last surgical procedure and remains disease-free.

Discussion

Inverted papilloma is a rare benign sinonasal tumor, characterized by a potentially invasive nature and relatively high recurrence rates which tend to vary with the extent of primary lesion and the choice of surgery [8, 9]. Both the cases presented here highlight this high recurrence rate. In the second case the patient was discharged too early without a proper endoscopic examination of his nasal cavity. This highlights both the high recurrence rate and the need for regular endoscopic examination for a considerable length of time. In our unit we would follow these patients for a period of at least 5 years.

The term inverted papilloma reflects its characteristic histological appearance of a covering epithelium that invaginates or inverts into the underlying stroma. It is more common in men and usually presents in the fifth and sixth decades of life [10]. Unilateral disease and unilateral nasal obstruction is the most common presenting feature [11, 12] but, as demonstrated in this article, bilateral disease does occur with reported rates of 2–7% [1, 6, 7]. The etiology of this multicentric appearance has yet to be fully understood. It has been suggested that bilateral and multicentric presentations could be the result of extension of the lesion through metaplasia of the adjacent mucosa [13]. However, as in our report, others have found bilateral involvement with no breach of the nasal septum and on opposite lateral nasal walls [14, 15]. In these cases bilateral involvement cannot be explained by metaplasia of adjacent mucosa.

Imaging plays an important role in the preoperative assessment of inverting papilloma as it allows for accurate planning of the intended surgical procedure. CT scanning is the initial modality of choice to evaluate the extent of disease [16]. Bony erosion can be present with inverted papillomas in as much as 44% [6] but raises concern over potential malignant change. More recently magnetic resonance imaging has been used to aid in the assessment and guide surgery as it can accurately distinguish between retained secretions and the soft tissue of a tumor [6, 16].

Traditional teaching has been to treat inverted papilloma with aggressive surgery in the form of a lateral rhinotomy and medial maxillectomy in an effort to limit recurrence [11, 12, 17, 18]. However, over the last decade many reports have emerged using endoscopic techniques [9, 19–22] with rates of recurrence averaging out at 13.4%. A trend towards utilizing the improved endoscopic technology, including navigational systems, has slowly emerged [23]. This approach avoids the problems of facial scarring and complications such as wound infection which has been reported in as many as 10% of lateral rhinotomy incisions [24].

Endoscopic surgery alone is not suitable for all cases. In one series of 104 cases when used in isolation the rate of recurrence was 22.4% which dropped to 16.2% when combined with open procedures for more extensive disease [19]. It has been proposed that endoscopic surgery alone is suitable for those lesions confined to the lateral nasal wall with or without extension into the ethmoid, maxillary and sphenoid sinuses [6] while tumors extending into the nasofrontal duct, orbit and frontal sinus may be better off with a combined procedure [6] as utilized in our second case as the tumor extended laterally in the frontal sinus. Outcome studies have suggested that endoscopic and combined endoscopic/external approaches are at least equal in their effectiveness as more traditional techniques but are associated with a reduced hospital stay and decreased morbidity [25]. If malignant progression is detected then there should be a very low threshold for open surgery. As with any procedure the outcome will also vary with the skill of the surgeon and we would recommend that definitive surgery for these tumors be carried out be experienced endoscopic sinus surgeons in a recognized center.

Recurrence rates have been quoted in the literature anywhere from as low 2% [11] to as high as 80% [3]. Obviously theses rates of recurrence depend very much on the extent of the original disease, the completeness of the resection, the tumor location and the biological variability of the tumor [6]. It follows that tumors with bilateral presentation will be more likely to recur as they are already multicentric and there is an increased chance of leaving residual tumor behind at surgery. This increased propensity to recur is well demonstrated in both cases illustrated in this report. Other features which have been associated with an increased risk of recurrence are smoking and younger age at presentation [26].

Malignant transformation and concurrent malignant disease is well recognized in this benign sinonasal tumor with rates in the literature ranging from 6 to 14.5% [2, 10, 27]. The factors which are responsible for this malignant transformation are as yet not understood. A number of studies have been performed in an attempt to discover predictive characteristics for malignant change [28, 29]. Eggers et al. [28] studied 93 cases of inverted papilloma with regards to both patient and tumor characteristics. They concluded that certain histological features, namely increased mitotic count and dyscariosis, along with male gender and older age group put patients at a higher risk of malignant change. Ingle et al. [29] studied 76 cases of inverted papilloma and found that a loss of the cell surface marker CD44 correlated with squamous cell carcinoma within the inverting papilloma but this did not have a predictive value. When malignant change has occurred radiotherapy has a role to play in the control of the disease [17, 30].

The treatment of inverting papilloma is essentially surgical. The surgical modality and access must be carefully considered and tailored according to the patient's condition. This case report illustrates the two major surgical approaches, complete endoscopic resection in the first case as opposed to case 2 where both external

and endoscopic resections were necessary. This difference in approach to the management was carefully considered because of the nature and anatomical location of the disease. It also highlights the importance of long-term follow-up with regular endoscopic examination in the outpatient setting.

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