Acta Clin Croat 2023; 62:387-390

doi: 10.20471/acc.2023.62.02.20

Case Report

INTRANASAL PLEOMORPHIC ADENOMA ARISING FROM THE LATERAL NASAL WALL

Ljiljana Jovančević[1,2], Aleksandra Fejsa Levakov[1,3] and Bojana Jovančević[1]

[1]University of Novi Sad, Faculty of Medicine, Novi Sad, Serbia;
[2]University Clinical Center of Vojvodina, Department of Otorhinolaryngology and Head and Neck Surgery, Novi Sad, Serbia;
[3]University Clinical Center of Vojvodina, Department of Pathology and Histology, Novi Sad, Serbia

SUMMARY – Pleomorphic adenoma is very rare in the sinonasal region, with the most common localization on the nasal septum, followed by lateral nasal wall. In the case presented, a 72-year-old woman was complaining of the right sided nasal obstruction without any other symptoms. The symptom started a year before and increased progressively. Anterior rhinoscopy revealed a mucosa-covered, smooth-surfaced, soft, polypoid, pale, grayish-pink in color mass in the right nasal cavity, approximately 2x2 cm in size. Nasal endoscopy showed the mass to have a broad base on the lateral nasal wall. Computerized tomography scan showed a homogeneous, solid soft tissue mass, 25x18x12 mm in size, which was attached to the lateral nasal wall, behind the nasal vestibule, just in front of the inferior turbinate. Endonasal endoscopic complete tumor excision was performed, during which some spillage of the tumor occurred. Histology diagnosis was pleomorphic adenoma of minor salivary glands. The patient was followed up on regular basis and had no tumor recurrence in the 6th postoperative year. Intranasal pleomorphic adenoma arising from the lateral nasal wall in front of the inferior turbinate is extremely rare, so the presented case is probably the first ever published.

Key words: *Pleomorphic adenoma; Nose neoplasms; Endoscopic endonasal surgery*

Introduction

Pleomorphic adenoma (PA) is the most common neoplasm of salivary glands. It occurs mostly in major salivary glands. In minor salivary glands, it can occur anywhere within their distribution in the upper aerodigestive tract, where the incidence is 8%-10% of PA cases[1,2].

Pleomorphic adenoma is rare in the sinonasal region. Rha *et al.* published a single institution case series combined with a comprehensive review of the literature[1]. Their search of the literature over a 50-year period yielded only 93 published cases of sinonasal PA (+8 cases of carcinoma *ex* PA), and the authors described seven cases of their own. They also found that sinonasal PA involves patients in the fifth decade of life on average, with some female predominance[1]. Patients usually presented within 1 year of the onset of symptoms, with the most frequent symptom being unilateral nasal obstruction, followed by epistaxis[1-4] . Karligkiotis *et al*. published a case series of 5 patients, suggesting a total number of less than 120 cases (arising from the nasal septum, lateral nasal wall, and nasopharynx) published so far[4]. In the sinonasal region, the most common localization of PA is nasal septum, followed by lateral nasal wall, usually the inferior turbinate[1-4] .

Correspondence to: *Assoc. Prof. Ljiljana Jovančević, MD, MS, PhD*, University of Novi Sad, Faculty of Medicine, Hajduk Veljkova 3, 21000 Novi Sad, Vojvodina, Serbia
E- mail: ljiljana.jovancevic@mf.uns.ac.rs

Received June 3, 2022, accepted July 11, 2022

Intranasal PA is a painless, slow-growing mass, so computerized tomography (CT) will show nonspecific benign features[5]. Usually, it is a well-defined soft-tissue mass, which can show adjacent bony displacement and expanding erosion. Magnetic resonance imaging (MRI) will show iso- to hyperintensity on T2-weighted images (WI) and hypointensity on T1-WI[5].

Complete surgical excision of the tumor is the treatment of choice for intranasal PA, which can be achieved with endonasal endoscopic surgical approach in most cases[1,2]. Approximately only 50 endoscopically treated cases have been reported in the literature so far[4].

On histology, intranasal, like any other PA, is a mixed (two-component) tumor. There is a cellular component comprising epithelial and myoepithelial cells, and mesenchymatous stroma (myxoid, hyaline, chondroid, and osteoid)[5]. Intranasal PAs have a more dominant epithelial component (*vs.* stromal components) than is seen in the major salivary gland PAs[3,5].

Case Report

A 72-year-old woman was referred to our Department of Otorhinolaryngology and Head and Neck Surgery for evaluation of unilateral nasal obstruction. She was complaining only of the right sided nasal obstruction without any other symptoms. The symptom started a year before and increased progressively.

Anterior rhinoscopy revealed a mucosa-covered mass in the right nasal cavity, approximately 2x2 cm in size. The mass was smooth-surfaced, soft, polypoid, pale, grayish-pink in color. Nasal endoscopy showed the mass to have a broad base on the lateral nasal wall (Fig. 1). CT scan showed a homogeneous, solid soft tissue mass, 25x18x12 mm in size, which was attached to the lateral nasal wall, behind the nasal vestibule, just in front of the inferior turbinate, with no intratumoral calcifications (Fig. 2). The mass pushed against the surrounding structures without invading them. The radiologist's diagnosis was a slow growing benign lesion.

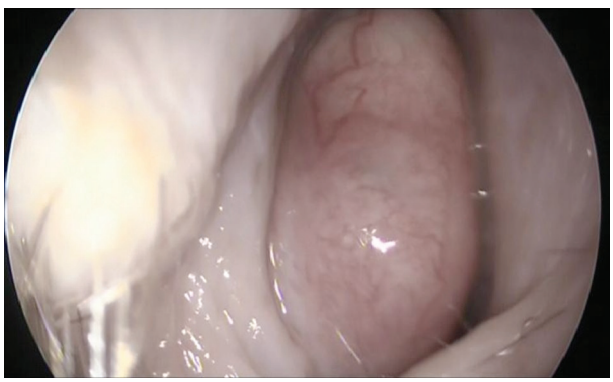

Fig. 1. Nasal endoscopy of the intranasal pleomorphic adenoma.

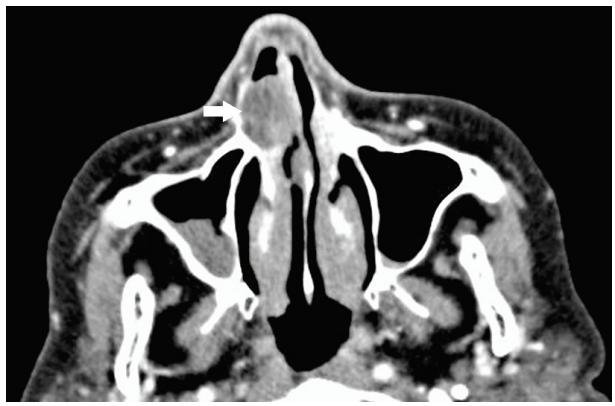

Fig. 2. Axial computed tomography scan of the intranasal pleomorphic adenoma (tumor marked with arrow).

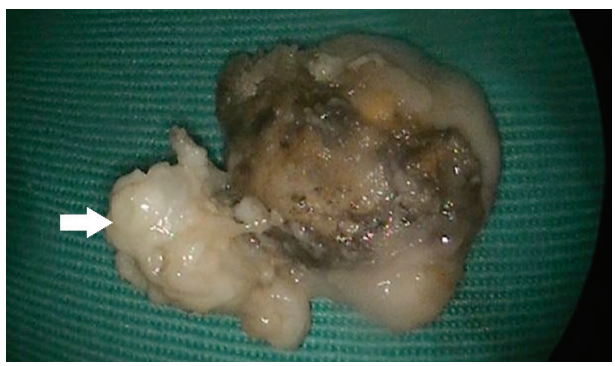

Fig. 3. Macroscopic view of the intranasal pleomorphic adenoma (tumor content marked with arrow).

Endonasal endoscopic complete tumor excision was performed under general anesthesia, during which some spillage of the tumor occurred. Macroscopically, it was a well-circumscribed, round, soft tumor, mostly gelatinous with homogeneous whitish-yellowish content and thin capsule (Fig. 3). The histopathologist made a definitive diagnosis of pleomorphic adenoma of minor salivary glands (Fig. 4).

The patient was discharged from the hospital one day after surgery without any complications. The patient was followed up on regular basis and

had no tumor recurrence at 5.5-year postoperative follow up (she was operated on in December 2016) (Fig. 5).

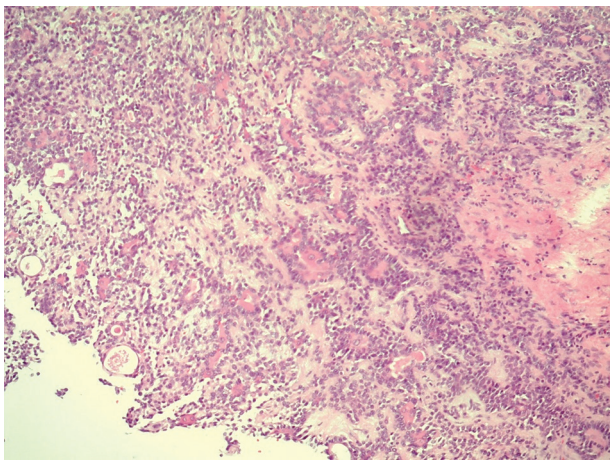

Fig. 4. Tumor histology: pleomorphic adenoma, a mixture of ductal epithelial cells, basal and myoepithelial cells, and variable amounts of stroma, both hyaline and chondromyxoid (hematoxylin-eosin, X100).

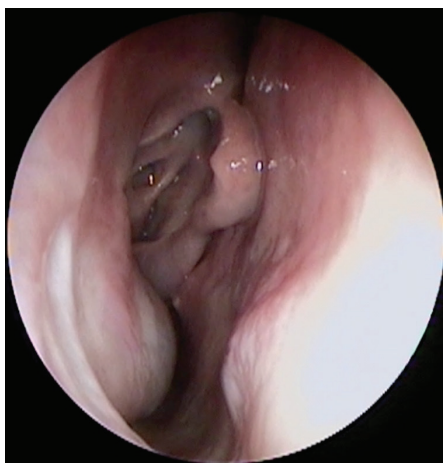

Fig. 5. Nasal endoscopy: postoperative follow up at 5.5 years (no tumor recurrence).

Discussion

Pleomorphic adenoma of the minor salivary glands can be found in the oral cavity (hard palate, soft palate, superior lip), ear, external nose and nasal cavity, paranasal sinuses, nasopharynx, lower eyelid, larynx, trachea, pterygopalatine fossa, etc.[1,2]. However, they are extremely rare in the nasal cavity[1,4].

The mean age of patients with intranasal PA is 45.4 years[1]; however, our patient was much older, 72 years of age. The most frequent symptom in these patients at presentation is unilateral nasal obstruction, as in our case, but they can also present with epistaxis, nasal mass, cheek or nasal swelling, external deformity, mucopurulent rhinorrhea, and epiphora[1-4].

Intranasal PAs are most often localized on the nasal septum, and according to Rha, it is found in 57.4% of all patients[1-3]. The lateral nasal wall is a possible localization, usually on the inferior turbinate[1,2]. In the literature available on PubMed database, we found no case of intranasal PA localized on the lateral nasal wall in front of the inferior turbinate, as in the case presented.

The natural history of intranasal PA is slow growth with conformity of the adjacent anatomy, as also seen on our patient's CT scan[1,3,6]. Endonasal endoscopic complete resection of the tumor was done. The reported case is typical considering its size (25x18x12 mm) and broad base. The mean size of intranasal PA is usually 3.6 cm (range 0.5-10 cm) and most of them have a broad base[1,3]. Cross-sectionally, they are well-circumscribed, round or lobular nodular tumors surrounded by a fibrous capsule of varying thickness, firm consistency, or in some cases, as in ours, are soft and myxoid (gelatinous)[1,5].

In postoperative follow up, the main problems with PA are the possibility of recurrence, malignant transformation, and metastasizing. The reported recurrence rate of intranasal PA (≤10%) is lower than that recorded in parotid PA[1,3].

During surgery, we had some tumor spillage. Tumor disruption and spillage have been reported as variables with an independent effect on parotid PA recurrence of punctured tumors (26.9%). Recurrence has been reported in 80% of cases with spillage[6]. Despite these data, our patient is still without recurrence after 5.5 years of follow up.

Malignant transformation of PA has been reported, meaning carcinoma *ex* PA and malignant mixed tumor[1,2,4]. The diagnosis of benign metastasizing PA of the salivary gland is extremely rare and still under debate[1,4].

Postoperative long-term follow up is necessary in all patients[1,2], so we will continue to follow up our patient.

Conclusion

Intranasal PA arising from the lateral nasal wall in front of the inferior turbinate is extremely rare, so the case presented may be the first ever published. Com-

plete surgical excision is the treatment of choice, if possible, by means of endoscopic endonasal surgery, paying special attention not to cause tumor spillage and puncture. The patients need long-term postoperative follow up.

Acknowledgment

The authors would like to thank Prof. Slobodan Mitrović, PhD, for his valuable assistance.

References

1. Rha MS, Jeong S, Cho HJ, Yoon JH, Kim CH. Sinonasal pleomorphic adenoma: a single institution case series combined with a comprehensive review of literature. Auris Nasus Larynx. 2019 Apr;46(2):223-9. DOI: 10.1016/j.anl.2018.08.003
2. Kuan EC, Diaz MF, Chiu AG, Bergsneider M, Wang MB, Suh JD. Sinonasal and skull base pleomorphic adenoma: a case series and literature review. Int Forum Allergy Rhinol. 2015 May;5(5):460-8. DOI: 10.1002/alr.21500
3. Compagno J, Wong RT. Intranasal mixed tumors (pleomorphic adenomas): a clinicopathologic study of 40 cases. Am J Clin Pathol. 1977 Aug;68(2):213-8. DOI: 10.1093/ajcp/68.2.213
4. Karligkiotis A, Bozkurt G, Pietrobon G, Battaglia P, Turri-Zanoni M, Chu F, Ansarin M, Ottini G, De Luca M, Uccella S, Castelnuovo P. Endoscopic endonasal resection of sinonasal and nasopharyngeal pleomorphic adenomas: a case series. Turk Arch Otorhinolaryngol. 2020 Sep;58(3):186-92. DOI: 10.5152/tao.2020.5382
5. Baron S, Koka V, El Chater P, Cucherousset J, Paoli C. Pleomorphic adenoma of the nasal septum. Eur Ann Otorhinolaryngol Head Neck Dis. 2014 Apr;131(2):139-41. DOI: 10.1016/j.anorl.2013.03.007
6. Riad MA, Abdel-Rahman H, Ezzat WF, Adly A, Dessouky O, Shehata M. Variables related to recurrence of pleomorphic adenomas: outcome of parotid surgery in 182 cases. Laryngoscope. 2011 Jul;121(7):1467-72. DOI: 10.1002/lary.21830

Sažetak

INTRANAZALNI PLEOMORFNI ADENOM PODRIJETLOM IZ LATERALNE STIJENKE NOSA

Lj. Jovančević, A. Fejsa Levakov i *B. Jovančević*

Pleomorfni adenom je veoma rijedak u sinonazalnoj regiji, pričem je najučestalija lokalizacija septum, zatim lateralna stijenka nosa. U prikazanom slučaju bolesnica u dobi od 72 godine se žalila na unilateralnu začepljenost nosa, bez drugih simptoma. Zapušenost nosa joj se javila godinu dana prije pregleda i progresivno se pogoršavala. Prednjom rinoskopijom joj je u nosu desno uočena izraslina prekrivena sluznicom, glatke površine, polipoidna, meke konzistencije, blijedo sivkasto-ružičaste boje, veličine oko 2x2 cm. Endoskopija nosa je utvrdila da je polazište izrasline široko i da se nalazi na lateralnoj stijenci nosa. Kompjutoriziranom tomografijom je utvrđena homogena, solidna, mekotkivna masa veličine 25x18x12 mm spojena na lateralnu stijenku nosa, odmah iza vestibula. a neposredno ispred donje nosne školjke. Načinjena je endonazalna endoskopska kompletna ekscizija tumora tijekom koje je došlo do djelomičnog istjecanja sadržaja tumora. Patohistološka dijagnoza je bila pleomorfni adenom malih žlijezda slinovnica. Bolesnica je redovito praćena i kontrolirana, nije bilo recidiva tumora u šestoj poslijeoperacijskoj godini. Intranazalni pleomorfni adenom s polazištem na lateralnoj stijenci nosa ispred donje nosne školjke je iznimno rijedak pa je prikazani slučaj vjerojatno prvi ikada publiciran.

Ključne riječi: *Pleomorfni adenom; Neoplazme nosa; Endoskopska endonazalna kirurgija*