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Case Report

Tricky nasal mass from meek polyp to intricate papilloma: A diagnostic dilemma

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ABSTRACT

Inverted papilloma is a benign epithelial growth arising from the underlying stroma of the nasal cavity and paranasal sinuses. The pathogenesis of this lesion upto date remains unclear. The tumor is known for its local invasiveness, rapid recurrence and link with malignancy. The recurrence rate of this tumor is usually too high that represents residual disease in most of the cases. Hence, it is mandate that the patient keeps a proper follow up on long term concerns. Here is one such patient, whose nasal mass seemed Antrochoanal polyp clinically but ended up as Inverted papilloma histopathologically.

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1. Introduction

Inverted papilloma (IP) is a rare benign neoplasm of the nasal cavity and paranasal sinuses. This tumor mainly occurs in male adults between 40-70 years of age. Three prominent characteristics make this tumor very different from other sinonasal tumors which are relatively strong potential for local invasion, high rates of recurrence and risk of carcinomatous changes. They are associated with malignancy 5-15% malignancy rates.¹⁻⁵ Here, is one such presentation in an adult male with a nasal mass which was both fiddly and deceptive.

2. Case Report

44 year adult male patient comes to ENT outpatient setting with nasal obstruction since 2 years. He also gives history of headache, anosmia, rhinorrhoea which was usually mucoid in nature, thick in consistency and also occasionally blood-stained as well as foul smelling, hemifacial pain on the left side. He also gives history suggestive of allergy such as watery nasal discharge, sneezing, hawking sensation in the

throat, pruritus in the nose and eyes. He also gives history of aural fullness. He had shown at several places which suggested only local consultation where he was temporarily treated thinking it to be recurrent URI. He is known hypertensive since 4 years for which he is on medication and is in control. No previous surgical history or any other systemic illness. On clinical examination, patient was stable and comfortable with vitals in normal limits. There were no abnormal findings detected on systemic examination.

On local examination of the nose, the external appearance and osteo-cartilagenous framework of nose was normal. There was no paranasal sinus tenderness. The columella, caudal septum and vestibule were normal. On Anterior rhinoscopic examination as depicted in Figure 1 A & B shows a single, pale, polypoidal sinonasal mass arising from the left middle meatus extending into mid-half of the left nasal cavity anteriorly while also reaching upto the choanae posteriorly blocking the entire posterior choanae. This mass was insensitive to pain and did not bleed on touch. The mass was free from the floor on probe test as was able to negotiate the mass all around except the lateral part from where the mass was seen arising (i.e 270° rotatory movements was only possible). The remaining

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structures that were visualised in the left nasal cavity such as left inferior turbinate, left inferior meatus and partly seen left middle turbinate were normal. There was minimal deviated nasal septum to left with bony septal spur. The right inferior turbinate, right inferior meatus were normal and visualised while right middle turbinate and right middle meatus was not visualised with the floor of the right nasal cavity being free and no polyps visualised. The Posterior rhinoscopic examination showed superior surface of soft palate, posterior end of nasal septum, choanae, posterior end of turbinates and meati, Eustachian tube opening, Fossa of Rosemuller, roof of nasopharynx were normal and seen on right side. But on left side, the mass had was totally blocking the choanae hampering the visualisation of these structures. To check for nasal patency, both cotton wool test and cold spatula test showed reduced movement and reduced misting on both sides, but more on left side. While the tests for olfaction (Pocket smell test) that is the 3 "C" which are Camphor, Coffee powder and Clove oil, the smell was reduced on both sides, more on left side of the nasal cavity.

The coronal section of CT PNS (plain) shows polypoidal mucosal thickening in bilateral maxillary, frontal, ethmoidal and sphenoid sinuses. Soft tissue also seen in B/L nasal cavity in the middle meatus suggestive of Sinonasal polyposis. There is deviation of nasal septum towards left with bony septal spur. No evidence of any bony erosion or destruction as shown in Figure 2C & D

Patient was worked up and fitness for surgery. Patient underwent Functional Endoscopic Sinus Surgery (FESS) under General Anaesthesia. Polyp was excised in toto from the left nasal cavity and sent for histo-pathological examination (HPE). Both sides, Anterior and Posterior Ethmoidectomy along with (R) Middle meatal antrostomy was done that is to clear the disease while retaining the normal sinus mucosa. The histopathology report says papillary structure lined by stratified squamous epithelium. Nuclei are regular with moderate cytoplasm. Sub-epithelium shows edematous stroma with compressed blood vessels. Foci of lymphocytic infiltrate and squamous epithelial islands are noted. No evidence of atypia was seen. Hence, features suggestive of Schneiderian Papilloma (Inverted Papilloma), which appeared as a diagnostic dilemma as shown in Figure 3 E, F & G.

3. Discussion

Inverted papilloma (IP) is a rare benign neoplasm of the nasal cavity and paranasal sinuses. This sinonasal tumor mainly occurs in adults with a male preponderance usually during the 5th decade but can range anywhere between 40-70 years of age with M:F ratio= 3:1. Three prominent characteristics make this tumor very different from other sinonasal tumors which are relatively strong potential for local invasion as well as local destruction

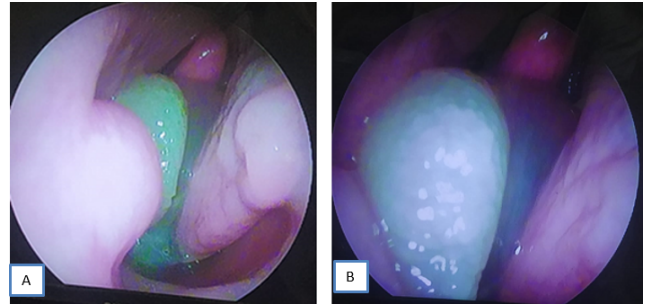


Fig. 1: A:&B: (close-up): Singular, pale, polypoidal sinonasal mass arising from the left middle meatus and occupying the left side of the nasal cavity as per the nasal endoscopic picture seeming like left Antrochoanal polyp.

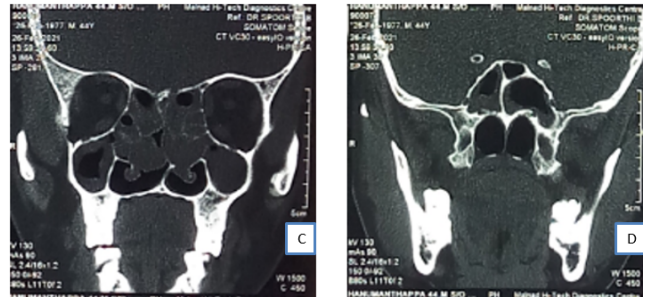


Fig. 2: C:&D: Coronal section of CT PNS (plain) shows polypoidal mucosal thickening in bilateral maxillary, frontal, ethmoidal and sphenoid sinuses. Soft tissue seen in B/L nasal cavity in the middle meatus suggestive of Sinonasal polyposis. There is deviation of nasal septum towards left with bony septal spur. No evidence of any bony erosion or destruction.

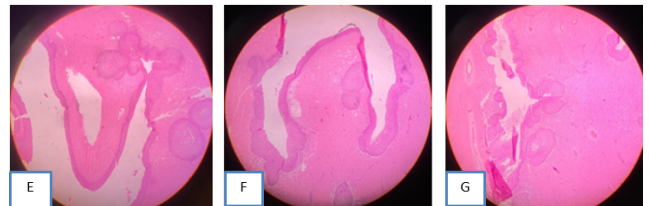


Fig. 3: E: F:&G: All the pictures showing the H & E sections of Schneiderian Papilloma.

of the surrounding structures, high rate of recurrence and risk of carcinomatous evolution. They are associated with malignancy 5-15% malignancy rates are most generally accepted.²⁻⁴ They are most commonly associated with Squamous cell carcinoma (SCC). There are 4 types of association- metachronous SCC, CA in situ within IP, synchronous lesions, malignant transformation. Clinically, unilateral nasal obstruction (58%), epistaxis (17%), nasal discharge (14%), bilateral nasal obstruction (12%), nasal mass (9%), sinusitis (9%), headache, diplopia, facial numbness, facial swelling and anosmia. It originates from Schneiderian membrane which has a neuro-ectodermal

origin. The tumor was first named after Conrad Victor Schneider in 1600 who demonstrated nasal mucosa: that produces catarrh and not CSF. It is also called Schneiderian papilloma, Schneider papilloma, Inverting papilloma, Inverted papilloma, Ringertz tumor, Epithelial papilloma, Transitional cell papilloma, Sinonasal papilloma (SP) (Ward in 1854). It is benign/ intermediate group i.e epithelial origin as per WHO classification and consists about 0.5-4% of sinonasal tumors.³⁻⁶

Its pathogenesis is unclear but although the probable etiology are allergy, chronic sinusitis and viral infections (propensity of HPV 6, 11). The most common site of origin is the lateral nasal wall and the mass is usually unilateral in nature. As per Krouse- the primary sites of this tumor are lateral nasal wall(82%), maxillary sinus(53.9%), ethmoid sinus(31.6%), septum(9.9%), frontal sinus(6.5%), sphenoid sinus(3.9%).⁷⁻⁹ It appears greyish white/pale pink fleshy firm to rubbery sometimes multiple polypoidal mass arising from lateral nasal wall. It has a tendency to recur after surgical removal and is associated with Squamous cell carcinoma in 10-15% cases. Clinically, presents with unilateral nasal obstruction, bleeding from the nose, unilateral nasal mass that resembles polyp.⁸

Clinical-pathological profile of sino-nasal masses (as per study by Lathi et al in Acta Otorhinolaryngol. 2011 Dec; 31(6): 372-377 shows nasal obstruction (97.3%), Rhinorrhoea (99.1%), Hyposmia (31.3%), Headache(16.9%), Intermittent Epistaxis(17.9%) and external deformity of nose and cheek. Krause and Som classified SP's as true nasal neoplasms and described them as true papillomas. Ringertz was the first to identify the tendency of SP's to invert into the underlying connective tissue stroma which differs from other types of papillomas.¹⁰

There are 3 types of gross appearance of this nasal mass.^{11,12} *Fungiform papillomas*(50%):¹¹ arise in the nasal septum and has exophytic growth pattern. It does not grow down into the underlying normal stroma. It is associated with HPV. It doesnot have tendency to recur. In these tumors, > 57% have HPV DNA positivity. Cylindrical papillomas(3%):¹² they have ragged, beefy appearance and histologically composed of columnar cells and pink cytoplasm. The nuclei are oval or round with many areas of mucin cysts. They actually act very similar to inverted papilloma having more chance for malignancy. They are seen to arise from lateral nasal wall/sinuses. Inverted papillomas(47%):^{11,12} The US National Cancer institute has defined inverted papilloma as a type of tumor in which the surface epithelial cells grow downward into the underlying supportive tissue. They arise from Schneiderian membrane which is an invagination of the olfactory ectoderm that occurs during the 4th week of embryonic development. The mucosa creates a transitional zone between the ectodermally derived respiratory epithelium of the nasopharynx and keratinising squamous epithelium with the nasal vestibule. It

is a locally aggressive sinonasal tumor that arises from nasal respiratory epithelium which undergoes metaplastic change and proliferation. They tend to arise usually from the lateral nasal wall.

Exophytic papilloma (tree like) and inverted papilloma (nodule like) are the 2 histological variety. Histologically,⁹ the appearance of epithelium inverting into the stroma with a distinct and intact basement membrane. It separates and defines the epithelial component from the underlying connective tissue. It may be associated with atypia, CA in situ, CA, SCC, transitional cell carcinoma, adenocarcinoma, mucoepidermoid carcinoma, verrucous carcinoma. It is grossly exophytic and polypoidal. It is more vascular than an inflammatory polyp. It is grey to pink. It is frond-like projection and inverts into epithelium and also inverts into underlying stroma. Epithelium is distinct from respiratory (pseudostratified ciliated columnar epithelium) where the mucosa of the sinonasal cavity may be squamous, transitional or respiratory. The epithelium is proliferative and thickened that lacks mucus secreting cells and eosinophils. The staging of this inverted papilloma that aids for surgery is by Krouse staging,¹¹ which is mentioned below as follows:

1. Type I: tumor totally confined to the nasal cavity. The tumor can be localised to 1 wall or region of the nasal cavity or can be bulky and extensive within the nasal cavity but must not extend into the sinuses or into any extranasal compartment. There must be no concurrent malignancy.
2. Type II: tumor involving the OMC, ES and /or medial portion of the MS with or without involvement of the nasal cavity. There must be no concurrent malignancy.
3. Type III: tumor involving the lateral, inferior, superior, anterior or posterior walls of MS, sphenoid sinus and/ or frontal sinus with or without involvement of medial portion of the MS, ES or nasal cavity. There must be no concurrent malignancy.
4. Type IV: all tumors with any extranasal/ extrasinus extension to involve adjacent, contiguous structures such as orbit, intracranial compartment, pterygomaxillary space. All tumors are associated with malignancy.

The diagnosis can be tricky at times, hence radiological investigation provides an aid in diagnosis. CT scan (coronal section) shows bone erosion, thinning and remodelling with hyperdense areas with linear calcification which turns out to be one of the important investigation of choice. Though etiology remains little implicit, this tumor has a strong relationship with human papilloma virus with 40% of reported cases, raising the suspicion of implication in the pathogenesis of inverted papilloma.³⁻⁷ Treatment of choice is surgery either by Endonasal endoscopic approach or External approach (Mid-facial degloving) depending on

extension and tumoral characteristics. Follow-up is critical and mandate to diagnose local relapse which is often early but may also be late. The significance of this pathology lies in its connotation with carcinoma, which may be diagnosed at the outset or at recurrence during follow-up. It is important to diagnose recurrence to enable early treatment, especially in case of associated malignancy.⁴⁻⁹

4. Conclusion

This case report highlights the misleading behaviour of inverted sinonasal papilloma. Here, this simple, meek polyp that appeared as Antrochoanal polyp clinically ended up as complex, intricate Sinonasal papilloma histopathologically. Although a benign lesion but the triple catastrophic features of this tumor such as local ferociousness, inclination for recurrence and raised chances for malignant transformation typically differentiates sinonasal papilloma from the rest of the neoplasms in the sinonasal region and poses a real challenge to both surgeons and clinicians. This also suggests that any clinical, naked eye finding could still be deceiving, hence histopathological diagnosis is till date considered very accurate and decisive. Plus, association of HPV and Sinonasal papilloma accurately indicates the necessity for its summarizing in identifying patients at risk for recurrence and also to rule out probable chances of malignancy.

5. Source of Funding

None.

6. Conflict of Interest

Norn

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