

Endoscopic Management of Intranasal Pleomorphic Adenoma: A Case Report

Siti Nursiah, Andrina Y.M. Rambe, Irnanda W.R. Nasution
Department of Otorhinolaryngology Head and Neck Surgery, Faculty of Medicine,
University of North Sumatra, Medan 20155, Indonesia

Abstract:- Pleomorphic adenoma is one kind of tumor which is the most common benign tumor of major salivary gland tumors. And can occur in the minor salivary gland tissue that can be found in the nasal cavity. Intranasal pleomorphic adenomas are usually formed by the mucous glands of the nasal septum. We found the pathology result of endoscopic surgery. The purpose of this study was to demonstrate an endoscopic approach to surgery for pleomorphic adenoma of the nasal septum. A 34-year-old woman presented with complaints of nasal congestion and occasional bleeding for at least 3 years. We provide treatment with endoscopic surgery to remove the tumor. In this case we resected the tumor and also the submucous part of the nasal septal to reach tumor-free margins. Microscopic examination shows epithelial tissue, mesenchymal composed of myxoid stroma and also myoepithelial component with H&E staining. The postoperative follow-up of the patient was good, and there were no complications found in the patient. Benign nasal tumors of the minor salivary glands or benign pleomorphic adenomas of the nose are rare tumors and should be considered in their management. A correct histological diagnosis can be confirmed. Treatment for this disease can be done endoscopically endonasal surgery.

Keywords:- pleomorphic adenoma, histopathology, nasal septum, endoscopic surgery.

I. INTRODUCTION

Pleomorphic adenoma is a benign tumor of the major salivary glands that is very frequent. Minor salivary glands in the nasal cavity, hypopharynx, and glands of the larynx or trachea can also be affected. Nonetheless, intranasal pleomorphic adenoma, particularly septal pleomorphic adenoma, is uncommon. Although most salivary glands are found in the lateral wall of the nose and concha, intranasal pleomorphic adenoma mainly arises from glands of the nasal septum mucosa.¹

Salivary gland tumors account for 3% approximately of all head and neck cancers. The benign mixed tumor of salivary glands, also known as Pleomorphic adenoma, occurs primarily in the major salivary glands, particularly 70 % from the parotid gland, and less frequently 15-25 % in the submandibular salivary glands and sublingual salivary glands, because of morphological differences with parts of the epithelial and mesenchymal components. Only about 8% until 10% of Pleomorphic adenoma arise from the small salivary glands. From the soft palate, hard palate, lip, lacrimal

gland, also external auditory canal, there have been a few cases documented.²

The tumor's pathophysiology is yet uncertain. The cause of nasal septal pleomorphic adenoma can be attributed to faulty embryonic ectoderm cell sites, remnants of the vomeronasal organs, possibly triggered by some viral infection, according to some theories. Intranasal pleomorphic adenomas differ from other salivary gland, pleomorphic adenomas in minor salivary gland as they contain a higher cellularity and an epithelial tissue with a low stromal component. This tumor also does not have a capsule.

Intranasal Pleomorphic Adenoma is an essential differential diagnosis entity despite its rarity in this site. Nasal congestion and nasal bleeding are the predominant symptoms, which are not typical. It is necessary to conclude the component of epithelial parts paired with mesenchymal for the histological diagnosis about a mixed tumor. The preferred treatment of this tumor is surgical resection.²

II. CASE REPORT

A 34-year-old woman, presented with main complaint of nasal obstruction since 3 years ago which is getting worse over time. The patient also had history of rhinorea mixed with blood and nasal finger picking. During ENT routine examination, found ears were normal, anterior rhinoscopy showed of the right nasal cavity was found mass covering the nasal cavity, pale color and hypervascular, inferior turbinates were difficult to examine, the nasal septum was not deviated, the left nasal cavity was normal. The oropharynx was normal. The CT scan of the paranasal sinus with contrast and the result was found anterior septum mass, homogenous lesion with soft density in the right nasal and the paranasal sinus were normal.



Fig 1 CT / Computerized Tomography scan of the paranasal sinus with contrast and the result was found anterior septum mass, homogenous lesion with soft density in the right nasal

The patient underwent endoscopic surgical resection and resected the tumor completely also resected the submucosa and the normal tissue of the nasal septum to prevent the tumor from recurrence and malignant transformation. Macroscopically, the specimens were pink and pale in color, consistency of the tumor is solid, with the surface lobulated and smooth. The mass was sent for histopathological examination, which result features nasal septal mass. Evaluation 1 week post operative, in the right nasal cavity was found crust cover the anterior nasal septum.



Fig 2 the specimens were pink and pale in color, solid in consistency, and lobulated smooth surface, diameter 1,5-2 cm



Fig 3 In the right nasal cavity was found crust cover the anterior nasal septum

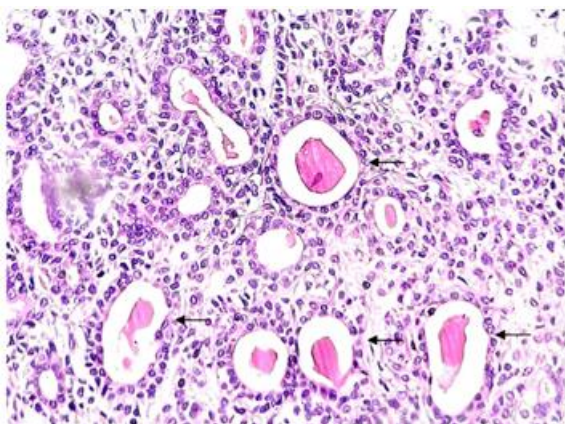


Fig 4 The glands are proliferating with shapes and sizes vary (arrows)

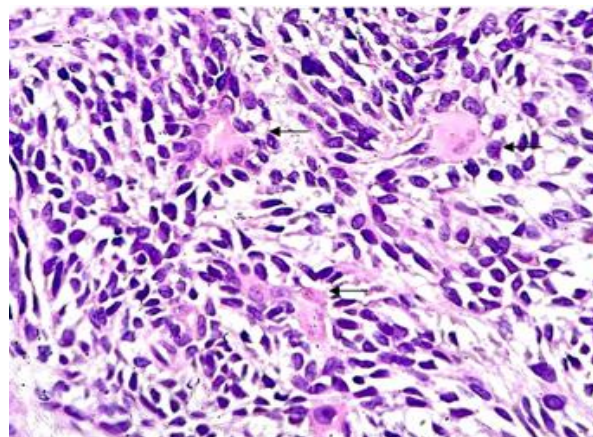


Fig 5 myoepithelial proliferation with nuclei within normal limits (arrows)

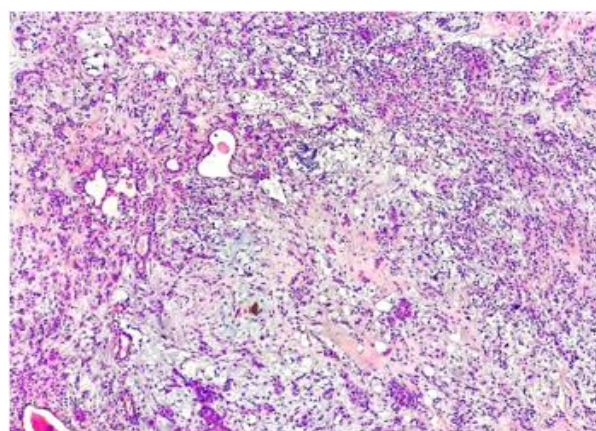


Fig 6 Myxoid mesenchymal components, with low normal stroma

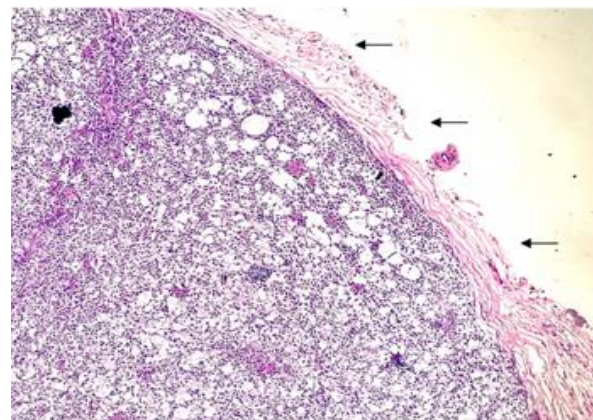


Fig 7 Capsule of the mass (arrows)

Histopathological result for the specimen was Pleomorphic Adenoma Intranasal. The microscopic shown partly pseudostratified columnar epithelial lining and partly cells squamous with nuclear morphology within normal limits. It is also seen that the glands are proliferating with shapes and sizes vary. Also seen myoepithelial proliferation with nuclei within normal limits. Stroma consists of a broad myxoid. Proliferative vessels. No signs of malignancy were found.

III. DISCUSSION

The tumors of salivary gland account for 3% approximately of all head and neck cancers. Pleomorphic adenomas are mostly produced by the major salivary glands (MSG), with only 8 to 10% produced by the minor salivary glands from the upper aerodigestive tract, which includes the nasal cavity, part of pharynx and larynx, trachea, external ear canal, and the glands of lacrimal.^{3,4} In spite of the fact that benign neoplasms are the most prevalent benign neoplasm in the minor salivary gland, about 15% of the establishment of these glands are benign, minor salivary glands are described by an advanced rate about 85% malignant kind of neoplasms.⁵ Because intranasal pleomorphic adenoma is very not common, from the statistic, only two significant series have been published. Between 1949 and 1974, Campagno and Wong examined 40 examples of intranasal mixed malignancies.⁶ There were 23 females and 17 males among the patients. The majority of the malignancies (25 from 40 patients) came from the mucosa of the nasal septal. Moreover, Intranasal pleomorphic adenoma showed no gender preference, unlike tumors from the major salivary glands, those are known to be more frequent in women. They were capable to gather data from follow-up 85% of the patients and identified a recurrence rate about 10%. (3 from 34 patients). There was no malignant transformation of the lesions in any of the patients. Suzuki later explained a case of intranasal pleomorphic adenoma and published a Japanese national study with 41 patients in 1990, however the female predominance was not statistically significant (1:1.18).⁷ Only four cases began in the lateral wall of the nasal cavity, whereas the other ninety percent of cases began in the nasal septum. One case (2.4 percent) recurred, while another developed a malignant change (2.4 percent). Vento published a nationwide Finnish study with ten cases in 2016, and the findings were comparable to the previous ones: the most of pleomorphic adenomas about 60 % arised from the nasal septum, female preponderance, no malignant transformation also no recurrence, however six cases were less than one year.⁸

Several explanations have been offered to describe the unusual development of nasal septal mucosa pleomorphic adenoma, despite the fact that seromucosal glands are generally present in the lateral side of nasal wall.

Vomer nasal organ, a tube lined with epithelium of cartilage septum that degenerates in the early fetal period, is said to be the source of the nasal septum, according to Stevenson PA.⁹ Septal PA, according to Matthew et al, is caused by ectoderm-derived embryonic epithelial cells that have been misplaced and transported into the septal region via the nasal pits.¹⁰ Evans and Cruickshank, on the other hand, believe that pleomorphic adenoma are epithelial in origin also form in fully mature salivary gland tissue rather than embryonic leftovers.¹¹

The role of Epstein-Barr virus (EBV), human cytomegalovirus (HCMV), human papilloma virus (HPV), and human herpes virus 8 (HHV-8), in the pathogenesis of pleomorphic adenoma has been explored. Atula showed EBV DNA positivity from a pleomorphic adenoma developing intranasal in 1998, but no HPV, HHV-8, or CMV DNA.¹² Malinvaud et al. described three examples of pleomorphic adenomas of the nasal septum, all of which had EBV-related blood antibodies and positive EBV-DNA detection in the tumor.¹³ In terms of carcinogen exposure in the environment, Zheng published a study with 41 cases of salivary gland neoplasms and 414 controls, finding the dust of silica exposure was linked to a 2.5-fold increased risk of salivary gland cancer,¹⁴ and Horn- Ross looked at 199 cases of salivary gland neoplasms that were linked to rubber workers exposed to nitrosamines.¹⁵

Clinical aspects of intranasal PA include a painless nasal swelling and the slow growing of the mass, thus clinical signs do not manifest until after a lengthy period of silence. The gradual severity of occasional epistaxis and unilateral nasal obstruction are the most prevalent symptoms. An outward swelling of the nasal pyramid, as well as pain, is less typical when the tumoral mass grows to a reasonably considerable size. It has the appearance of a smooth, with a soft substance and pink-pale lobulated mass. The lack of ulceration and invasion of neighboring structures suggest that the mass is benign.

Intranasal pleomorphic adenomas differ from other salivary gland, pleomorphic adenomas in minor salivary gland as they contain a higher cellularity and an epithelial tissue with a low stromal component. This tumor also does not have a capsule.¹ As in this case the histopathological pattern of the minor salivary gland tumor has epithelium dominant rather than the stromal component and also surrounded by incomplete non-invaded fibrous capsule.

Polyps of the nose, angiofibromas, osteomas, papillomas, squamous cell carcinoma, adenocarcinoma, mucoepidermoid carcinoma, adenoid cystic carcinoma, olfactory esthesioneuroblastoma, and melanoma are all other diagnosis of benign and malignant tumors. Computed tomography can show the extent of the tumor's invasion, or lack thereof, into the surrounding tissue, as well as, in many circumstances, the tumor's starting place.

When the capsule is disrupted and direct contact with around of healthy tissue is found, surgical removal including surrounding normal tissue is the best choice to prevent recurrence. Local recurrence, malignant transformation, and metastasis are the main dangers. After surgical excision, recurrence rates range from 0% to 8%, with numerous recurrences increasing the chance of cancerization. In the absence of resection, the probability of malignant transformation is 6% and is expected to be 1.5 percent within 5 years.¹⁶ At three years' follow-up, Compagno and Wong found 3 occurrences of local recurrence in 40 patients (7.5 percent).⁶

IV. CONCLUSION

We reported a case of nasal septal pleomorphic adenoma. However quite rare, intranasal pleomorphic adenomas should be considered when a slow-growing unilateral mass of the nasal cavity is found. The diagnosis of nasal septal pleomorphic adenoma is not only very rare in terms of the location but not least also in demonstration on anterior rhinoscopy and tomographic scans. Accurate diagnosis in these cases can be achieved through endoscopic surgical resection and specified examination of histopathology. The patient was treated with an endoscopic surgical resection of the mass. Pleomorphic Adenoma requires long-term setting of the follow-up, both endoscopic and radiologic, to eliminate the development of persistent illness into malignant.

REFERENCES

- [1]. Wierzchowska M, et al. Rare benign pleomorphic adenoma of the nose: short study and literature review. *Videosurgery Miniinv* 2015; 10 (2): 332–336
- [2]. Henriques V, et al. Septal Nasal Pleomorphic Adenoma: Case Report And Literature Review. *Acta Otorrinolaringol. Gallega* 2017; 10(1): 1-7
- [3]. Eveson JW, Cawson RA. Tumours of the minor (oropharyngeal) salivary glands: a demographic study of 336 cases. *J Oral Pathol.* 1985;14(6):500-9.
- [4]. Sciandra D, Dispenza F, Porcasi R, Kulamarva G, Saraniti C. Pleomorphic adenoma of the lateral nasal wall: case report. *Acta Otorhinolaryngol Ital.* 2008;28(3):150-3.
- [5]. Saade RE, Bell DM, Hanna EY. Benign neoplasm of the salivary glands. In *Cummings Otolaryngology, Head and Neck Surgery*, 6th Edition. Ed. Saunders; 2015. p 1245–1246
- [6]. Compagno J, Wong RT. Intranasal mixed tumors (pleomorphic adenomas): a clinicopathologic study of 40 cases. *Am J Clin Pathol.* 1977;68(2):213-8.
- [7]. Suzuki K, Moribe K, Baba S. [A rare case of pleomorphic adenoma of lateral wall of nasal cavity - with special reference of statistical observation of pleomorphic adenoma of nasal cavity in Japan]. *Nihon Jibiinkoka Gakkai Kaiho.* 1990;93(5):740-5.
- [8]. Vento SI, Numminen J, Kinnunen I, Rautiainen M, Tarkkanen J, Hagstrom J, et al. Pleomorphic adenoma in the nasal cavity: a clinicopathological study of ten cases in Finland. *Eur Arch Otorhinolaryngol.* 2016;273(11):3741-5.
- [9]. Stevenson HN. Mixed tumor of the septum. *Ann Otol Rhinol Laryngol.* 1932;41:563–570
- [10]. Matthew S, Ersner MD, Saltzman M. A mixed tumor of the nasal septum. Report of a case. *Laryngoscope.* 1944;54:287–296
- [11]. Evans RW, Cruickshank AH: *Epithelial tumors of the Salivary Glands*. Vol. 1 of the series *Major Problems in Pathology*. Philadelphia, W. B. Saunders, 1970.
- [12]. Atula T, Grenman R, Klemi P, Syrjanen S. Human papillomavirus, Epstein-Barr virus, human herpesvirus 8 and human cytomegalovirus involvement in salivary gland tumours. *Oral Oncol.* 1998;34(5):391-5.
- [13]. Malinvaud D, Couloigner V, Badoual C, Halimi P, Bonfils P. Pleomorphic adenoma of the nasal septum and its relationship with Epstein-Barr virus. *Auris Nasus Larynx.* 2006;33(4):417-21.
- [14]. Zheng W, Shu XO, Ji BT, Gao YT. Diet and other risk factors for cancer of the salivary glands: a population-based case-control study. *Int J Cancer.* 1996;67(2):194-8.
- [15]. Horn-Ross PL, Ljung BM, Morrow M. Environmental factors and the risk of salivary gland cancer. *Epidemiology.* 1997;8(4):414-9.
- [16]. Baron S, et al. Pleomorphic adenoma of the nasal septum. *European Annals of Otorhinolaryngology, Head and Neck diseases* (2014) **131**, 139—141