Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Otolaryngology

Soft Palate Pleomorphic Adenoma of a Minor Salivary Gland: An Unusual Presentation

Mkheittirate Yaacoub[1*], Arioua Abdelilah[1], Afellah Mohamed[1], Oudidi Abdelatif[1], Naouar Ouattassi[1], Mohamed Ridal[1], Najib Benmansour[1], Hammas Nawal[2], Mohammed Nouredine EL Alami El Amine[1]

[1]Department of Otolaryngology and Cervicofacial Surgery; Hassan II University Hospital, Fes, Morocco
[1]Anatomy, Surgery and Anesthesiology Laboratory, University of Sidi Mohammed Ben Abdellah, Fez, Morocco
[2]Anatomopathology Laboratory; Hassan II University Hospital Center – Fes, Morocco

DOI: 10.36347/sjmcr.2024.v12i05.049 | **Received:** 12.04.2024 | **Accepted:** 15.05.2024 | **Published:** 17.05.2024

***Corresponding author:** Mkheittirate Yaacoub
Department of Otolaryngology and Cervicofacial Surgery; Hassan II University Hospital, Fes, Morocco

Abstract **Case Report**

Tumors of salivary glands are less frequent in the head and neck region. Pleomorphic adenoma (PA) is a benign tumor of major and accessory salivary glands. The soft palate is one of the most frequent sites among accessory salivary glands. This tumor can increase in size and cause oropharyngeal obstruction with a risk of asphyxia, endangering the patient's life. Treatment is based on complete tumor excision and it's sometimes difficult. Histopathological examination confirms the diagnosis. In this study, we present a case of a large pleomorphic adenoma of the accessory salivary glands located in the soft palate, successfully treated with intraoral excision surgery after an initial tracheostomy.

Keywords: Pleomorphic Adenoma; Oral Surgery; Salivary glands; Tracheostomy; Soft palate; MRI.

INTRODUCTION

Tumors of the salivary glands represent 3% of head and neck tumors [1]. Pleomorphic adenoma (PA) is the most common tumor of the salivary glands, accounting for approximately 40% to 70% of all tumors of the major and accessory salivary glands [2]. PA is a benign mixed tumor composed of epithelial and mesenchymal cells arranged in various morphological patterns, surrounded by fibrous tissue [3]. The parotid gland is the most commonly affected site. Among accessory salivary glands, the palate is the most frequently involved site, accounting for nearly 60% of cases. Lips, cheeks, and gums are relatively rare sites [4]. Treatment is based on complete tumor excision. Histopathological examination confirms the diagnosis [5, 6].

In this study, we present a case of a large pleomorphic adenoma of the accessory salivary glands located in the soft palate, successfully treated with intraoral excision surgery.

CASE REPORT

Our case involves a 62-year-old patient with no significant medical history, who presented with the appearance of a left intraoral mass evolving over 4 years. This swelling is painless, gradually increasing in size, associated with solid dysphagia, rhinolalia, and increased respiratory discomfort with exertion, prompting the patient to seek consultation at our facility.

Clinically, the patient is in good general condition and eupneic at rest. On intraoral examination, a mass with healthy mucosa, firm consistency, irreducible, measuring approximately 6cm in its longest axis, is found, pushing back the soft palate and the left tonsil. It protrudes into the oropharyngeal space (Figure 1). Cervical palpation reveals no satellite lymphadenopathy. The rest of the clinical examination is unremarkable.

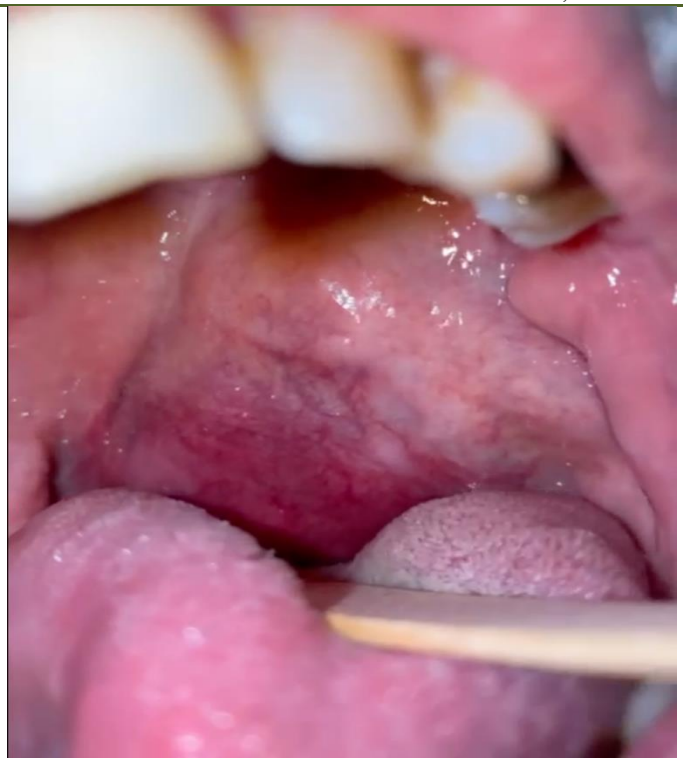

Figure 1: Mass pushing back the soft palate and the left tonsillar fossa, with narrowing of the oropharyngeal space

A cervicofacial CT scan was performed, revealing a large tumor mass measuring 76×71×55mm, with heterogeneous enhancement after contrast administration. It appears to arise from the palate and the left tonsil, with extension into the nasopharynx and the homolateral choana, causing significant narrowing of the oropharyngeal lumen. This tumor also shows close relationships with the left vascular axis. There is no evidence of bone lysis of the palate or the pterygoid processes (Figure 2: Image A and B).

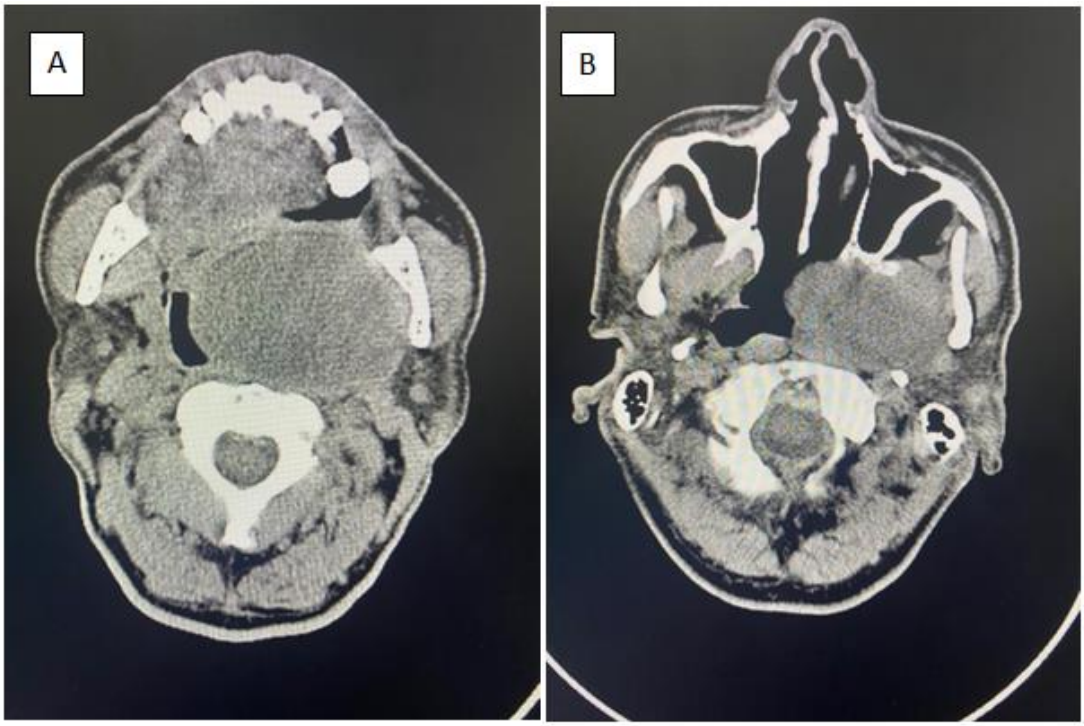

Figure 2: Axial facial CT scan: A- Well-defined large tissue mass with narrowing of the oropharyngeal space. B- Extension of the palatal process into the nasopharynx and the left choana

A cervicofacial MRI was performed to better characterize the relationships of the mass. It revealed a well-defined left lateral pharyngeal tumor process, with irregular contours in some areas described as isosignal on T1-weighted images, heterogeneous hyperintensity on T2-weighted images, non-restrictive diffusion, enhancement after contrast administration, unchanged on fat saturation sequences, measuring 74x46x76 (Transverse × Anteroposterior × Height). It exerts significant mass effect on the oro- and hypopharynx, nearly completely filling the pharyngeal lumen. It comes into contact with the left choana and infiltrates the palatoglossus muscle. It approaches the ascending branch of the mandible in the masticator space as well as the medial extension of the parotid gland, without signs of invasion. The mass also contacts the medial and lateral pterygoid muscles, without signs of invasion. The signal characteristics of the mass are consistent with the diagnosis of a pleomorphic adenoma Figure 3 (Image A and B).

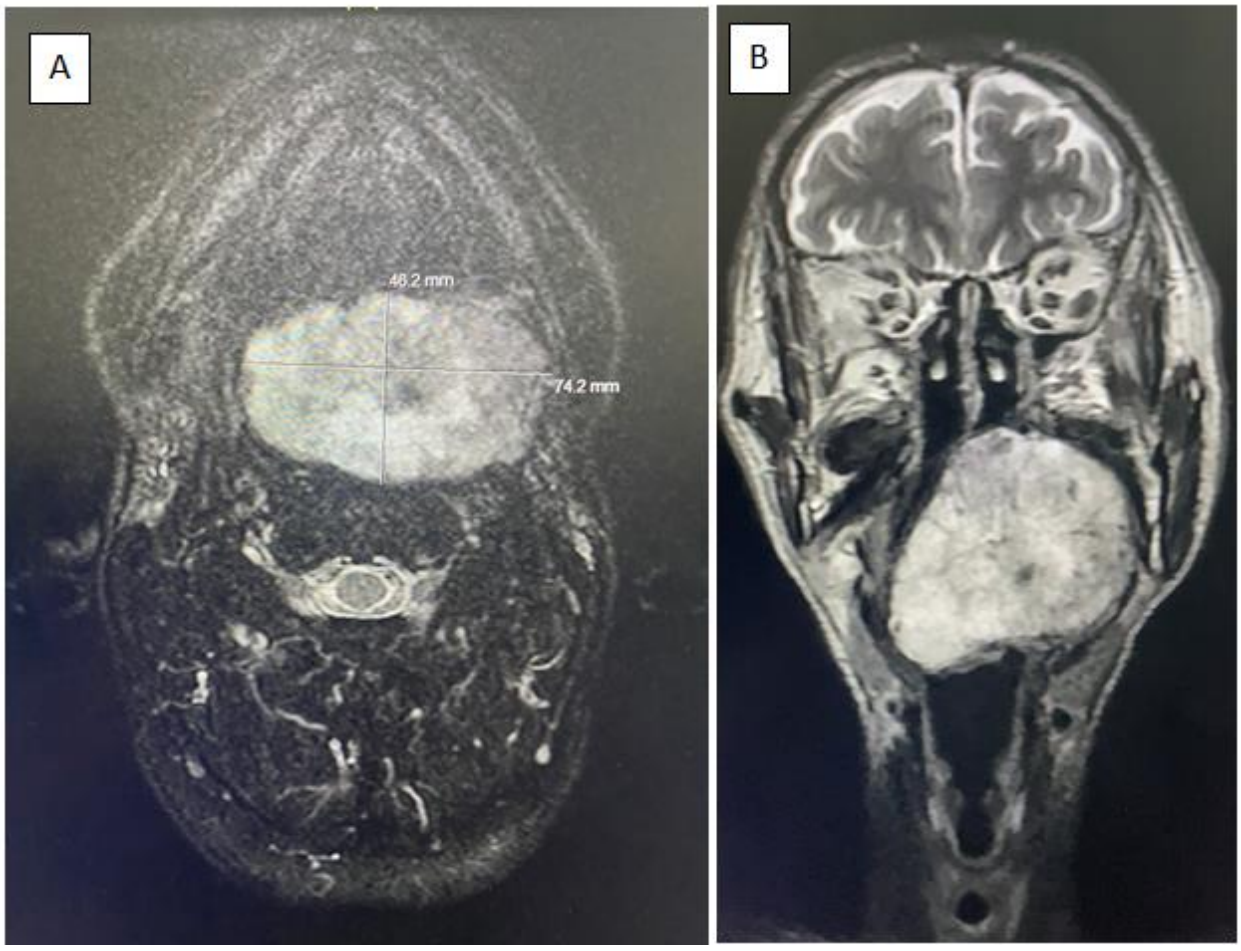

Figure 3: Facial MRI: A- T1-weighted sequence: Well-defined left lateral-pharyngeal tissue process with irregular and homogeneous contrast enhancement. B- T2-weighted sequence: Significant mass effect on the oropharynx, with near-total filling of the pharyngeal lumen

A biopsy of the mass was performed after establishing a tracheotomy due to the significant narrowing of the oropharyngeal caliber by the tumor. The histopathological results confirmed a pleomorphic adenoma.

Surgery was performed via an intraoral approach, allowing for a peri-capsular en bloc excision of the tumor, with placement of a nasogastric tube for feeding while ensuring proper healing of the surgical site (Figure 4 & 5).

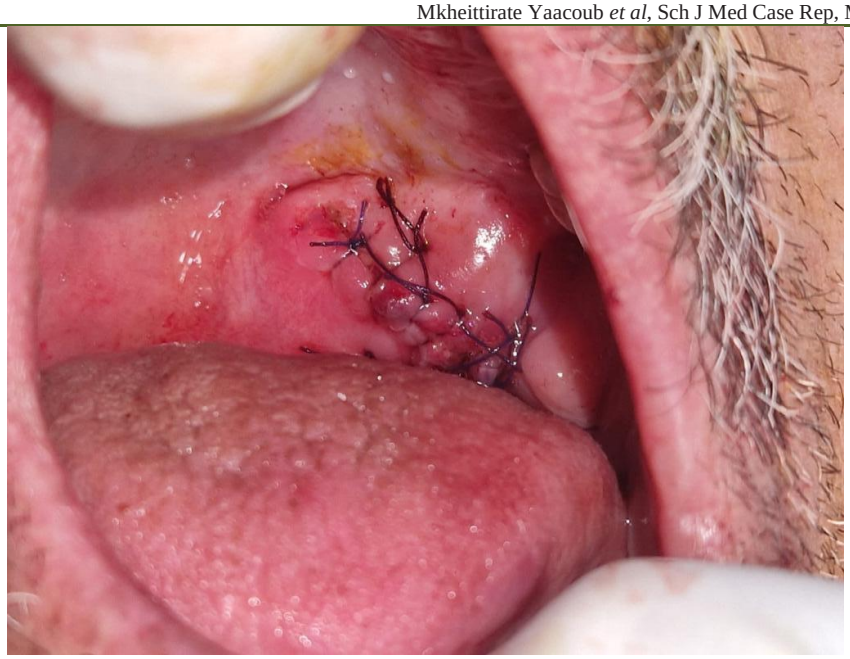

Figure 4: Intraoral approach

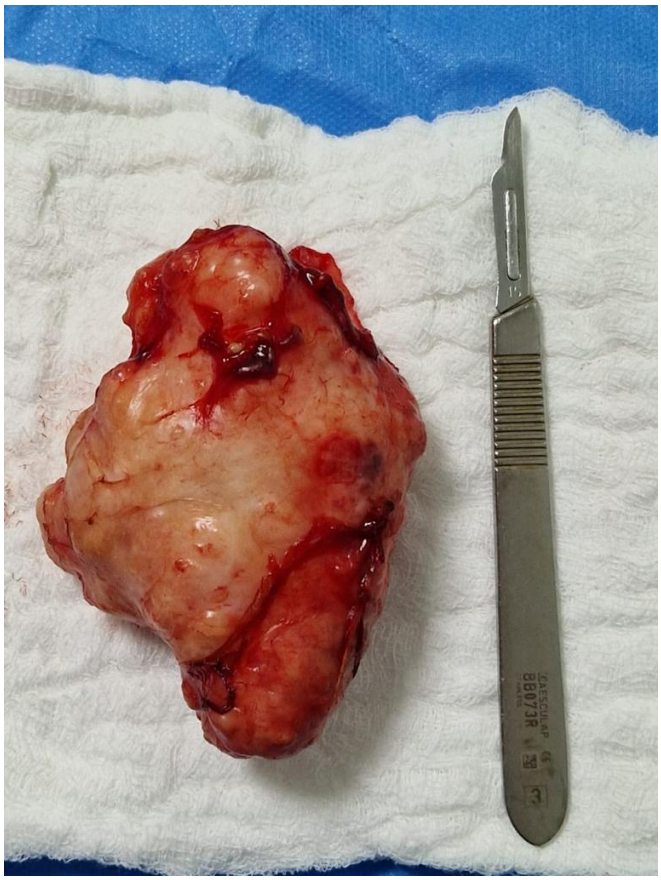

Figure 5: Surgical specimen

Postoperative recovery was uneventful, with removal of the nasogastric tube after 7 days, decannulation performed 10 days after the procedure, with gradual closure of the tracheotomy orifice. There was no recurrence observed during an 18-month follow-up period (Figure 6).

Final histopathological examination concluded a pleomorphic adenoma without signs of malignancy (Figure 7).

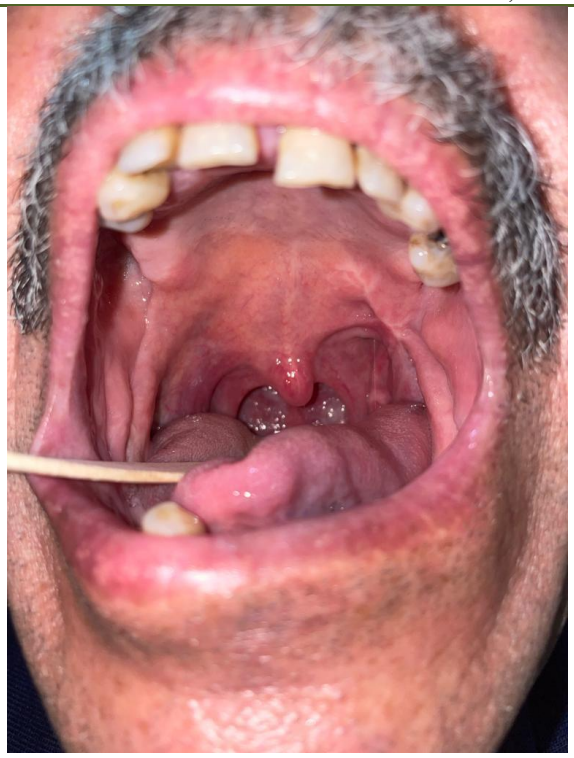

Figure 6: Postoperative assessment after 12 months

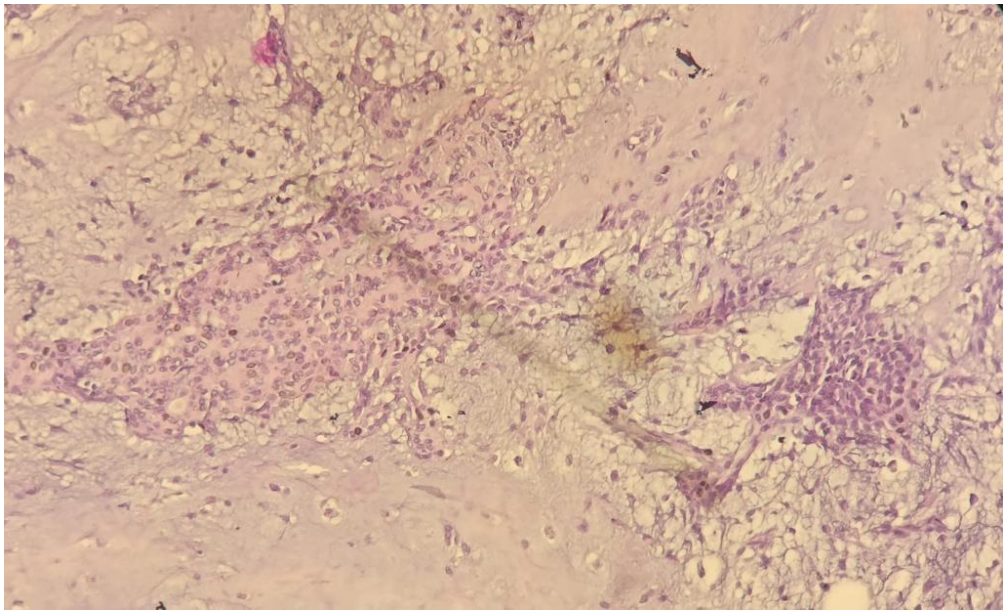

Figure 7: Tumor proliferation consisting of a triple component: epithelial, myoepithelial, and chondroid (H&E stain, 200x magnification)

DISCUSSION

Tumors of accessory salivary glands represent 15% to 20% of salivary gland tumors [7, 8]. Pleomorphic adenoma is the most common tumor (50%) of both major and accessory salivary glands [9]. Approximately 80% of pleomorphic adenomas develop in the parotid gland, 8% in the submandibular gland, and 7% in accessory salivary glands. It represents the most frequent histological type (70.6 to 100%) of benign tumors of accessory salivary glands with a predilection for the palate [10].

Pleomorphic adenomas of accessory salivary glands more often affect women than men, with varying ratios in different studies, ranging from 1/1.1 [1] to 1/3.2 [11]. The preferred age of onset for pleomorphic adenoma varies between 30 and 40 years [12]. Furthermore, our case involves a male patient with a late discovery at the age of 62.

These benign tumors grow slowly and are only discovered when they become large. Clinical symptoms depend on the size and location of the tumor [13]. In the oral cavity, a painless swelling beneath healthy mucosa is often described [12-14]. Pleomorphic adenoma of the soft palate presents as a mass embedded in the half-arch. Sometimes, it deviates the uvula or pushes back the anterior pillar of the tonsil.

Pleomorphic adenoma of the palate often occurs postero-laterally, confined between the bony vault and the thick, healthy fibromucosa. In the case of a malignant tumor, it distorts the region into a "glass clock" shape.

Due to the risk of malignant transformation of accessory salivary gland tumors, appropriate preoperative diagnostic investigations are recommended. Von Stempel et al. [15] suggest that MRI is useful for imaging submucosal palatal lesions such as pleomorphic adenoma because they typically exhibit low signal intensity on T1 and high signal intensity on T2, often with a low signal fibrous capsule. MRI can evaluate perineural extension and provides excellent soft tissue definition. They also recommend craniofacial computed tomography (CT) to evaluate bone erosion, which could indicate a more aggressive malignant lesion. In our case, a CT-MRI couple was also performed before surgery due to the tumor's size and the need to identify its extensions and any major blood vessels, allowing for proper surgical approach planning.

Fine needle aspiration or a core biopsy often provides a rapid and reliable diagnosis for a trained cytopathologist. Sensitivity ranges from 73% to 93%, and specificity from 85% to 98% in different series [16].

Treatment is primarily surgical. Complete surgical excision with removal of the covering mucosa and coagulation of the tumor bed will provide a definitive diagnosis and allow for the treatment of this remarkable salivary gland tumor. This complete excision is made difficult, especially with large tumors, due to the absence of a capsule and infiltration of accessory salivary glands between muscle fibers and adipose tissue. It may require the use of reconstruction flaps in cases of significant mucosal excision [5, 17].

Macroscopically, the tumor is nodular, well circumscribed, and often encapsulated by a connective tissue sheath. It is typically whitish-gray, sometimes translucent on cut section. Its consistency varies, being firm, soft, or gelatinous. The pleomorphic nature refers to a rich architectural diversity contrasting with the monomorphism of the epithelial and myoepithelial cells composing it.

These cells are typically regular and cytologically "reassuring." An important diagnostic feature is the observation of a distinctive stroma, which typically appears myxoid, sometimes with cartilaginous or bony differentiation [18, 19]. Highly cellular areas are sometimes observed (referred to as "cellular pleomorphic adenoma"). A capsule is most often observed, except sometimes in pleomorphic adenomas with primarily mucoid stroma or in oral or nasal locations [20]. Capsular breaches, incomplete capsules, pseudopods, and satellite nodules are common [21], but they do not necessarily indicate malignancy. However, they complicate the assessment of tumor resection quality and seem to facilitate local recurrence.

The incidence of malignant transformation or carcinoma ex-pleomorphic adenoma (CXPA) is rare. It occurs in less than 7% of pleomorphic adenomas in accessory salivary glands, mainly located in the palate [22].

Its prognosis is generally good, but remains marked by a high risk of recurrence after surgery and carcinomatous degeneration, necessitating early surgical management and regular surveillance [23].

CONCLUSION

Pleomorphic adenoma is a heterogeneous benign tumor of the salivary glands. Its extra-parotid locations are rare [24]. Extra-parotid locations are mainly dominated by the submandibular gland and accessory salivary glands of the palate and lips. Each location is distinct in its clinical characteristics, diagnostic methods, and surgical management [25, 26]. Literature review confirms regarding this tumor, on one hand, its progressive and invasive nature in case of surgical abstention, and on the other hand, its tendency for locoregional recurrence. Complete excision is imperative [27].

Regular locoregional surveillance after excision should be systematic for several years [23].

REFERENCES

1. Eveson, J. W., & Cawson, R. A. (1985). Tumours of the minor (oropharyngeal) salivary glands: a demographic study of 336 cases. *Journal of Oral Pathology & Medicine*, *14*(6), 500-509. 10.1111/j.1600 0714.1985.tb00522.x.
2. Lopes, M. A., Kowalski, L. P., Cunha Santos, G., & De Almeida, O. P. (1999). A clinicopathologic study of 196 intraoral minor salivary gland tumours. *Journal of oral pathology & medicine*, *28*(6), 264-267. 10.1111/j.1600-0714.1999.tb02036.x.
3. Jorge, J., Pires, F. R., Alves, F. A., Perez, D. E. C., Kowalski, L. P., Lopes, M. A., & Almeida, O. P. (2002). Juvenile intraoral pleomorphic adenoma: report of five cases and review of the literature. *International journal of oral and maxillofacial surgery*, *31*(3), 273-275. 10.1054/ijom.2002.0206

4. Cohen, M. A. (1986). Pleomorphic adenoma of the cheek. *Int J Oral Maxillofac Surg*, 15, 777-779. 10.1016/S0300-9785(86)80123-5
5. Nouri, H., Raji, A., Elhattab, Y., Rochdi, Y., & M'barek, B. A. (2006). Adénome pléomorphe de la voûte palatine. À propos d'un cas. *Rev d'Oto rhinolaryngologie et de chirurgie cervico-faciale*, 302, 22-23.
6. Gassab, E., Berkaoui, A., Kedous, S., Korbi, A., Khairedoine, N., Harrathi, K., ... & Gassab, A. (2009). Adénome pléomorphe à localisation extra-parotidienne. *Journal Tunisien d'ORL et de Chirurgie Cervico-Faciale*, *22*, 36-37.
7. Varghese, B. T., Sebastian, P., Abraham, E. K., & Mathews, A. (2003). Pleomorphic adenoma of minor salivary gland in the parapharyngeal space. *World journal of surgical oncology*, *1*(1), 1-3.
8. Becelli, R., Frati, R., Cerulli, G., Perugini, M., Frati, A., & Lannetti, G. (2001). Pleomorphic adenoma of the minor salivary glands of the palate. *Journal of Experimental & Clinical Cancer Research: CR*, *20*(1), 25-28.
9. Clauser L, Mandrioli S, Dallera V, Sarti E, Galie M, Cavazzini L. Pleomorphic adenoma of the palate, J Craniofac Surg 2004;15(6):1026-9.
10. Toida, M., Shimokawa, K., Makita, H., Kato, K., Kobayashi, A., Kusunoki, Y., ... & Shibata, T. (2005). Intraoral minor salivary gland tumors: a clinicopathological study of 82 cases. *International journal of oral and maxillofacial surgery*, *34*(5), 528-532.
11. Isacsson, G., & Shear, M. (1983). Intraoral salivary gland tumors: a retrospective study of 201 cases. *Journal of Oral Pathology & Medicine*, *12*(1), 57-62.
12. Pires, F. R., Pringle, G. A., de Almeida, O. P., & Chen, S. Y. (2007). Intra-oral minor salivary gland tumors: a clinicopathological study of 546 cases. *Oral oncology*, *43*(5), 463-470.
13. Just, P. A., Miranda, L., Elouaret, Y., Meatchi, T., Hans, S., & Badoual, C. (2008, December). Classification des tumeurs des glandes salivaires. In *Annales d'Otolaryngologie et de Chirurgie Cervico-faciale* (Vol. 125, No. 6, pp. 331-340). Elsevier Masson.
14. Hakeem, A. H., Hazarika, B., Pradhan, S. A., & Kannan, R. (2009). Primary pleomorphic adenoma of minor salivary gland in the parapharyngeal space. *World journal of surgical oncology*, *7*, 1-4.
15. Von Stempel, C., Morley, S., Beale, T., & Otero, S. (2017). Imaging of palatal lumps. *Clinical radiology*, *72*(2), 97-107.
16. Baglin, A. C., & Wassef, M. (2007, November). Cytoponction des glandes salivaires: le pour et le contre. In *Annales de Pathologie* (Vol. 27, pp. 78-80). Elsevier Masson.
17. Verma, K., & Kapila, K. (2002). Role of fine needle aspiration cytology in diagnosis of pleomorphic adenomas. *Cytopathology*, *13*(2), 121-127.
18. Barnes, L., Eveson, J., Reichart, P., & Sidransky, D. (2005). World Health Organization classification of tumours. Pathology and genetics of head and neck tumours. Lyon: IARC Press.
19. Ellis, G. L., & Auclair, P. L. (1996). Atlas of tumor pathology. Third series, fascicle 17. Tumors of the salivary glands. Washington: Armed forces institute of pathology.
20. Uro-Coste, E. (2007, November). Particularités des tumeurs des glandes salivaires accessoires. In *Annales de Pathologie* (Vol. 27, pp. 76-78). Elsevier Masson.
21. Zbären, P., & Stauffer, E. (2007). Pleomorphic adenoma of the parotid gland: histopathologic analysis of the capsular characteristics of 218 tumors. *Head & Neck: Journal for the Sciences and Specialties of the Head and Neck*, *29*(8), 751-757.
22. Akan, H., Yildiz, L., & Ünal, R. (2008). Carcinoma ex pleomorphic adenoma of the minor salivary gland with pulmonary metastasis. *Diagnostic and Interventional Radiology*, *14*(1), 3-5.
23. Lowry, T. R., & Heichel, D. J. (2004). Pleomorphic adenoma of the hard palate. *Otolaryngology—Head and Neck Surgery*, *131*(5), 793-793.
24. Cwalina, P., Skorek, A., Narozny, W., & Stankiewicz, C. (2002). Pleomorphic adenoma of minor salivary glands. *Otolaryngologia Polska= The Polish Otolaryngology*, *56*(6), 737-740.
25. Boumdini, H., Lebbar, K., & Chaouir, S. (2003). Adénome pléomorphe de l'espace parapharyngé: à propos d'un cas. *Rev Med du Maghreb*, *112*, 5-9.
26. El Kohen, A., Essakalli, L., Kzadri, M., Sefiani, S., & Andaloussi, Z. S. (2007). Adénome pléomorphe de la base de langue. *Revue de Stomatologie et de Chirurgie Maxillo-faciale*, *108*(3), 215-217.
27. Baek, B. J., Lee, J. Y., Bae, C. H., Kang, E. G., & Cho, H. D. (2009). Pleomorphic adenoma of the nasal vestibule. *Otolaryngology—Head and Neck Surgery*, *141*(4), 535-536.