

## Case Series

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# Exploring rare entities of sinonasal tumours from diagnosis to treatment: case series

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## ABSTRACT

Tumours arising from the nose and paranasal sinuses are diverse in nature and some are rarely reported in literature. Sinonasal tumours, though rare, represent a clinical challenge due to their complex presentation. Herein this case series studies about three unique cases of sinonasal masses i.e. sinonasal myxoma, esthesioneuroblastoma, follicular lymphoid hyperplasia which presented at our institution and their management. These nasal masses, presented with similar clinical features like nasal obstructions or with epistaxis, were diagnosed through clinical examinations, imaging, and histopathology, and were managed accordingly. Due to the delayed time of presentation in different age groups with common clinical features, the chances of misdiagnosis at an early stage are common. This necessitates for an early intervention and a multidisciplinary approach to manage such rare cases.

**Keywords:** Sinonasal myxoma, Esthesioneuroblastoma, Follicular lymphoid hyperplasia, Sinonasal tumors

## INTRODUCTION

Sinonasal tumours account for less than 3% of head and neck cancers and 0.8% of all human cancers.<sup>1</sup> Approximately 55% originate in the maxillary sinuses, 35% in the nasal cavity, 9% in the ethmoid and 1% in the frontal and sphenoid sinuses.<sup>1</sup>

Sinonasal tumours are uncommon and represent a heterogeneous group of head and neck lesions.<sup>2</sup> Paranasal sinus cancers include: squamous cell carcinoma (SCC), which comprises 50% to 80% of malignancies in this region; adenocarcinoma (ACA); adenoid cystic carcinoma; olfactory neuroblastoma; sinonasal undifferentiated carcinoma (SNUC); minor salivary gland tumours; melanoma; sarcomas; and other rare tumours.<sup>2</sup> According to the American Joint Committee on Cancer (AJCC), sinonasal malignancies are classified under the headings, “maxillary sinus, nasal cavity, and ethmoid sinus” whereas esthesioneuroblastoma is categorized under the heading, “brain and spinal cord.” sinonasal

malignancies have an incidence of 0.5–1 per 100 000 per year.<sup>3,4</sup>

Exposure to carcinogens like wood dust, asbestos is a significant risk factor for around 40% of reported sinonasal malignancies.<sup>5</sup> Malignant tumours in this area can occur at any age but when occupational factor is excluded, the male to female ratio is approximately 2:1.<sup>5</sup> The prognosis of Sinonasal malignancy is generally very poor with 5-year overall survival rate.

Extension into the orbit is a predictor of recurrence-free, disease-specific and overall survival. Majority of the tumour will involve erosion of the lamina, abutting the periosteum.<sup>5</sup> Given their often-poor prognosis, early detection and intervention are critical.

This study aims to present cases of rare sinonasal tumours that shared common symptoms, highlighting the importance of differential diagnosis.

## CASE SERIES

### Case 1

A 3-year-old male child presented to the outpatient department with complaints of bilateral nasal obstruction and intermittent episodes of nasal bleeding for the past two months. The nasal bleeding was about 5–10 ml per episode. The obstruction was progressive, affecting the child's breathing. Physical examination revealed a pinkish, pale, polypoidal mass in the left nasal cavity, extending posteriorly into the choana and nasopharynx. The nasal septum was significantly deviated to the right.

A computed tomography (CT) scan of the nose and paranasal sinuses (PNS) showed a well-defined, heterogeneously enhancing mass in the left nasal cavity with the following extensions: anteriorly, into the nasal cavity, posteriorly, through the choana into the nasopharynx and oropharynx as shown in Figure 1c. Superiorly, abutting the cribriform plate and body of the sphenoid bone and laterally remodelling the medial wall of the left maxillary sinus as shown in Figure 1a.

The mass was suspicious for an aggressive, though benign, pathology (Figure 1b). A biopsy revealed sinonasal myxoma, a rare benign mesenchymal tumour known for its locally invasive behaviour. A nasal endoscopy and biopsy were reported as extra gnathic sinonasal myxoma. The child underwent a medial maxillectomy with a lateral rhinotomy approach, and the mass was completely excised.

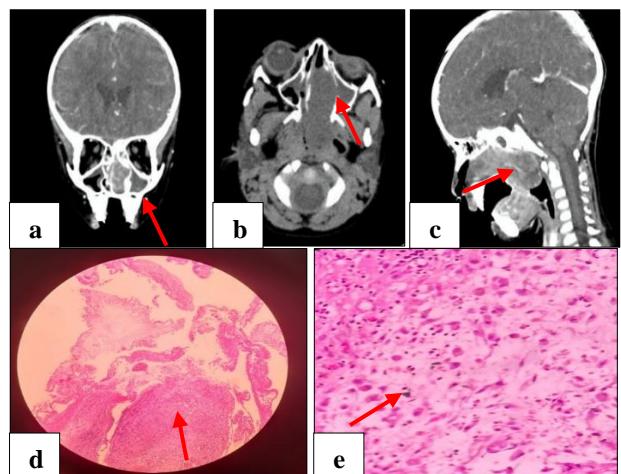
The resected tissue was sent for a frozen section examination reported as – myxoma as shown in Figure 1d. Final histopathology confirmed the diagnosis. Post-operative recovery was smooth, with no recurrence on follow-up imaging (Figure 1e).

### Case 2

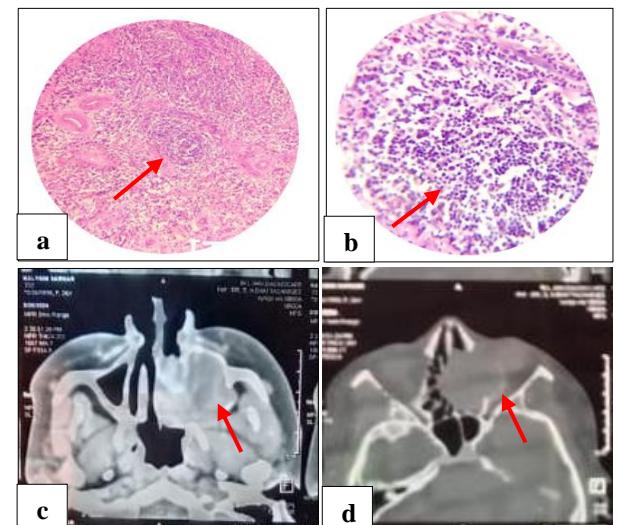
A 33-year-old female presented with progressive bilateral nasal obstruction and intermittent epistaxis from the left nasal cavity for six months. The patient also complained of watery discharge from her left eye, which had worsened over the previous few weeks.

Clinical examination revealed a pale, polypoidal mass in the left nasal cavity, which bled upon touch. Imaging via CT scan revealed a soft tissue mass filling the left maxillary sinus, causing displacement of the nasal septum shown in Figure 2d and erosion of adjacent bony structures as shown in Figure 2c.

Based on these findings, the patient was taken up for endoscopic sinus surgery. A fleshy mass was resected from the left maxillary sinus, and histopathological examination revealed follicular lymphoid hyperplasia (FLH) as shown in Figures 2a and b.



**Figure 1:** (a) CECT nose and PNS showing remodelling of maxillary sinus, (b) CECT nose and PNS showing Mass pushing the septum to the right, (c) CECT nose and PNS showing mass extending into the nasopharynx, (d) hematoxylin/eosin staining section showing myxoid stroma in 10X magnification, and (e) hematoxylin/eosin staining section myxoid stroma in 40X magnification.

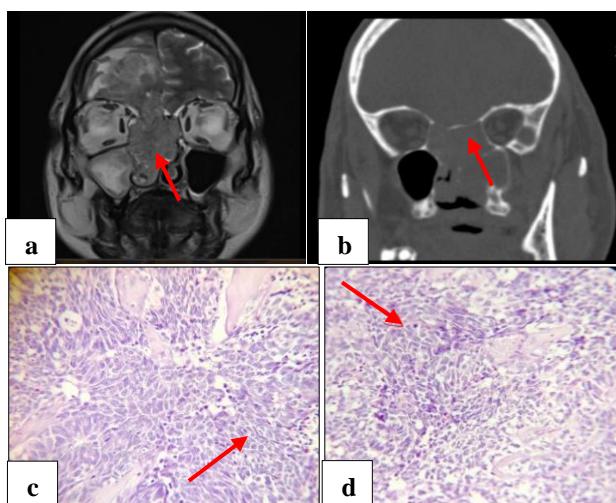


**Figure 2:** (a) Hematoxylin/eosin staining showing sub epithelium tissue shows lymphoid infiltration with few follicles of varying size (10X magnification), (b) hematoxylin/eosin staining showing lymphoid follicles in 40X magnification, (c) CECT nose and PNS showing mass in the left maxillary sinus causing adjacent bony erosions, and (d) CECT nose and PNS showing mass pushing the septum to the opposite side.

### Case 3

A 40-year-old male presented with progressive left-sided nasal obstruction and facial pain for three months. The patient also reported episodes of nasal bleeding and excessive tearing from his left eye. A detailed clinical examination revealed a polypoidal mass in the left nasal

cavity. CECT (Figure 3b) and magnetic resonance imaging (MRI) scans of nose and paranasal sinuses (Figure 3a) showed a large mass in the left nasal cavity and ethmoid sinus, with extension through the cribriform plate into the anterior cranial fossa, causing a midline shift of the brain. A biopsy of the mass confirmed the diagnosis of esthesioneuroblastoma, a rare malignant tumour originating from the olfactory neuroepithelium as seen in Figure 3c. Immunohistochemistry was done and was positive for synaptophysin, CD56, Pan CK and Ki67 which was suggestive for a high-grade neuroendocrine tumour of the sinonasal region. The patient was referred for a multidisciplinary treatment approach, including surgery followed by radiation therapy due to the intracranial extension of the tumour.



**Figure 3: (a) T2 W MRI showing intracranial extension of the mass, (b) CECT nose and PNS showing erosion of the cribriform, and (c) hematoxylin/eosin staining showing poorly differentiated carcinoma in 40X magnification.**

## DISCUSSION

Sinonasal tumours, though rare, can present significant diagnostic and therapeutic challenges due to their variable clinical presentations. They are often misdiagnosed as more common benign conditions such as chronic sinusitis or nasal polyps, which can lead to delays in diagnosis. As seen in these three cases, a wide range of pathology can manifest as common symptoms like nasal obstruction and epistaxis.

### *Sinonasal myxoma*

Sinonasal myxomas are extremely rare tumours, especially in the paediatric population. True extra gnathic osseous myxomas are extremely rare.<sup>6</sup> Myxomas are benign mesenchymal tumors of uncertain etiology.<sup>7</sup> Macroscopically, it is a gray to white, smooth, mucoid or gelatinous, unencapsulated mass. having characteristic clinical presentation of a painless swelling of the affected area, usually the nasolabial or paranasal region.<sup>8</sup>

Despite their benign nature, they are locally destructive and can cause significant morbidity due to bone erosion. Standard treatment of myxoma is surgical en bloc resection with a small margin of normal tissue, which can be difficult due to involvement of local structures and risk for disfigurement. Thus, conservative total resection, sparing critical structures and preserving organ function, appears to be the treatment of choice. Local recurrence occurs in approximately 25% of patients, and usually within 5 years. Therefore, sinonasal myxomas in the paediatric population present a challenging diagnostic dilemma because of the low incidence of the disease, histological similarities shared with other neoplasms and poor compliance.<sup>9</sup>

### *Follicular lymphoid hyperplasia*

Follicular lymphoid hyperplasia (FLH) is a very rare though benign reactive process of an unknown pathogenesis that may resemble a follicular lymphoma, clinically and histologically.<sup>10</sup> It occurs more frequently in older patients (mean age of 62 years) and it also presents a tendency to be more common in women.<sup>10</sup> It is also known as nodular lymphoid lesion or pseudo lymphoma, is a non-neoplastic lymphoproliferative disease. Initially reported by Adkins in 1973. FLH in submucosal tissues of the oral and maxillofacial areas are rare.<sup>11</sup> FLH involves various organs like the skin, orbit, nasopharynx, larynx, thyroid, lungs, gastrointestinal tract, breasts, spleen, pancreas, and liver. The sites of occurrence in the oral and maxillofacial regions are the palate, tongue, salivary gland, and cheek region. FLH is recognized as a non-neoplastic proliferative lymphoid lesion and shows very uncommon features. A definitive diagnosis of FLH can only be established through clinical and histopathological evaluation.<sup>11</sup> Histologically, FLH consists of well-circumscribed lymphoid follicles with distinct germinal centres and mantle zones. The germinal centres contain a mixture of small and large lymphoid cells, including cleaved and uncleaved cells, along with scattered macrophages. The parafollicular area contains varying numbers of B and T lymphocytes, along with immunoblasts. Immunohistochemically, FLH shows positivity for markers such as CD20, CD21, CD10, CD79a, and Bcl6.<sup>11</sup> The primary treatment for FLH is surgical excision, although radiotherapy has been applied in a few cases. In our case, the mass originated from the maxillary sinus, causing erosion of the medial maxillary wall and the anterior and lateral walls of the maxilla, leading to a provisional diagnosis of carcinoma maxilla. The patient underwent total tumor resection under general anesthesia, and histopathological examination revealed FLH. After six months of follow-up, the patient showed complete healing with no signs of recurrence.

### *Esthesioneuroblastoma*

Esthesioneuroblastoma (ENB) also known as olfactory neuroblastoma is a rare malignant neoplasm arising in the roof of nasal cavity.<sup>12</sup> ENB is a rare malignant neuro-

ectodermal tumour of the upper nasal cavity.<sup>13</sup> According to the WHO, ENB is defined as “a malignant neuro-ectodermal tumour that is assumed to originate from olfactory receptor cells present high in the nasal cavity”.<sup>14</sup> It accounts for 2–6% of intranasal tumours.<sup>11</sup> Common presenting symptoms of Esthesioneuroblastoma include nasal obstruction, epistaxis, facial pain, diplopia, proptosis, and anosmia. Dulguerov and Calca terra further modified the grading according to the sinus involvement (Table 1).<sup>15</sup> Its incidence is approximately 0.4–1/1,000,000 population per year in the adult population.<sup>16</sup> It is claimed to originate from the sphenopalatine ganglion, Jacobson’s vomeronasal organ, ectodermal olfactory placode, Loci’s ganglion, sympathetic ganglia of the nasal

mucosa, neuroepithelial cells of the olfactory membrane and the ectopic olfactory epithelium in the nasal mucosa.<sup>16</sup> Due to similar clinical presentation as chronic sinusitis, it is rarely picked up at an earlier stage of the disease. In the reported case, the patient presented with epistaxis and headaches for six months and developed altered sensorium during the hospital stay. MRI revealed the mass extending to the anterior cranial fossa, causing edema and midline brain shift. A multidisciplinary approach led to the diagnosis of a high-grade neuroendocrine tumor with multiple metastases. Immunohistochemistry (IHC) confirmed the diagnosis, and the patient was managed with radiation oncology. Due to the advanced stage at the time of diagnosis, the prognosis remained poor.

**Table 1: Dulguerov and Calca terra’s modified grading according to the sinus involvement.**

Stages	Grading
<b>T1</b>	Tumour involving the nasal cavity and/or paranasal sinuses, excluding the sphenoid, sparing most superior ethmoidal cells
<b>T2</b>	Tumour involving the nasal cavity and/or paranasal sinuses, including the sphenoid, with extension to or erosion of the cribriform plate
<b>T3</b>	Tumour extending into the orbit or protruding into the anterior cranial fossa
<b>T4</b>	Tumour involving the brain

## CONCLUSION

Due to similar clinical presentations of these tumours, delayed time of presentation and rarity in their diagnosis, it urges the clinician to diagnose it at an earlier stage and to chalk out a plan through a multidisciplinary approach for a better prognosis.

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