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OLGU BİLDİRİSİ / CASE REPORT

A case of pleomorphic adenoma of the inferior turbinate

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Alt konka yerleşimli bir pleomorfik adenom olgusu

Pleomorfik adenom majör tükürük bezlerinden kaynaklanan en sık tümördür. Boyunda, kulakta, mediastende ve burun dış kısmında görülmesine rağmen, bu tümörlerin intranazal lokalizasyonu çok nadir olup, çoğunlukla nazal septum kaynaklıdır. Lezyon nereden kaynaklanırsa kaynaklansın, tedavideki ana yaklaşım cerrahidir. Bu makalede, 4 yıldır sol nazal kavitede obstrüksiyon şikayeti olan 33 yaşındaki kadın hasta sunuldu. Endoskopik nazal kavite değerlendirilmesinde, sol nazal kavitenin sol alt konkadan kaynaklanan kitlesel lezyon ile oblitere olduğu izlendi. Lezyonun ekspansif yapısından dolayı sol alar insizyon uygulandı. Cerrahi sonrası 18 aylık takibi yapıldı. Literatüre bu tip neoplazmların yerleşim yerleri açısından dikkatlice göz atıldığında, alt konka yerleşimi pek alışılmamış olduğundan bu olguyu sunmayı uygun bulduk.

Anahtar Sözcükler: Pleomorfik adenom, inferior nazal konka.

Abstract

Pleomorphic adenoma is the most frequent tumor of the major salivary glands. Although it has also been reported to be present in the neck, ear, mediastinum, external nose and nasal cavity. Intranasal localization of this lesion is very rare and mainly originates from the nasal septum. From wherever the lesion originates, the main treatment modality should be surgical. In this article, we present a 33 year-old woman with a 4-year history of left-sided nasal obstruction. Endoscopic examination of the left nasal cavity showed that the mass seemed to originate from left inferior turbinate and protrude to the left nasal cavity. Due to the expansile nature of the lesion, a left alar incision was performed. She was followed-up after surgery for 18 months. A careful review of the literature has revealed that this is a highly unusual site for such neoplasms. For this reason, we approved to present this case.

Key Words: Pleomorphic adenoma, inferior nasal turbinate.

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Introduction

Pleomorphic adenoma is the most frequent form of benign tumor in the major and minor salivary glands, but rare in the nasal cavity, pharynx, larynx, trachea and lacrimal gland.[1-3] These tumors arise most commonly in the major salivary glands; the minor salivary glands are affected in only 8% of the cases.[4]

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Pleomorphic adenoma detected in nasal cavity is extremely rare. Furthermore, many of the reported intranasal pleomorphic adenomas are originated from the nasal septum.[5] Inferior nasal turbinate is a highly unusual for pleomorphic adenoma. Review of the literature show that pleomorphic adenoma mainly arises from the mucous membrane of the nasal septum, although most of the minor salivary and serous gland are located in the lateral nasal wall. Pleomorphic adenoma of the nasal cavity tends to present with symptoms of unilateral nasal obstruction and epistaxis. Other presenting complaints include nasal swelling, epiphora and mucopurulent rhinorrhea. The diagnosis of pleomorphic adenoma of the nasal cavity is difficult because of its frequent coincidence with the nasal polyps or septum deviation.

The potential risk of malignancy transformation of the pleomorphic adenoma is not very high and estimated amount is about 6%.[5,6] But the risk of malignancy transformation is increased with the delayed diagnosis of the tumor. Treatment of intranasal pleomorphic adenomas consists of surgical excision, with wide margins when feasible, to avoid recurrence.

In this study, we present a rare case of an intranasal pleomorphic adenoma originated from the left inferior nasal turbinate. A careful review of the literature has revealed that this is a highly unusual site for such neoplasms. For this reason, we approved to present this case.

Case Report

A 33-year-old woman who had complained of left-sided nasal obstruction for 4 years was referred to the our ENT Department. She has been previously went to doctor one-year ago. The operation was recommended by the doctor but she ignored the recommendation. Her complaints were not relieved and they increased. Anterior rhinoscopic examination of the patient revealed a vascularized mass obstructing on the left nasal cavity. There was an elastic hard, oval and mucosa-covered mass (Figure 1). No abnormal finding was noted in the other ENT areas. Endoscopic examination of the left nasal cavitiy showed that the mass seemed to originate from lateral nasal wall and protrude to the left nasal cavity. The mass has been expanded nasomaxiller and nasolabial sulcus. Paranasal computed tomographic evaluations revealed a well circumscribed solid and expansile polypoid lesion which was originated from the left inferior turbinate, and obstructed left nasal cavity. The size of the lesion was 28 mm x 21 mm x 20 mm. The lesion displaced the lateral nasal wall laterally and did not show any evidence of destruction of the bony structure of the left maxillary sinus (Figure 2). Later, punch biopsy was made. Histopathological examination revealed that fibromyxoid stroma and the epithelial component composed of monotonous round cells was of ductal

Figure 1. Anterior rhinoscopic appearence of the lesion. [Color figure can be viewed in the online issue, which is available at www.turkarchotolaryngol.org]

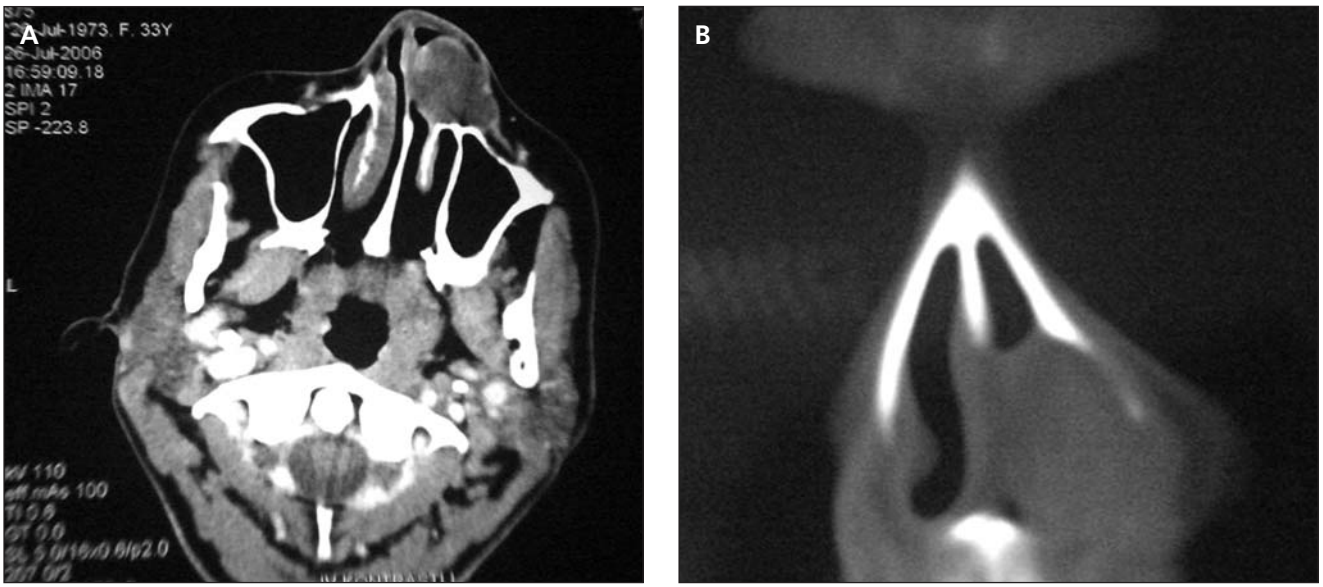

Figure 2. Axial (**A**) and coronal (**B**) computed tomographic images of the lesion.

nature. Tumoral cells did not show cellular atypia. According to histopathological examination, the lesion was reported as pleomorphic adenoma. Due to expansile nature of the lesion, left alar incision was performed under general anesthesia. Intraoperatively, it was detected that the mass was originated from the left inferior turbinate. The mass did not extend into the left maxillary sinus. The mass was removed totaly with partial excision of the inferior turbinate (Figure 3). The histopathological examination of the specimen confirmed the preoperative diagnosis (Figure 4).

Figure 3. Surgical specimen of the lesion. [Color figure can be viewed in the online issue, which is available at www.turkarchotolaryngol.org]

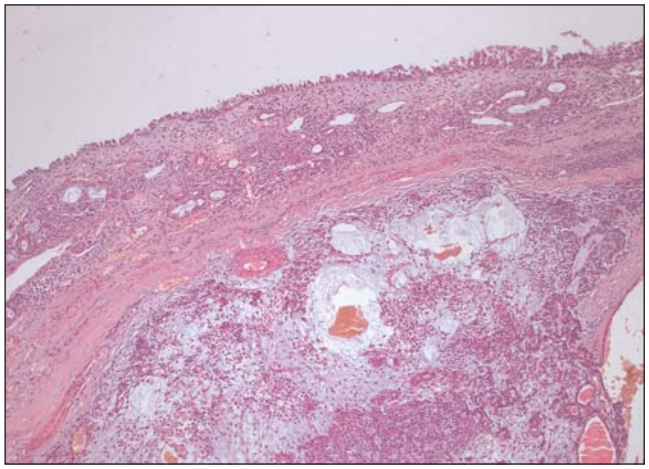

Figure 4. Histopathologic appearance of the lesion (**HE x10**). [Color figure can be viewed in the online issue, which is available at www.turkarchotolaryngol.org]

The patient was discharged without any complication 3 days after the surgery and informed about the risk of recurrence. She was followed-up after surgery for 18 months. The symptoms were resolved and no evidence of recurrence was observed (Figure 5).

Figure 5. The endoscopic view of the patient after 18-month follow-up period. [Color figure can be viewed in the online issue, which is available at www.turkarchotolaryngol.org]

Discussion

The vast majority of pleomorphic adenoma arises in the major salivary glands. Those occurring in the minor salivary glands usually arise in the hard and/or soft palate, although other sites, including the nasal cavity, external auditory canal, larynx, pharynx, trachea, lacrimal glands, the capsule of thyroid and parathyroid glands, sternoclavicular joint, mandibula and cervical soft tissues have rarely been reported.[5,7-9]

Cases of pleomorphic adenoma in the nasal cavity have been described by Compagno and Wong,[10] Suzuki et al.[2] and Wakami et al.[11] This type of tumor generally originates from the septal mucosa even though the seromucosal glands are mostly located in the lateral nasal wall. The most common site of origin was the bony cartilaginous septum.[12] In Polish otolaryngological literature, 8 cases of pleomorphic adenoma located in the nasal cavity have been described. In 5 cases, tumor arose from the nasal septum; in the remaining 3 cases, from the lateral nasal wall.[13] Ozturk et al.,[14] Castello et al.[15] and Clark et al.[16] have described a case of pleomorphic adenoma located in nasal cavity arose from the nasal septum, lateral nasal wall and middle turbinate, respectively. All case reports located in nasal cavity have been shown a Table 1.

As previosly stated, a careful review of the literature has revealed that inferior turbinate is a highly unusual site for such neoplasms. Baraka et al. presented a case of pleomorphic adenoma, originating from the anterior edge of the inferior turbinate, measuring 1-1.5 cm and obstructing the right nasal cavity. Only two cases have been reported in English literature. Ünlü et al. presented a case of pleomorphic adenoma, originated from the middle 1/3 of the right inferior turbinate and about 3.5 cm in diamater.[5,17,18]

Pleomorphic adenoma of the nasal cavity tends to present with symptoms of unilateral nasal obstruction or epistaxis. In Compagno, Suzuki and Wakami's series, the most frequent complaints were nasal obstruction and epistaxis in 70% and 60% respectively. Other existing complaints include nasal swelling, epiphora and mucopurulant rhinorrhea. Locoregional and distant metastases have been described in the literature.[2,7,10,11,15]

Nasal endoscopy typically demonstrates a unilateral polypoidal lesion that bleeds easily. A computed tomography scan demonstrates a smooth, localized lesion entire nasal cavity causing smooth bony expansion, suggesting a slow, benign process,

Table 1. Pleomorphic adenoma cases located intranasal cavity.

Author name	Publishing date	Age (years)	Gender	Side of lesion	Intranasal localization
Compagno and Wong[10]	1977	16-74	Female predominance	No difference	25/40 nasal septum, 8/40 lateral nasal wall, others not found
Baraka et al.[18]	1984	No full text available	No full text available	Right	Anterior edge of the inferior turbinate
Suzuki et al.[2]	1990	43	Woman	Right	Lateral nasal wall
Castello et al.[15]	1996	45	Man	Right	Lateral nasal wall
Wakami et al.[11]	1996	21 51	Woman Man	Right Left	Nasal septum Lateral nasal wall
Clark et al.[16]	1999	41	Woman	Right	Middle turbinate
Ünlü et al.[5]	2003	39	Woman	Right	Inferior turbinate
Tahlan et al.[4]	2004	55	Man	Right	Nasal septum
Mackle et al.[7]	2004	31	Man	Right	Nasal septum
Kumagai et al.[1]	2004	32	Woman	Left	Nasal septum
Narozny et al.[13]	2005	21-69	5 men, 3 women	Left	5/8 nasal septum, 3/8 lateral nasal wall
Öztürk et al.[14]	2008	65	Woman	Right	Nasal septum

rather than the destructive bony pattern associated with malignancy.[12]

The diagnosis of pleomorphic adenoma of the nasal cavity is difficult because of its frequent coincidence with the nasal polyps or septum deviation. Computed tomography, which showed the site of tumor's origin, its extension and its penetration into surrounding structures, is very helpful.[13]

Treatment of intranasal pleomorphic adenomas consists of surgical excision, with wide margins when feasible, to avoid recurrence. The surgical approach includes endoscopic removal, lateral rhinotomy and even facial degloving, depending on the location of the tumor.[15] However, some intranasal benign tumors, which were successfully treated by endoscopic transnasal surgery have been reported in the literature.[12,19,20] In our case, the size of the tumor was not suitable for endoscopic resection. So due to the expansile nature and tendency of the tumor to bleed and to recur, we preferred left alar incision. This surgical technique has an advantage of wide exposure and direct approach to the nasal cavity. And this type of approach provided good local intraoperative control. But this technique has a cosmetic disadvantage to other approaches.

Clinical follow-up is important in view of the potential for recurrence, malignant transformation.[21,22] In Compagno, Suzuki and Wakami' s series, the rate of malignancy transformation was 2.5-10% and had a female predominance. Compagno and Wong applied surgical treatment to 40 intranasal pleomorphic adenoma cases and followed them for a 7.5-year period. The rate of recurrence was reported 5%.[2,10,11] Due to the nonaggressive nature of the tumor, its total excision may result in a lower risk of recurrence. It is well known that insufficient resection will cause a recurrence. The recurrence rate of

pleomorphic adenoma depends almost on the adequacy of the primary excision. Most of these recurrence will appear during the first 18 months after surgery, but it is possible to re-emerge over an exceedingly long period (50-years or more).[22] In our reported case, there was no evidence of recurrence after 18 months postoperatively.

References

1. **Kumagai M, Endo S, Koizumi F, Kida A, Yamamoto M.** A case of pleomorphic adenoma of the nasal septum. *Auris Nasus Larynx* 2004;31:439-42.
2. **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. *Nippon Jibiinkoka Gakkai Kaiho* 1990;93:740-5.
3. **Kumar SS, Reddy PS, Prabhakar G.** Pleomorphic adenoma of the nasal septum–a case report. *Indian J Pathol Microbiol* 2004;47: 397-8.
4. **Tahlan A, Nanda A, Nagarkar N, Bansal S.** Pleomorphic adenoma of the nasal septum: a case report. *Am J Otolaryngol* 2004;25: 118-20.
5. **Ünlü H, Çelik O, Demir MA, Eskiizmir G.** Pleomorphic adenoma originated from the inferior nasal turbinate. *Auris Nasus Larynx* 2003;30:417-20.
6. **Shaheen OH.** Benign salivary gland tumors, In: Kerr AG, editor. Scott Brown's Otolaryngology. 6th ed. London: Butterworth Heinemann; 1997. pp. 5/20/1-18.
7. **Mackle T, Zahirovic A, Walsh M.** Pleomorphic adenoma of the nasal septum. *Ann Otol Rhinol Laryngol* 2004;113:210-1.
8. **Suzuki K, Baba S, Miyamoto N.** Pleomorphic adenoma of the external auditory canal in Japan, with a case report. *Auris Nasus Larynx* 1991;18:271-9.
9. **Ferlito A, Baldan M, Andretta M, Blandamura S, Pesavento G, Piazza M.** Implantation of parotid pleomorphic adenoma in the upper neck. *ORL J Otorhinolaryngol Relat Spec* 1991;53:165-76.
10. **Compagno J, Wong RT.** Intranasal mixed tumors (pleomorphic adenomas): a clinicopathologic study of 40 cases. *Am J Clin Pathol* 1977;68:213-8.
11. **Wakami S, Muraoka M, Nakai Y.** Two cases of pleomorphic adenoma of the nasal cavity. *Nippon Jibiinkoka Gakkai Kaiho* 1996;99: 38-45.
12. **Yiotakis I, Dinopoulou D, Ferekidis E, Manolopoulos L, Adamopoulos G.** Pleomorphic adenoma of the nose. *Rhinology* 2001;39:55-7.
13. **Narozny W, Kuczkowski J, Mikaszewski B.** Pleomorphic adenoma of the nasal cavity: clinical analysis of 8 cases. *Am J Otolaryngol* 2005;26:218.
14. **Öztürk E, Sağlam O, Sönmez G, Cüce F, Haholu A.** CT and MRI of an unusual intranasal mass: pleomorphic adenoma. *Diagn Interv Radiol* 2008;14:186-8.
15. **Castello E, Caligo G, Pallestrini EA.** Case report: pleomorphic adenoma of the lateral nasal wall. *Acta Otorhinolaryngol Ital* 1996; 16:433-7.
16. **Clark M, Fatterpekar GM, Mukherji SK, Buenting J.** CT of intranasal pleomorphic adenoma. *Neuroradiology* 1999;41:591-3.
17. **Carriero E, Almadori G, Cadoni G.** Pleomorphic adenoma of the lower turbinates: clinical case and review of literature. *Acta Otorhinolaryngol Ital* 1997;17:377-81.
18. **Baraka ME, Sadek SA, Salem MH.** Pleomorphic adenoma of the inferior turbinate. [Abstract] *J Laryngol Otol* 1984;98:925-8.
19. **Ünlü HH, Ünlü Z, Ayhan S, Eğrilmez M.** Osteochondroma of the posterior nasal septum managed by endoscopic transnasal transseptal approach. *J Laryngol Otol* 2002;116:955-7.
20. **Cho KJ, el-Naggar AK, Mahanupab P, Luna MA, Batsakis JG.** Carcinoma ex-pleomorphic adenoma of the nasal cavity: a report of two cases. *J Laryngol Otol* 1995;109:677-9.
21. **Freeman SB, Kennedy KS, Parker GS, Tatum SA.** Metastasizing pleomorphic adenoma of the nasal septum. *Arch Otolaryngol Head Neck Surg* 1990;116:1331-3.
22. **Rosai J.** Skin. In: Rosai J, editor. Ackerman's surgical pathology. 8th ed. St Louis: Mosby Year Book; 1996. p. 820-1.

Conflict of interest statement:
No conflicts declared.

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