

International Journal of Medical Science and Current Research (IJMSCR) Available online at: www.ijmscr.com Volume1, Issue 2, Page No: 89-95 July-August 2018 IJMSCR

Bilateral Recurrent Inverted Papilloma a Surgical Challenge: Case Report and Review of Literature

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Type of Publication: Original Research Paper Conflicts of Interest: Nil

ABSTRACT

Introduction: Inverted papilloma is a locally aggressive benign tumor with a rare bilateral nasal involvement. Although benign, it is characterized by high rate of recurrence, associated risk of malignancy and tendency of multicentricity which led to intriguing and controversial management.

Case summary: We present a case report of a male patient who underwent multiple nasal surgeries by endoscopic route for bilateral nasal inverted papilloma and presented with recurrence of the disease. Patient was further managed with an external approach where bilateral lateral rhinotomy with medial maxillectomy and complete excision of the disease was done to prevent recurrence.

Discussion: The ideal surgical management of bilateral, recurrent inverted papilloma is controversial due to its high rate of recurrence and tendency of multicentricity. They can be effectively managed by various surgical approaches with importance of identification of the site of origin and appropriate preoperative planning. We report one such case of bilateral inverted papilloma with several surgical procedures in the past and had recurrences. We also review the current literature and highlight the issues on the management.

Keywords: Bilateral nasal inverted papilloma, recurrent inverted papilloma, Inverted papilloma, bilateral recurrent inverted papilloma.

INTRODUCTION

Inverted papilloma is a rare benign tumor of the nose and paranasal sinuses. It originates from the scheniderian nasal mucosa and constitutes .5-4% of all sinonasal tumors.^[1,2,3] It is seen in all the age groups with 4 to 5 times more common in males with more prevalence in Caucasians in their 5th and 6th decade and has an incidence of .75 to 1.5/ 100cases.⁴

Although benign it is a tumor of surgical challenge because of its aggressive growth potential, local invasive property, multicentricity (12%), high rate of recurrence In 10% cases of inverted papilloma has cellular atypia with squamous cell carcinoma.^[4,6]Inverted papilloma is usually confined to one side of nasal cavity in 91-99% cases⁷ and bilateral involvement is extremely rare and to best of our knowledge there are only 30 reported cases in the English literature [Table 1].

Treatment of choice in inverted papilloma is surgery with importance of preoperative radiological localization of primary site of attachment to reduce the incidence of recurrence. Although the type of surgical approach is controversial in literature because of the characteristic features of the tumor and for bilateral lesions combined approaches are required to provide adequate exposure. In this article we describe a rare such case of bilateral sinonasal inverted papilloma with recurrence and importance of

combined approach in successful removal and review of literature.

CASE REPORT

A 41 year old male presented to our department of ENT with the complaints of bilateral progressive nasal obstruction left side more than right, headache and nasal discharge for 6 years. He underwent two nasal surgeries in the past at another institution during the past 9 years. Anterior rhinoscopy examination revealed a fleshy pink polypoidal mass filling bilateral nasal cavity. A computed tomography scan showed a soft tissue mass in the left side of nasal cavity involving the ethmoid region and frontal sinus. Bilateral sphenoid sinuses were free of the disease. Primary site of attachment could not be seen.

Intraoperatively a fleshy polypoidal mass was arising from the middle meatus area on both the sides of nasal cavity. The mass was seen extending posteriorly to involve the ethmoidal area on the left side. Absent middle turbinate with syenechiae was seen between the right turbinate and septum on the right side due to previous surgery shown in figure 1 and 2. Patient underwent bilateral lateral rhinotomy with wide exposure and complete excision of the entire residual tumor to prevent the recurrence. The postoperative period was uneventful and patient had unremarkable recovery. Histopathology revealed the mass to be inverted papilloma. The follow up of the patient in the last six months showed no recurrence.

DISCUSSION:

Inverted papilloma term reflects its characteristic histological appearance of epithelium that invaginates or inverts into the underlying stroma. It is called Schneider, papilloma, ewing papilloma, transitional cells papilloma, epithelial papilloma virus cancer, transitional cell benign tumor and papillomatosis.^[1,2,6]This diversity in nomenclature reflects a part of misunderstanding of the tumor behavior.

It is a rare tumor and constitutes only .5-4% of all sinonasal tumor.^[1,2] Inspite its rarity, the tendency of frequent recurrence, association with malignancy and local aggressive nature make this a surgical challenge. The discussion is further intensified by its bilateral presentation, seen only in 4% cases.⁸It is suggested that bilateral and multicentric involvement can occur due to extension of lesion through

metaplasia of adjacent mucosa.⁸ However, Astor et al.⁹ reported independent bilateral inverted papilloma. Similarly Perzin et al.¹⁰reported a case of bilateral inverted papilloma with squamous cell carcinoma with no communication seen between the two.in our case also there was no invasion through the septum and on opposite lateral walls.

It mainly arises from the nasal cavity, lateral wall and paranasal sinuses, with primary involvement of paranasal sinuses seen only in 5% cases.^[6,11] Till date to best of our knowledge only 30 cases have been reported in medical literature. Of these cases only one is iatrogenic in nature and two cases have primary sphenoid of frontal involvement and sinus respectively. Surgical excision is generally regarded as the treatment of choice. Preoperative radiological investigation is important for planning an appropriate surgical approach. Precision of surgery and postoperative rate of recurrence is reduced by localizing the primary site of attachment of the tumor and need to be drilled during the surgical excision of the tumor It is characterized by a cone shaped area on contrast enhanced scan due to hyperostosis of bone and has a preoperative predictive value of 95%.³⁵ Although in our case we could not identify this site of attachment due to previous multiple surgery.

The approach and extent of surgery is the debatable subject. Main goals of any surgical approach are adequate exposure for complete resection of the tumor. unobstructed postoperative view for surveillance and minimal cosmetic deformities and functional disabilities. The accepted choice is complete wide local excision. Several external surgical procedures like lateral rhinotomy, medial maxillectomy, and the midfacial degloving approach has been proposed for extended IPs in an effort to limit recurrence which ranges from 0- 11% with these radical surgeries.^[6,12,13] It is 41%- 78% with conservative procedures like nasal polypectomy, cadwell luc surgery.²

In the last decade endoscopic sinus surgery has emerged with the recurrence rate of 13.4%.¹⁴ However; it is not suitable for all the cases. It is suitable for lesions confined to lateral nasal wall with or without extension into paranasal sinuses, while tumors extending to the nasofrontal duct, orbit and frontal sinus may be done with a combined external and endoscopic procedure.

Dr. Payal Kamble et al. International Journal of Medical Science and Current Research (IJMSCR)

Of the reported cases of bilateral nasal inverted papilloma one was excised with bilateral endoscopic medial maxillectomy approach and another one using the combined open and endoscopy further using diode laser for excision. In our patient two independent lesions were seen arising from the middle meatus and extending posteriorly to the ethmoid and sphenoid sinus on the left side. With the recurrence of the tumor a bilateral lateral rhinotomy approach was planned combined with endoscopic visualization and it provided a wide exposure, good accesses and wide working space to nasal cavity, ethmoid and maxillary sinus with complete excision of the residual tumor done. Powered instruments like microdebrider was used for assisted and complete clearance of the diseases Post-operative period was uneventful and follow up of the patient in 1 year months post-surgery doesn't show any recurrence.

The surgical decision should take into account the extent, volume and lesion location. Sunet al.³⁴ suggested that location of IP, secondary surgery, AJCC stage of IP/SCC and the choice of surgical method for advanced IP, incomplete or inadequate removal of tumor are the direct risk factors of IP recurrence. The management of risk factors involves precise identification of the tumor attachment site, anatomical anomalies in sinonasal region; careful planning of surgical procedures and well- planned postoperative follow up.

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TABLES AND FIGURES

Table 1: Review of cases of bilateral nasal cavity inverted papilloma

$(NC\mbox{-}nasal\ cavity, lt\mbox{-}left, rt\mbox{-}right, B/L\mbox{-}bilateral, ITF\mbox{-}infratemporal\ fossa)$

No	Year	Author (et al)	Extension of disease	Surgical approach
1	1981	Perzin	NC	transnasal excision
2	1982	Chatterji	NC,ethmoid,maxillary	B/L transnasalethmoidectomy
3	1990	Phillips	NC	Lateral rhinotomy+medialmaxillectomy
4	1991	Nielsen	NC, lt maxillary sinus	Not mentioned
5	1994	Mccary	NC,ethmoids,orbit	Lateral rhinotomy+supraorbital craniotomy
6	1996	Shohet	NC,frontal,rt maxillary	Osteoplastic surgery
7	1996	Hosal	NC, ethmoids, maxillary sinus	B/L lateral rhinotomy+medialmaxillectomy
8	1997	Tsue	aplastic maxillary sinus	Endoscopic excision
9	1998	Sham	ethmoids	Endoscopic excision
10	2002	Yiotakis	NC,ethmoids,sphenoid	Endoscopic excision
11	2003(2)	Baruah	 NC,ethmoids,maxillary ,sphenoids NC,etmoids,maxillary, sphenoids,ITF 	1.Endoscopic excision
				2.Endoscopic excision
12	2005(2)	Panda nk	 Nc,maxilla,ethmoid,sp henoid NC,maxillary,ethmoids ,sphenoids 	 B/L lateral rhinotomy midfacialdegloving
13	2007	Panda nk	Ethmoids,frontal,orbit,in tracranial	Craniofacial resection
14	2008	Salmone	NC,etmoids,.frontal,sphe noid,rt maxillary sinus	Endoscopic excision

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15	2010	Murphy	NC	Open+endoscopic procedure
16	2011	Cabrezio	NC, maxillary ethmoid	Endoscopic excision
17	2012	Kodama	NC, maxillary, ethmoid ,sphenoi	Endoscopic medial maxillectomy
18	2013	Agarwal	NC, frontal, ethmoid rt maxillary	Lateral rhinotomy=endoscopic exposure=diode laser
19	2013	Pathak	NC, lt maxillary, lt sphenoid B/L ethmoid	Endoscopic excision
20	2014	Keskin	Frontal sinus	Endoscopic excision
21	2015	Subramaniam	NC, B/L maxillary ,ethmoid ,frontal	Endoscopic excision
22	2017	Oztruk	NC, maxillary	Endoscopic medial maxillectomy

Figure 1: Endoscopic view of nasal cavity



A) Right side



B) Left side

Figure 2: CECT scan of Nose and PNS



A) Coronal view



Figure 3: Intra operative view



A) Right side

B) Left side

C) Coblator in use



A) 1st post operative day



B) 1 year follow up





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