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Skull Base Inverted Papilloma: A Case Report

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Abstract

Inverted papilloma is an uncommon primary nasal. Despite its benign nature, this tumor represents three typical characteristics: a high propensity of recurrence, local aggressiveness and association with malignancy. Inverted papilloma can reduce the patient's quality of life due to compromised nasal function, extension to orbit and brain even in absence of malignancy. We are presenting a case of 66 year old male patient with bilateral inverted papilloma with intracranial extensions after multiple recurrences operated 10 times over a period of 28 years, with extensive resection of disease every time but without any evidence of malignancy till date still showing fast and aggressive recurrences.

Introduction

Inverted papilloma is a benign epithelial growth extending into the underlying stroma of the nasal cavity and paranasal sinus. These tumors arise from the Schneiderian respiratory membrane that lines the nasal cavity and paranasal sinuses [1]. Schneiderian papillomas can be classified in into 3 types: exophytic (everted or fungiform), inverted, and oncocytic (columnar or cylindrical) [2].

In 1854 Ward first reported this type of tumor in nasal cavity [3]. Ringertz et al. [4] in 1938 was the first to identify endophytic growth patterns of IPs with its characteristic tendency to invert into underlying stroma and called it "inverted papilloma". Hyams [2] divided papillomas of sinonasal tract into three histological categories due to their pattern of growth: (a) Fungi form (everted) papilloma, (b) Oncocytic schneiderian papillomas, (c) Inverted papillomas.

Inverted papilloma (IP) is an uncommon lesion that accounts for 0.5–4% of all primary nasal tumors [5]. It affects all ages, most commonly males [6] in the fifth to the seventh decades of life [7]. The most frequent sites are the lateral nasal wall near the middle turbinate or ethmoid recesses and the maxillary sinuses. The nasal septum, frontal and sphenoid sinuses are rarely affected. Common sites of intracranial spread include the cribriform plate, fovea ethmoidalis, and orbits [8]. The tumor is well known for its invasiveness, tendency to recur and association with malignancy [9].

Case Presentation

A 66 year old Hindu male patient presented to department of ENT of SMS hospital with few months history of bilateral nasal fullness, purulent nasal discharge, posterior nasal drip, proptosis of left eye and headache. No history of tobacco smoking, alcohol intake or allergy was present.

Over past 28 years patient was operated 10 times for bilateral nasal mass and different approaches were used at different centers by experienced surgeons. Patient underwent Transnasal resection of tumor twice (year1989, 1991), Moore's lateral rhinotomy excision of tumor (year1993, 1995), Midfacial degloving approach (year1999), Endoscopic excision of tumor (year2001, 2003), Bilateral lateral rhinotomy with craniofacial resection (year2005), Endoscopic excision with frontal sinustomy by external approach (year2009).

Histopathological diagnosis of the first operation was nonspecific inflammation of the nasal mucosa, while the pathological diagnosis of rest of the surgeries was Inverted Papilloma. Patient was also advised radiotherapy but he refused for it.

Clinical examination of the patient revealed left eye proptosis with diplopia of left eye, left side frontal bossing, and normal vision. There was no cranial nerve palsy no cervical lymphadenopathy. On performing Anterior Rhinoscopy we found single nasal cavity with absence of nasal septum. Minimal lesion was present along lateral nasal wall. Nasal endoscopy showed single nasal cavity with absence of nasal septum, middle turbinate, inferior turbinate. Papilloma like mass along lateral

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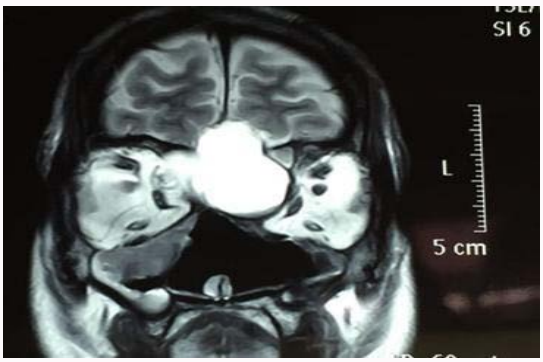

Figure 1: Coronal section MRI of nose and pns showing cystic mass occupying frontoethmoid region with intracranial extension (arrow).

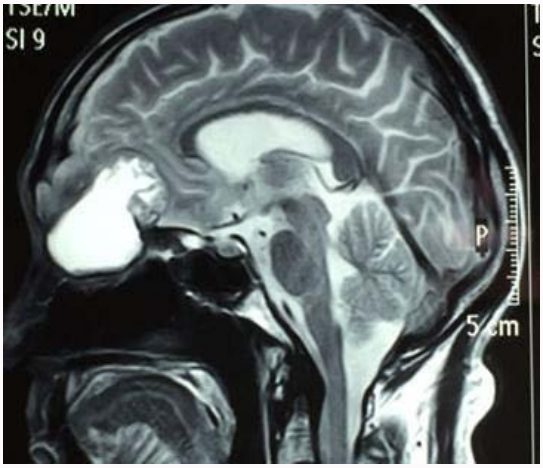

Figure 2: MRI saggital cuts of nose and paranasal sinus showing cystic mass occupying frontoethmoid region with intracranial extension.

nasal wall and a cystic mass seen hanging from roof of nasal cavity.

Since recurrent Inverted Papilloma was suspected patient was elected for CT and MRI of head and neck areas which showed heterogeneous opacification of mass in right maxillary sinus, with minimal lesion in left maxillary sinus and nasal cavity. Nasal septal perforation due to prior endonasal resections was also observed (Figure 3).Cystic mass occupying frontoethmoid region with intracranial extension (Figure 1 and 2).

Under general anesthesia patient underwent endoscopic excision of tumor from lateral nasal wall and maxillary sinus along with aspiration of fluid from the cyst. Histopathological study of the resected specimen as well as cytology of the fluid confirmed the diagnosis of Inverted Papilloma without evidence of malignancy. Further HPV studies of the patient came negative.

Discussion

Inverted papilloma can be defined as a group of benign neoplasm arising from the sinonasal (Schneiderian) mucosa and is composed of squamous or columnar epithelial proliferation with associated mucous cells. Schneiderian papillomas represent <5% of all sinonasal tract tumors. Inverted papillomas occur along the lateral nasal wall (middle turbinate or ethmoid recesses), with secondary extension into the paranasal sinuses. They may originate in paranasal sinus with/ without involving nasal cavity. Typically, the schneiderian papillomas are unilateral; bilateral papillomas may also occur. In our case it

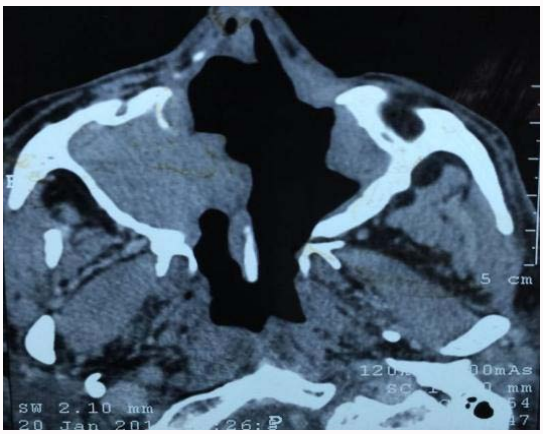

Figure 3: Contrast enhanced CT axial sections showing single nasal cavity with minimal lesion along lateral nasal wall and maxilla.

was bilateral inverted papilloma since beginning. Symptoms vary according to the site of occurrence and include airway obstruction, epistaxis and a symptomatic mass or pain.

The etiology of inverted papilloma is still unclear. There have been many causes suggested such as allergy, chronic sinusitis, viral infections, and inflammation, HPV [10].

Significant association has been identified between the presence of human papilloma virus DNA in inverted papilloma and recurrence after surgical resection [11]. HPV 16 and 18 were found to be related to the malignant transformation of Inverted Papilloma [12]. In addition to HPV, studies have showed that *p21* and *p53*, probably coupled epidermal growth factor receptor (EGFR; ErbB-1), transforming growth factor alpha (TGF-α), Topoisomerase II- alpha are predictive of malignant transformation [13]. The observation that Inverted Papilloma tend to recur after incomplete surgical removal supports investigation suggested that Inverted Papilloma is a true neoplasm arising from single progenitor cell and the recurrence represents growth of the residual clone [14]. In our study we found that the patient was HPV negative and molecular studies could not be done because of unavailability of such studies at our centre but in future if such studies are available then we will get them done.

Differential diagnosis of Inverted Papilloma includes antrochoanal polyp, nasal cavity squamous polyp, allergic fungal sinusitis, fibrous dysplasia, giant cell granuloma, juvenile angiofibroma, nasal glioma, meningoencephalocele, mucocele, mucus retention cyst, Thornwaldt's cyst, grossly enlarged adenoids, SCC, lymphoma, adenocarcinoma, esthesioneuroblastoma. Sinonasal inflammatory polyps are clinically similar but histopathologically epithelial alterations are seen in inverted papillomas and not in the inflammatory polyps. Unilateral tumor localization involving the lateral nasal wall and the middle meatus is a diagnostic clue to Inverted Papilloma.

CT and MRI are techniques of choice for pretreatment staging in IP. Due to nature of tumor histology occurring in paranasal sinuses, CT offers superior bony definitions and MRI gives superior soft tissue delineation [15].Unilateral opacification of the paranasal sinuses is typical CT finding of inverted papilloma. Bony changes on CT imaging of inverted papilloma are useful for predicting tumor origin and recurrence sites. Focal hyperostosis, bony struts or osteitis detected on preoperative CT can predict with high degree of accuracy the site of origin [16]. Erosion, remodeling and widening of the

natural orifices of the sinuses on a CT scan are useful signs indicating IP.

MRI is superior to CT scan in distinguishing papillomas from inflammation. With gadolinium enhancement, MRI demonstrates perineural invasion and dural or intracranial involvement very well. MRI is useful for planning an appropriate surgical approach, and for selecting cases that can be managed by endoscopic approaches, resulting in lower rates of tumorrecurrence and morbidity [17]. Dynamic MR imaging can differentiate accurately recurrent IP from postoperative changes. On T1- weighted images, sinonasal papillomas appear slightly hyperintense to muscle; on T2- weighted images, sinonasal papillomas have intermediate intensity.

Intracranial extension and dural penetration is rare and often associated with recurrent disease [18]. Tumor may spread by direct invasion of bone and cartilage to involve related structures, but the walls of the nasal cavity and paranasal sinuses also contain numerous foraminae and fissures transmitting important neurovascular bundles.

The primary and preferred treatment of inverted papilloma is surgery. Precise determination of sites tumor origin and attachment during the operation, strict application of selection criteria, proper preoperative evaluations, intra-operative determination of extent and attachment of the tumor, close endoscopic follow up, expert application of endoscopic techniques, meticulous use of subperiosteal dissection in the involved areas, wide removal of the tumor origin along the subperiosteal plane as well as drilling the underlying bone [19], complete removal of all the diseased mucosa with creation of wide cavities, and long term regular follow up evaluation are the key elements to the successful treatment. Limited involvement of the skull base can be successfully achieved by endoscopic excision. Endoscopic resection is associated with shorter hospital stay, shorter operative time and lesser morbidity. External approach has been gold standard for sinonasal tumor removal [20], but is associated with several side effects, including facial scars, intracranial and extracranial complications, long hospital stay. The external surgical approach is adequate for tumors extending to the brain, orbit and maxillary sinus.

"Recurrence" actually represents residual disease in most cases. The magnitude of the recurrence is directly proportional to the completeness of removal with the best results obtained by techniques that afford the best operative exposure.

There is also association of malignancy with inverted papilloma. Malignancy was found to be associated with bilateral inverted papilloma, histologic multicentricity, a predominance of mature squamous epithelium, severe hyperkeratosis, absence of inflammatory polyps among the papillomas. In our case it was bilateral inverted papilloma since beginning with presence of focal squamous epithelial cells in HPR with 10 times recurrence over 28 years but still there were no signs of malignancy.

Conclusion

This case report presents Inverted papilloma with intracranial extension after multiple recurrences. Here the Inverted papilloma was bilateral which is less common and a risk factor for malignancy but still no malignancy reported till date. High rate of recurrence, local aggressiveness and association of malignancy makes it necessary to perform radical surgery for this benign tumor. Aggressive treatment of intranasal inverted papilloma is the most important factor in preventing intracranial presentation. Because of the malignant potential of the disease long term follow up of the disease is also mandatory. HPV studies came negative in our case, but due consideration must be given to such studies i.e. HPV and molecular studies so that a better understanding of alterations in epithelial cell proliferation and cell cycle regulation in inverted papilloma may lead to adjuvant medical therapies to decrease recurrence rates and improve treatment.

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