Bilateral aggressive inverted papillomas: A changing scenario

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Abstract

Objectives The ideal management of bilateral, recurrent inverted papillomas is intriguing and often controversial. We describe the ideal management of such tumors.

Design Retrospective, case series

Methods The three patients in this series inspite of several surgical procedures in the past had recurrences and behaved in an aggressive fashion. The patients underwent surgical excision by either bilateral lateral rhinotomy, midfacial degloving or craniofacial approaches.

Results One patient who underwent excision by midfacial degloving approach had recurrence. This was removed by lateral rhinotomy approach. Two patients were subjected to radiotherapy due to the presence of mitotic figures on histopathology. All the patients are asymptomatic at 6 months follow-up.

Conclusion Aggressive inverted papillomas may have varied presentation. They could have multiple recurrences, bilateral presentation or intracranial extension. They can be effectively managed with bilateral lateral rhinotomy or

cranio-facial resection. Adjuvant radiotherapy has a role in controlling recurrences.

Keywords Bilateral papilloma · Intracranial extension · Bilateral lateral rhinotomy

Introduction

Inverted papilloma was classified as a true nasal neoplasm (“true papilloma”) in 1935 by Kramer and Som [1], distinguishing it from inflammatory nasal polyps. Later, Ringertz et al [2] described the propensity of this lesion for locally invasive growth. There is diversity in nomenclature of this tumor reflected as a part of misunderstanding of this tumor’s behaviour. In spite of its rarity, the interest has been sparked by its tendency to recur frequently and association with squamous cell carcinoma [3, 4]. This is further intensified by its bilateral presentation, since the inverted papilloma are usually unilateral [5]. Some cases of inverting papilloma may also behave more aggressively resulting in intracranial extension. Because of clinical resemblance between the papillomatosis and inflammatory polypoid, the correct diagnosis is usually possible on histopathological examination.

Recognition of the predilection for recurrence and association with malignancy have led to evolution of guidelines for the treatment of inverting papilloma. Surgical excision is generally regarded as the treatment of choice, but the approach and extent of surgery have been the subject of debate. Many workers have endorsed lateral rhinotomy and wide resection as the procedure of choice for all cases of inverted papilloma [6, 7]. Of late, there has been a growing trend towards endoscopic removal of these tumours. However, the endoscopic technique may be grossly ineffective, notably in extensive cases or if bilateral extension is present. Endoscopic surgery by less experienced surgeon may also contribute to recurrences.

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We report three cases of aggressive inverted papilloma (two bilateral) who inspite of several surgical procedures in the past had many recurrences and behaved in an “aggressive” fashion. The management of these cases along with review of literature (Table 1) is discussed.

Case Report 1

A 55 years old man was referred to our hospital with complaints of bilateral nasal obstruction and epistaxis. He had been operated seven times between 1989 and 2003. He had undergone three times non endoscopic endonasal polypectomy, twice excision by lateral rhinotomy approach and twice endoscopic excision. Patient presented with recurrence of the tumor within 5 months of the last surgical procedure. Clinical examination revealed pinkish polypoidal mass lesion occupying both the nasal cavities. A computerized tomographic (CT) scan showed tumor involving bilateral nasal cavities, bilateral maxillary, ethmoidal and sphenoidal sinuses (Fig. 1). The tumor was eroding the cribriform plate with intracranial extension. There was also destruction of the posterior table of frontal sinuses on both the sides with intracranial extension. A complete tumor excision was achieved by a bilateral lateral rhinotomy approach. The whole tumor was excised along with bilateral medial maxillectomy (Fig. 2). The dura was healthy. He was subjected

to radiotherapy in view of atypia and brisk mitotic activities on histopathology (Fig. 3) and multiple recurrences. The patient is tumor free for 6 months after the procedure, without any adverse sequelae of surgery.

Case report 2

A 37-year-old man presented with bilateral nasal obstruction and nasal bleeding for the last 2 ½ years. Clinical examination revealed pinkish polypoidal mass lesion occupying both the nasal cavities. Contrast enhancing CT scan of paranasal sinuses showed involvement of both maxillary, ethmoidal and sphenoidal sinuses along with destruction of medial wall of maxilla. A biopsy from the lesion revealed histopathological features of inverted papilloma. All other investigations including a skiagram of the chest were normal. The tumor was exposed by a mid facial degloving approach and excised completely by bilateral medial maxillectomy. Following surgery, patient remained asymptomatic for about 10 months. Thereafter, he had recurrence of the lesion involving the right side of paranasal sinuses. He was operated again by a lateral rhinotomy approach. Postoperatively, the patient has been doing well at 3 months follow-up. Excised tumor tissue did not exhibit any evidence of malignancy on histopathology examination.

Fig. 1 Preoperative CT scan of bilateral inverted papilloma showing intracranial extension at cribriform plate and posterior table of frontal sinus.

Fig. 2 Postoperative CT scan of bilateral inverted papilloma showing complete clearance of disease from intracranial part.

Fig. 3 Microphotography with low power magnification of inverted papilloma showing brisk mitotic activity reaching superficial layers (arrows) (H&Ex10).

Insert- high power magnification showing mitosis (arrows) (H&Ex20).

Fig.4 Preoperative CT scan of inverted papilloma involving left frontal sinus, orbit and extending intracranially.

Insert - Postoperative CT scan after craniofacial resection

Table 1 Bilateral inverted papilloma, its management and outcome

S. No.	Year	Author (et al) (ref. no)	Extend of disease (bilateral)	Surgical approach	Associated malignancy	No. of Recurrence	Radiotherapy	Follow-up-free of diseases
1.	1981	Perzin [21]	NC	Transnasal excision	SCC	–	RT given	Died after 3 months (leukemia)
2.	1982	Chatterji [5]	NC, ethmoid, maxillary	B/L transantral ethoidectomy	–	–	–	1 year
3.	1990	Phillips [3]	NC	Lateral rhinotomy + medial maxillectomy	–	–	–	–
4.	1991	Nielsen [4]	NC, (L) maxillary sinus	Not mentioned	SCC	Not mentioned	Not mentioned	Not mentioned
5.	1994	McCary [22]	NC, ethmoids, orbit	Lateral rhinotomy+ supraorbital craniotomy	–	–	–	49 months
6.	1996	Shohet [23]	NC, Frontal sinus, ® maxilla	Osteoplastic surgery	–	–	–	2 years
7.	1996	Hosal [15]	NC, ethmoids, maxilla	B/L lateral rhinotomy + medial maxillectomy	–	–	–	6 months
8.	1997	Tsue [24]	Aplastic maxillary sinus	Endoscopic excision	–	–	–	Not mentioned
9.	1998	Sham [9]	Ethmoids	Endoscopic excision	–	–	–	58 months
10.	2002	Yiotakis [25]	NC, ethmoids, sphenoid	Endoscopic excision	–	–	–	1.5 years
11.	2003	Baruah [26]	NC, ethmoids, maxillary, sphenoids, ITF	Endoscopic excision	–	1	–	5 years
12.	2003	Baruah [26]	NC, ethmoids, maxillary, sphenoids, ITF	Endoscopic excision	–	3	–	1 year
13.	2005	Panda NK (index Case 1)	NC, maxillary, ethmoids, sphenoids	B/L lateral rhinotomy	–	7	RT given	6 months
14.	2005	Panda NK (Index Case 2)	NC, maxillary, ethmoids, sphenoids	Midfacial degoving	–	1	–	6 months

Case Report 3

A 53 years old female was referred to us with complaints of bilateral nasal obstruction and epistaxis. In the past, she had been operated three times elsewhere. She underwent excision of the tumor with an extended lateral rhinotomy approach. The patient presented again within 3 months of the surgical procedure with complaints of headache, ipsilateral proptosis and epistaxis. The CT scan showed an extensive lesion involving the ethmoids, frontal sinuses and extending into orbit and intracranially to the anterior cranial fossa (Fig. 4). A craniofacial resection of this tumor was accomplished by a team of neurosurgeons and otolaryngologists. The patient remained asymptomatic for 1 year before presenting again with mild epistaxis. She had a recurrence of lesion in the ipsilateral ethmoids which was removed again. In view of the recurrent, aggressive nature and presence of mitosis on histopathology, she has been subjected for radiation therapy.

Discussion

The local aggressiveness, high rate of recurrence, associated malignancy and tendency to multicentricity are the clinical characteristics of inverted papilloma that have recently led most workers [7, 8] to advocate radical surgical removal of these tumors. The incidence of recurrence has been observed directly related to the method of surgical treatment. The limited excision by intranasal polypectomy, turbinectomy or the Caldwell-luc has resulted in high recurrence rates of 73% [6] to 78% [7]. On the contrary, lateral rhinotomy performed in conjunction with medial maxillectomy is accompanied by a marked reduction in the incidence of recurrences, the figures varying from 0 – 13% [6–8].

Over the past century, management of inverted papilloma has remained a controversial subject. The various surgical approaches in vogue are: external approaches (lateral rhinotomy or midfacial degloving with medial maxillectomy) and endoscopic endonasal excision [9].

It is obvious that surgical access and excision is better with open approaches and hence less chances of recurrence. Most surgeons would therefore adopt the radical external approaches as the first line treatment. (Cummings and Goodman [6]; Phillips et al [3]) although others have recommended the endonasal or the limited external approaches for limited disease (Woodson et al [10]; Lawson et al [11]). Currently the lateral rhinotomy approach is regarded by most surgeons as the gold standard against which all surgical procedures for sinonasal inverted papilloma must be measured. But none of the authors have described an ideal approaches for extensive bilateral lesion involving paranasal sinuses with intracranial extension. There is no reference standard for such extensive lesions.

Selection of therapy depends on an accurate radiologic assessment of the extent of tumor. Lawson et al [11] advocated an intranasal or transantral ethmoidectomy for lesions of lateral nasal wall involving the inferior or middle turbinate and or meatus, with limited extension into the ethmoid labyrinth and antrum. Candidates for the conservative intranasal surgery ideally should not have had any previous surgery. On the contrary, some reports appear to support radical surgery consisting of lateral rhinotomy with medial maxillectomy [12, 13]. The recurrence rates reported ranges from 0–14% with this method [13]. The technique of the lateral rhinotomy with medial maxillectomy is suitable for the resection of lesions that arise from the lateral nasal wall and extend to the ipsilateral ethmoid and maxillary sinuses [13]. Lateral rhinotomy with septotomy is the modification of this approach that includes the transaction of the nasal septum. Despite the improved exposure to the both nasal cavities and ethmoids, this does not provide enblock resection of bilateral lesions [14]. Biller et al [14] described the superior rhinotomy technique for resection of bilateral ethmoid neoplasms that extend into the anterior cranial fossa. Although this approach provides bilateral enbloc resection of ethmoid labyrinth, medial wall of maxilla and cribriform plate, it should be considered only for patients with intracranial extension.

Bilateral lateral rhinotomy approach can be ideal for extensive bilateral lesions [15]. It gives an excellent exposure for adequate removal of the tumor. It can even be employed for limited intracranial extensions through the frontal sinus (Case 1). Midfacial degloving has been developed by the association of sublabial and rhinoplastic incisions, with or without osteotomies of nasal bone and frontal process of maxilla [16]. In our study, Case 2 underwent excision by midfacial degloving approach. In spite of this, he presented with recurrence. We feel that this was due to the lesion situated high up in the ethmoids. Mid facial degloving approach may thus have a limitation in lesions situated high up, such lesions can be removed with lateral rhinotomy approach.

To circumvent the complications of external approaches, endoscopic intranasal procedures were proposed. Waitz and Wigand [17] concluded that endoscopic elimination

of inverted papillomas can be accomplished even with large lesions involving the posterior ethmoidal sinus, the naso frontal duct, or sphenoidal sinus, with recurrence rate of 17%, but results were biased by selecting patients with smaller and less aggressive lesions. More and more workers [17, 18], are now proposing endoscopic approach for inverted papillomas. Endoscopic approach, however, may have certain limitations. It has a limited role in bilateral aggressive papillomas as was the cases in the present series. All the three patients in the present series had endoscopic excision during some period of their management. We therefore are of the opinion that endoscopic surgery may not be an answer for aggressive bilateral lesions.

Kamel et al [18] believed that the most important factor in the choice of the surgical procedure is the involvement of maxillary sinus, and divided into two distinct groups according to the location. The lesions originating from the lateral nasal wall or nasal septum, are ectodermal in origin, have a localised attachment and in most cases are pedicled, can be controlled by conservative endoscopic endonasal excision. The second group includes lesions having diffuse attachment located lateral to the lateral nasal wall, i.e., involving maxillary sinus with or without nasal extension, which could be controlled by radical transnasal endoscopic medial maxillectomy. But what constitute a safety margin in endoscopic surgical resection is a contentious issue. However, tumor involving the anteromedial aspect of the maxillary sinus presents technical difficulty, particularly when the nasolacrimal duct is in the way. Another limitation for the endoscopic approach is more extensive tumor involvements of the frontal sinus, supra-orbital ethmoid region and intracranial extension, which pose an access problem as in our Case 3. It is important to note that even the lateral rhinotomy or midfacial degloving approach alone may not be able to offer sufficient access to these areas. These type of lesions should be ideally dealt by craniofacial resection (Case 3). There should be no hesitation in resorting to external approaches if there is any doubt about achieving complete resection which should always be the primary goal of management.

Concern exists that the removal of medial maxilla on both the sides may result in nasal deformity by reducing the lateral nasal support. However, the nose supported by its midline skeleton is not at risk of collapse. Another area of concern is, if bilateral incisions would compromise the blood supply of the nasal skin. As the external nasal branch of anterior ethmoid artery supplies the skin along the dorsum to the tip [19], it is unlikely that postoperative necrosis would occur. Additionally, zigzag incision along the dorsum between the medial canthus and nasal ala, provides better cosmetic results with less scar formation [19]. The role of adjuvant radiation therapy for a benign lesion is poorly defined [20, 21]. However, the association of malignancy or histopathological evidence of focal dysplasia, atypia and increase in mitotic figures, which happened in

two of our cases necessitates adjuvant radiation therapy to control further recurrences [21].

To conclude, aggressive inverted papillomas may have varied presentations. They may have extensive bilateral disease or multiple recurrences with intracranial extension. They can be effectively managed with bilateral lateral rhinotomy or a craniofacial resection for intracranial extension. Adjuvant radiotherapy has a role in controlling recurrences in patients exhibiting dysplasia on histopathology.

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