CASE REPORT

A Rare Presentation of Extra Nasopharyngeal Angiofibroma in the Hard Palate and Maxillary Sinus: A Case Report

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Abstract

Background: Juvenile nasopharyngeal angiofibroma (JNA) is a rare, histologically benign but locally aggressive vascular tumor predominantly affecting adolescent males. Extra nasopharyngeal angiofibroma (ENA), a less common subtype, differs significantly in clinical behavior, anatomical distribution, and demographic profile. ENA rarely arises from the hard palate, particularly in pediatric populations. Case Presentation: We report a rare case of ENA in a 14-year-old male presenting with persistent facial pain and swelling following minor trauma. Clinical examinations revealed facial asymmetry and intraoral swelling without classic symptoms such as nasal obstruction or epistaxis. Imaging demonstrated a destructive soft-tissue mass originating from the left hard palate and extending into the inferomedial maxillary sinus. Complete surgical excision under general anesthesia was performed without the need for preoperative embolization. Histopathological analysis confirmed angiofibroma, and the postoperative course was uneventful with no recurrence at follow-up. Conclusion: This case highlights an uncommon presentation of ENA in a pediatric patient involving the hard palate with maxillary sinus extension—an exceptionally rare site. Given the atypical symptoms and location, ENA should be considered in the differential diagnosis of pediatric facial masses. Early imaging and histopathological confirmation are key for appropriate management.

Keywords: Extra nasopharyngeal angiofibroma • hard palate tumor • pediatric facial swelling • angiofibroma case report • maxillary sinus invasion

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Introduction

Nasopharyngeal angiofibroma (NA) is an uncommon, non-malignant tumor with locally invasive behavior, primarily affecting adolescent males. It typically develops near the sphenopalatine foremen, originating from the posterolateral wall of the nasal cavity or the nasopharynx. Although histologically benign, JNA tends to infiltrate surrounding structures, including the paranasal sinuses, orbital region, and intracranial space. The most frequent presenting symptoms include unilateral nasal blockage and recurrent nosebleeds. Other associated clinical features may involve headaches, facial asymmetry or swelling, loss of smell, cranial nerve dysfunction, ear-related symptoms, and orbital complications.[1]

Because of their locally aggressive behavior and challenging anatomical location, juvenile nasopharyngeal angiofibromas (JNAs) can be associated with considerable morbidity. Although surgical removal remains the primary curative treatment—favoring endoscopic techniques over traditional open surgery—these procedures are complicated by the risk of significant intraoperative bleeding. One strategy to minimize blood loss during surgery is preoperative tumor embolization.[2]

Extranasopharyngeal angiofibroma (ENA) represents a rare subset of vascular tumors that differs significantly from the more commonly recognized juvenile nasopharyngeal angiofibroma (JNA) in both clinical presentation and behavior. Unlike JNA, which typically arises in the nasopharynx and shows a strong male predilection, ENA can develop in various regions throughout the upper aerodigestive tract, with the nasal septum being the most frequently affected site. Additionally, ENA exhibits a more balanced male-to-female ratio (approximately 2.13:1), contrasting with the almost exclusive male occurrence seen in JNA. Clinically, ENA often presents with more rapidly progressing symptoms, necessitating a distinct diagnostic and therapeutic approach. These differences underscore the

importance of recognizing ENA as a separate clinical entity rather than a variant of JNA [3].In this article, we report a rare case of extranasopharyngeal angiofibroma (ENA) originating from the hard palate with infiltration into the maxillary sinus—an exceptionally uncommon site and pattern of involvement for this tumor.

Case Presentation

A 14-year-old male presented to the ENT outpatient department at Dr. Ruth K.M. Pfau Civil Hospital, Karachi, with a one-year history of persistent pain and swelling on the upper left side of his face [Figure 1-(a)&(b)]. The pain was sharp, continuous, and radiated toward the left ear. Over time, the symptoms progressively worsened but showed partial improvement following the extraction of upper left teeth (teeth 3–8). The onset of symptoms was traced back to facial trauma sustained during play, when the patient was accidentally struck on the left side of his face by a ball. There was no associated history of fever or nasal obstruction; however, the patient did report mild episodes of nasal bleeding.

On clinical examination, mild facial asymmetry was noted due to fullness in the left maxillary region (Figure 1-a). Intraoral inspection revealed diffuse swelling in the left buccal and upper vestibular areas, which were mildly tender on palpation. There were no signs of cervical lymphadenopathy, jaundice or anemia and nasal examination was unremarkable. Initial laboratory tests, including a complete blood count (CBC), revealed a hemoglobin level of 11.2 g/dL, which is slightly below the reference range for males (11.5–16 g/dL), suggesting mild anemia. The total leukocyte count was 9.5×10^9/L and the platelet count was 240×10^9/L—both within normal limits. The hematocrit was measured at 31.2%. Overall, these values fall within acceptable limits for a 14-year-old male. Serum biochemistry results were also unremarkable, with a creatinine level of 0.5 mg/dL (slightly below the normal range of 0.9–1.3 mg/dL) and a blood urea nitrogen (BUN) level of 10 mg/dL, which is within the normal range (6–20 mg/dL). X-ray of the face (water's view) showed involvement of left maxillary region and facial asymmetry (Figure 1-b)



Figure 1. (a)Intraoral inspection of the face showing diffuse swelling and hyperemia in the left buccal mucosa. (b)X-ray of the face (water's view) showing involvement of left maxillary region and facial asymmetry

Unenhanced coronal CT exhibited erosion of lateral aspect of left hard palate (asterisk) with an associated ill-defined soft-tissue mass extending into the inferomedial left maxillary antrum. (Figure 2-a)

Plain CT scan of the maxillofacial region in axial view showed a destructive soft tissue density mass lesion of the left hard palate invading the left maxillary sinus. (Figure 2-b)

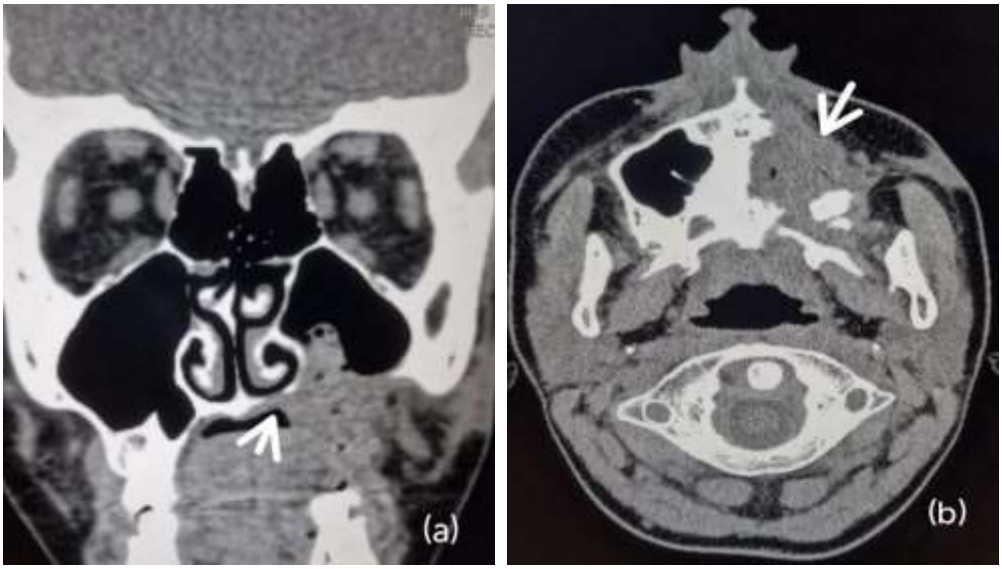


Figure 2. (a)Unenhanced coronal CT showing soft tissue mass extending into inferomedial maxillary antrum. (b) Plain CT of maxillofacial region in axial view showing soft tissue density mass lesion of left maxillary sinus.

The patient was taken for surgical excision of the lesion under general anesthesia. Intraoperatively, a vascular, non-encapsulated mass was identified involving the upper alveolar and buccal spaces, with displacement of three maxillary teeth.

Histopathological examination of the excised tissue revealed a grey-brown, irregular soft tissue mass measuring 2.5 × 1.5 × 0.5 cm. Microscopy showed a Vaso formative lesion with heavy chronic inflammatory infiltrate, abundant plasma cells, and areas covered with squamous and respiratory epithelium (Figure 3-b). Foci of necrotic mucoid material were also observed (Figure 3-a). The overall features were strongly suggestive of angiofibroma, with no evidence of malignancy or granuloma formation.

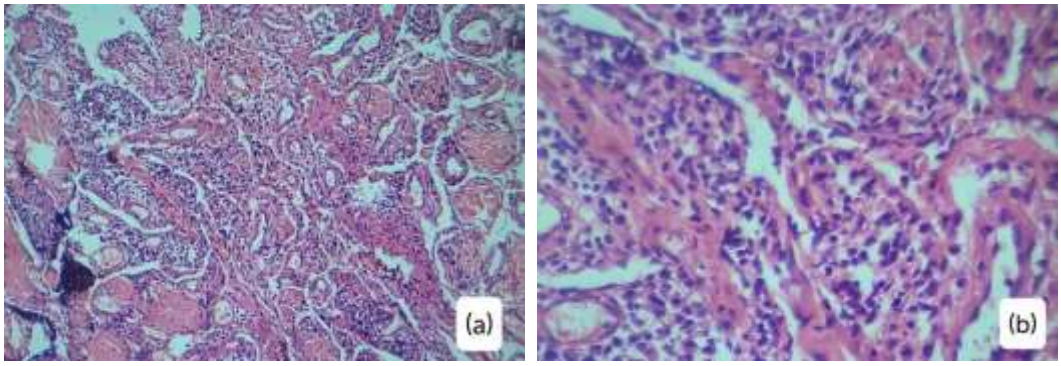


Figure 3. *[Caption to be added by the authors]*

A diagnosis of extra nasopharyngeal angiofibroma (ENA) was established based on clinical, radiological, and histopathological findings. The patient underwent complete surgical excision of the lesion, with minimal intraoperative blood loss and no complications. The postoperative course remained uneventful, with satisfactory wound healing and no evidence of neurological or functional deficits. The patient was discharged on the third postoperative day in stable condition. Follow-up at two weeks showed complete resolution of facial swelling and no signs of recurrence.

Discussion

Adolescent males can develop a rare vascular tumour called nasopharyngeal angiofibroma (NA). Extra nasopharyngeal angiofibromas (ENAs) are a rare finding that often affects women. Depending on the tumor site, it can present with nonspecific symptoms and an aggressive growth pattern. Their clinical presentation is more muddled, and it may take longer for a diagnosis to be made after symptoms start [4]We report a rare case of extra nasopharyngeal angiofibroma in a 14-year-old boy that started in the left hard palate, a very uncommon location, and extended into the inferomedial aspect of the maxillary sinus. It presented with atypical symptoms, notably lacking the classic features such as nasal obstruction or epistaxis, thereby posing a significant diagnostic challenge due to its unusual location and clinical presentation.

Because they are so uncommon, angiofibromas outside of the nasopharyngeal region may go undiagnosed. There have been reports of extra nasopharyngeal angiofibromas developing from a variety of locations in the head and neck. The nasal septum, maxillary sinus, ethmoid sinus, larynx, sphenoid sinus, cheek, conjunctiva, oropharynx, retromolar area, middle turbinate and inferior turbinate, external nose, hard palate, external ear, lacrimal sac, carotid bifurcation, esophagus, trachea, facial nerve, middle cranial fossa, and infratemporal fossa have all been identified as their possible origins [5] Windfuhr et al. [6] searched Medline and Google Scholar for literature and created a thorough list of 174 ENA cases till 2017. According to descriptions, these tumors most frequently develop along the nasal septum, inferior turbinate, and maxillary sinus. A few more cases of ENAs have been reported up to 2025 in various location [6–8]stating the hard palate as one of the most exceptional sites.

ENAs can exhibit locally aggressive behavior, including bony erosion, orbital invasion, Intracranial extension, and soft tissue infiltration. A case of maxillary extra nasopharyngeal angiofibroma has been reported with erosion of the anterior maxillary wall and thinning of the anterior wall of the pterygopalatine fossa in a middle-aged woman. [9] We report a case of ENA originating in the left hard palate invading into the inferomedial maxillary sinus, which has not been previously described in pediatric cases.

According to studies, compared to nasopharyngeal angiofibromas, ENAs have a more balanced sex ratio (male-to-female ratio: 2.13:1) and a wider age distribution (8-87 years). The majority of extra nasopharyngeal angiofibroma (ENA) cases occur in adult women, while pediatric cases are much less common. In a study by Szymańska et al, it has been reported that with a mean age of presentation of about 28

years, females accounted for about 25–26% of all ENA cases, indicating a preference for middle-aged adults and a different demographic pattern from juvenile nasopharyngeal angiofibromas. [4]

Pediatric extra nasopharyngeal angiofibromas are exceedingly rare, as in the literature review by Yan YY et al. Revealed that only 45 cases of the Pediatric age group have been reported so far, with 13.3 % girls and all others were boys. [10].Only one case of ENA originating from the posterior hard palate in a 14-year-old Pediatric patient has been reported so far. It shows no signs of invasion in the surrounding tissues and presents with the symptom of an Ulcerated mass with oral bleeding [11]. While our case reports ENA in a 14-year-old boy originating from the lateral hard palate invading into the inferomedial maxillary sinus and presenting with uncommon symptoms like dull pain and facial swelling, no epistaxis.

ENA are usually presented with uncommon symptoms depending on the tumor site, making diagnosis difficult. In our case, the patient presented with only dull pain and facial swelling, showing no typical signs of epistaxis, making the diagnosis challenging. For an accurate diagnosis, a specimen suspected of being angiofibroma must undergo precise histopathological examination. When angiofibroma is suspected, a biopsy should be avoided as it may result in excessive bleeding [12]

Extra nasopharyngeal angiofibromas are difficult to diagnose radiologically because of their multiple locations, so computed tomography and magnetic resonance imaging are helpful techniques to ascertain the location and spread of ENA.ENA contrast enhancement is typically only moderate or nonexistent because tumors frequently have poor vascularity [12]

In our case, Unenhanced coronal CT showed erosion of the lateral aspect of the left hard palate, with an associated ill-defined soft-tissue mass extending into the inferomedial left maxillary antrum.

The preferred course of treatment for ENA is total surgical excision [13] .Due to their lower vascularity, ENAs rarely require preoperative embolization [5]. In our case, the lesion was completely surgically removed while under general anesthesia.

Conclusion

This case highlights an unusual presentation of a 14-year-old child developing an extra nasopharyngeal angiofibroma that extends from the hard palate into the maxillary sinus. The diagnosis was difficult because of its rare location and lack of common symptoms like nasal blockage or epistaxis. Accurate identification required careful histological analysis and imaging. Early detection and total surgical removal prevent recurrence and effective treatment.

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Ethical considerations

Our institution does not require ethical approval for reporting individual cases or case series or short communication.

Conflict of interest

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Data availability

All data required for the study are included in the article. No new data is generated in this study.

Consent

Written informed consent is taken from patient parents for publication.

Patient consent

Informed consent was obtained.

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Conflict of interest

None

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