OPEN ACCESS
Global Journal of Research in Dental Sciences
ISSN: 2583-2840 (Online)
Volume 05 | Issue 02 | March – April | 2025
Journal homepage: https://gjrpublication.com/gjrds/
Case Report

A Synchronous Presentation of Pleomorphic Adenoma and Maxillary Polyp in An Adolescent Patient: A Rare Case Report and Clinical Insights

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DOI: 10.5281/zenodo.14965682 **Submission Date: 25 Jan. 2025 | Published Date: 04 March 2025**

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Abstract
Pleomorphic adenoma (PA) is a common benign salivary gland tumor that can arise from either major or minor salivary glands. While it is most frequently observed in older age groups, it is rarely seen in children and young adolescents. The concurrent occurrence of a pleomorphic adenoma with a maxillary sinus polyp is yet another uncommon presentation that poses unique diagnostic and therapeutic challenges. This case report discusses a 17-year-old female patient with the synchronous presentation of pleomorphic adenoma of the palate and a maxillary sinus polyp, detailing the clinical presentation, diagnostic workup, treatment strategy and follow-up. The rare coexistence of pleomorphic adenoma of the palate and maxillary sinus polyp highlights the need for comprehensive diagnostic assessment in the head and neck region. Prompt identification and complete surgical removal of the pleomorphic adenoma are vital to prevent recurrence or malignant transformation while addressing the maxillary polyp alleviates related symptoms. Early detection and appropriate intervention are key to achieving favorable outcomes and preserving oral function and esthetics in affected patients.

Keywords: *Pleomorphic adenoma; palate; maxillary polyp; minor salivary gland neoplasm.*

INTRODUCTION
Synchronous tumors are the simultaneous occurrence of two or more distinct tumors in the same individual.[1] Synchronous tumors of salivary gland occurrence are uncommon and often associated with malignant forms.[2,3] Their management requires accurate diagnosis, multidisciplinary coordination, and tailored treatment to address each lesion effectively.

Conversely, Salivary Gland Tumors (SGTs) are extremely rare in children, comprising less than 5% of cases compared to adults. Pleomorphic adenoma (PA) is the most common type of salivary gland tumor, typically appearing in the 4th and 5th decades of life, and its occurrence in children and young adolescents is exceptionally rare.[4] PA has a slight female predilection and favors the palate as the most common site accounting for about 60% of the cases typically presenting as a slow-growing, firm, and painless mass.[5,6] In contrast, maxillary sinus polyps, also referred to as Antro choanal polyps (ACP), are benign growths originating from the mucosal lining of the maxillary sinus, often leading to nasal obstruction or sinus-related symptoms.[7]

Imaging studies, such as CT and MRI, are instrumental in distinguishing the nature and extent of both lesions, facilitating accurate diagnosis and surgical planning. Complete surgical excision with adequate margins is the treatment of choice for PA to prevent recurrence and potential malignant transformation. Management of the maxillary sinus polyp typically involves endoscopic sinus surgery. This report emphasizes the significance of comprehensive evaluation and tailored treatment approaches in pediatric cases involving synchronous benign lesions in the head and neck region.

CASE REPORT

A 17-year-old female patient presented to the dental clinic with the chief complaint of swelling concerning the left upper first molar region for about 3 weeks. The patient had no associated medical history. Intraoral examination revealed an oval-shaped growth in the left hard palatal region concerning the tooth region, measuring approximately 3x3 cm. The growth extended anteriorly to within 2 mm of the palatal rugae, posteriorly to 3 mm from the posterior palatal seal, medially to 5-7 mm from the marginal gingiva, and laterally involving the mid-palatine raphe. The overlying mucosa appeared intact with a bluish discoloration (Figure 1A). On palpation, the growth was edematous, had a smooth surface, and was fixed to the underlying mucosa. Clinical differential diagnoses included odontogenic cysts and tumors, soft tissue tumors, and salivary gland tumors.

Imaging studies, including CT and MRI of the neck, revealed a well-defined lesion originating from the soft tissue lining the undersurface of the left side of the hard palate, measuring approximately 23 x 20 x 16 mm. The radiographic examination also revealed an incidental finding of an 18 x 18 mm retention cyst located on the floor of the left maxillary sinus (Figure 1B). Additionally, lymph node enlargement was observed, measuring 24 x 10 mm in the left submandibular region and 10 x 6 mm in the submental region.

Fine-needle aspiration cytology (FNAC) of the palate revealed characteristics consistent with a salivary gland neoplasm, suggesting pleomorphic adenoma. After which a wide local surgical excision and reconstruction was planned.

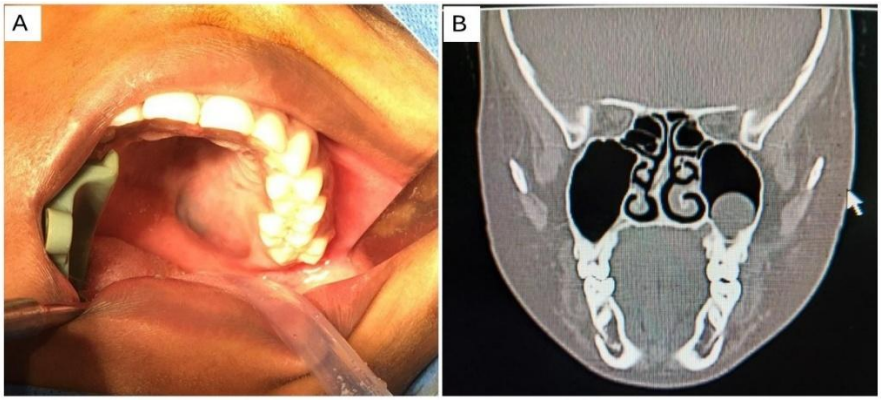

FIGURE 1: (A) Photograph shows the site and extent of the lesion. **(B)** CT -coronal view showing a unilateral presentation of polyp in left maxillary sinus with adjacent scalloping of the palatine process of maxilla with thinning of the floor of the left maxillary sinus.

After obtaining the patient's informed consent, general anesthesia was administered, and nasotracheal intubation was performed. An incision was made along the palatal region using electrocautery (Figure 2A). A palatal flap was raised, exposing the bone, after which bone guttering was performed, and the lesion was excised along with vital tissue (Figure 2B, C, & D). Additionally, through a high vestibular incision, a window was created in the palatal region to access and remove the maxillary sinus polyp. The defect was closed using the buccal fat pad (Figure 2E & F) and hemostasis was successfully achieved.

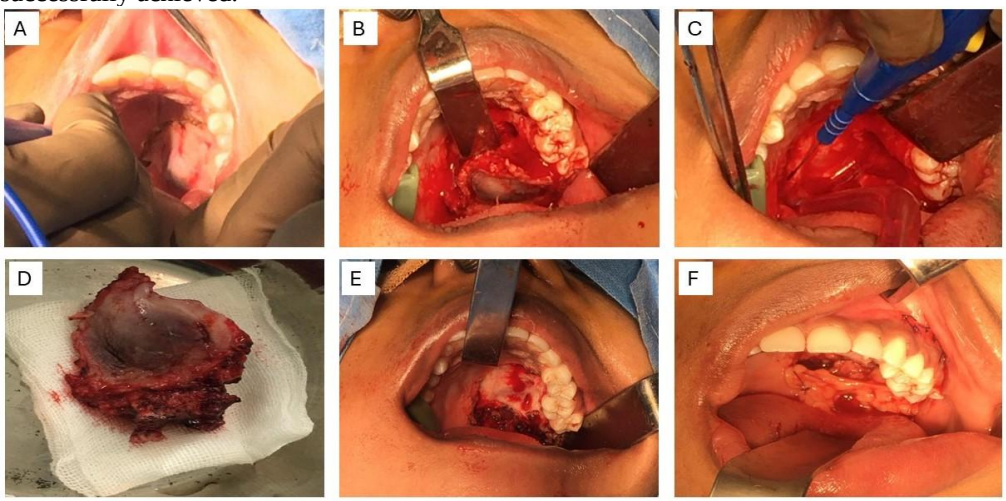

FIGURE 2: (A) Photograph shows the incision placed using electrocautery. **(B&C)** Intraoperative images showing the raising of the palatal flap and degloving of the lesion. **(D)** Photograph showing the postsurgical specimen of pleomorphic adenoma. **(E&F)** Photograph showing the reconstruction of the defect area with the buccal pad of fat.

Histopathological examination from the excisional biopsy of the palatal region revealed nests of epithelioid cells with uniform nuclei, surrounded by myoepithelial cells in a myxoid background, along with dilated ductal structures. No evidence of malignancy was observed. Tissue sections from the maxillary sinus region showed ciliated columnar epithelial lining with connective tissue composed of collagen fibers and numerous dilated blood capillaries. Based on these findings, a final diagnosis of pleomorphic adenoma of the palate and a benign cystic lesion of the maxillary sinus was established.

Postoperatively, the patient developed a palatal defect (Figure 3A). An obturator was provided to cover the defect, facilitating functional and aesthetic restoration. Satisfactory healing was observed during subsequent follow-up visits and the patient is under constant follow-up for two years with no signs of recurrence. (Figure 3B, C&D).

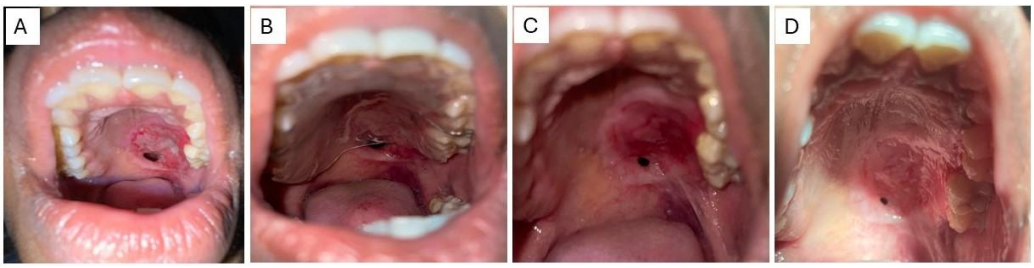

FIGURE 3: (A) Postoperative follow-up image. **(B, C&D)** An obturator was given to cover the palatal defect.

DISCUSSION

Salivary gland neoplasms account for about 6%–8% of all head and neck cancers.[8] As the most common benign neoplasm of the salivary glands, PA represents 70% of cases, with a higher predilection for females.[9] Typically, pleomorphic adenomas are seen in individuals between the third and fifth decades of life, with a mean age of 44.14 years.[10] However, in our case, the patient was only 17 years old. PA is infrequently seen in children, with involvement of the minor oral salivary glands being even rarer. The palate is the most common intraoral site, followed by the upper lip and buccal mucosa.[4,11] Pleomorphic adenoma typically presents as a painless, slow-growing, firm mass. When it involves the palate, it may appear fixed to the underlying bone but does not invade it. Other differential diagnoses must be considered when involving the palate.[12]

Investigative procedures like CT and MRI are crucial for determining the site and extent of lesions. In the present case, they also uncovered an additional lesion in the maxillary sinus. FNAC is strongly recommended as a preliminary diagnostic approach to differentiate benign and malignant tumors.[13] PA is characterized histologically by a combination of epithelial, myoepithelial, and mesenchymal elements, giving it a mixed appearance. The epithelial cells are arranged in diverse patterns embedded within a chondromyxoid or fibro myxoid stromal matrix. The complex and heterogeneous architecture is a hallmark feature of pleomorphic adenoma.[10]

On the contrary, ACP is a benign, solitary polypoid lesion that primarily affects adults and is uncommon in children. It arises from hypertrophy of the mucous membrane in the maxillary sinus antrum and extends through the maxillary sinus ostium into the nasal cavity and choana. They typically exhibit a distinctive pear-shaped structure, with the thicker portion often containing a large cystic space.[7] Several theories suggest that the development of Antro choanal polyp (ACP) may be linked to chronic inflammatory conditions, either allergic or infectious.[7,14] Histopathological examination reveals a central cystic cavity lined by respiratory epithelium surrounded by a homogeneous stroma.[15]

Surgical excision remains the primary treatment for PA of the palate and ACP, followed by close monitoring.[16]

CONCLUSION

The coexistence of PA of the palate and ACP is a rare clinical occurrence, highlighting the need for comprehensive diagnostic evaluation in the head and neck region. Considering the patient's age, prompt identification and complete surgical removal of the pleomorphic adenoma are vital to prevent recurrence and malignant transformation, while managing the sinus polyp addresses related symptoms. A multidisciplinary approach and diligent follow-up are key to achieving optimal outcomes in these complex cases.

ACKNOWLEDGMENT

There is no source of financial support and industry affiliations for all those involved.

CONFLICT OF INTEREST

The authors declare that there are no conflicts of interest related to this case report.

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CITATION

V.S Sakthivel, K. Sumithra, S. S. Kumar, Avinash JB, S. Chandana Priya, & K Janani. (2025). A Synchronous Presentation of Pleomorphic Adenoma and Maxillary Polyp in An Adolescent Patient: A Rare Case Report and Clinical Insights. In Global Journal of Research in Dental Sciences (Vol. 5, Number 2, pp. 7–10). https://doi.org/10.5281/zenodo.14965682