International Journal of Otolaryngology and Head & Neck Surgery, 2024, 13, 251-257
https://www.scirp.org/journal/ijohns
ISSN Online: 2168-5460
ISSN Print: 2168-5452

Pleomorphic Adenoma of the Salivary Glands: Two Atypical Locations

Sidibé Youssouf[1,2], Diarra Abraham[1,3,4], Samaké Djibril[4,5*], Sanogo Boubacar[2,6], Traoré Abdoul Moumine[2,6], Diarra Kassim[4,6], Coulibaly Bourama[7], Diakité Youssouf[8], Keita Mohamed Amadou[2,6]

[1]Department of ENT and Head and Neck Surgery Mother-Child University Hospital "Le Luxembourg", Bamako, Mali
[2]Faculty of Medicine and Odontostomatology, Bamako, Mali
[3]Department of ENT and Head and Neck Surgery, The Mali-Gavardo Hospital, Bamako, Mali
[4]Ministry of Higher Education and Scientific Research, Bamako, Mali
[5]ENT and Head and Neck Surgery Department of the Commune V Reference Health Center (CSRef CV), Bamako, Mali
[6]Department of ENT and Head and Neck Surgery at CHU Gabriel Touré, Bamako, Mali
[7]Department of Anatomy and Pathological Cytology, Gabriel Touré University Hospital Center, Bamko, Mali
[8]Department of Oral Surgery, Sikasso Regional Hospital, Sikasso, Mali
Email: *samakedjibi@yahoo.fr

How to cite this paper: Youssouf, S., Abraham, D., Djibril, S., Boubacar, S., Moumine, T.A., Kassim, D., Bourama, C., Youssouf, D. and Amadou, K.M. (2024) Pleomorphic Adenoma of the Salivary Glands: Two Atypical Locations. *International Journal of Otolaryngology and Head & Neck Surgery*, **13**, 251-257.
https://doi.org/10.4236/ijohns.2024.133022

Received: April 1, 2024
Accepted: May 28, 2024
Published: May 31, 2024

Abstract

Pleomorphic adenoma is the most common tumor of the salivary glands, accounting for approximately 50% of salivary gland lesions. It develops mainly in the salivary glands: parotid (80%), submaxillary (10%), sublingual (1%) and in the accessory oral-pharyngeal glands (9%). The aim of this work was to report 2 cases of pleomorphic adenoma of atypical location and then to discuss the difficulties linked to its diagnostic and therapeutic management in the Malian context. They were a 40-year-old man and a 72-year-old woman. They were admitted to the ENT department of the "Luxembourg Mere-infant" hospital for oropharyngeal swelling for the first and swelling of the palate for the second. The clinical expression was a swelling in both cases, of a hard, mobile consistency with healthy mucosa on their surface. The remainder of the physical examination was unremarkable. The diagnosis of a tumor of the oropharynx and palate was made following clinical radiological examinations. The histopathological examination of the surgical specimen made it possible to make the diagnosis of pleomorphic adenoma of the palate and the left palatine tonsil. The follow-up surgery was straightforward, with a favorable outcome. Pleomorphic adenomas are relatively rare benign tumors of the accessory salivary glands; their clinical expression remains swelling. Therapeutic management is surgery and the diagnosis is confirmed by histopathological examination.

Keywords

Pleomorphic Adenoma, Palate, Tonsil, Mali

1. Introduction

Pleomorphic adenoma, formerly called mixed tumor, is the most common tumor of the salivary glands, representing approximately 50% of salivary gland lesions [1]. It is mainly found in the main salivary glands: the parotid glands (80%), the submaxillary glands (10%), the sublingual glands (1%) and the oral-pharyngeal accessory salivary glands (ASG) (9%) [1]. In GSA, pleomorphic adenoma, develops mainly in the palate, followed by the labial region [1] [2]. These atypical localizations in the GSA are distinguished from those of the parotid by their clinical, histological and therapeutic characteristics [3]. We report 2 cases of pleomorphic adenoma of atypical location and then discuss the difficulties linked to its diagnostic and therapeutic management in the Malian context.

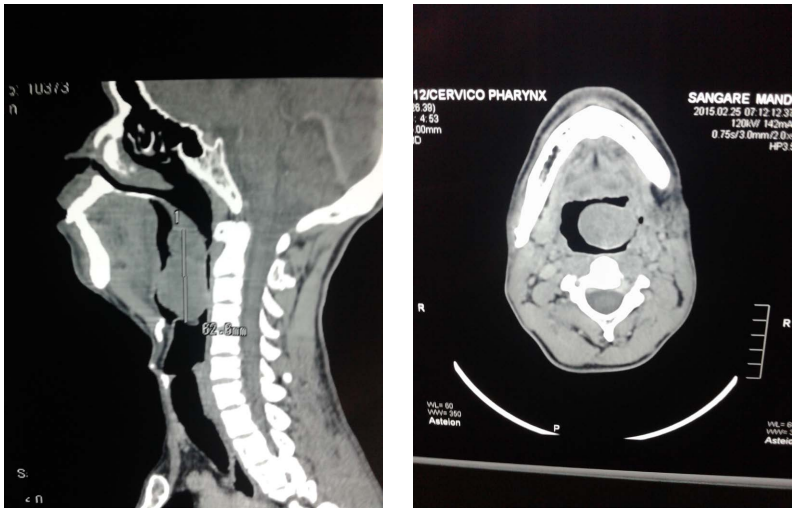
2. Observations

2.1. Observation 1

This was a 40-year-old man with a surgical history of sub-maxillectomy in 1996 for pleomorphic adenoma. He was admitted on February 10, 2015 to the ENT department of the "Luxembourg mere-infant" university hospital for dysphagia. The onset of symptoms dates back around 3 years, marked by dysphagia to solids of progressive appearance and permanent evolution. It was associated with dysphonia, dyspnea and unquantified weight loss. Several traditional treatments were carried out without success. The general condition was rated 2 according to the WHO activity index. The weight was 65 kg and there were signs of malnutrition. Examination with the tongue depressor found a large mass of hard consistency almost completely occupying the oropharynx. It was attached to the left posterior lateral pharyngeal wall. The palatine tonsil was poorly individualized. The remainder of the physical examination was unremarkable. A pharyngo-laryngeal CTscann showed an oblong homogeneous hypodensity measuring approximately 62.6 mm in diameter at the level of the hypopharynx with an implantation base on the left lateral wall of the oropharynx (**Figure 1**).

Thus, we hypothesized a tumor of the oropharynx (left tonsil) extending to the hypopharynx and an indication for surgery was made. In the operating room, under general anesthesia, we performed a right tonsillectomy then a left tonsillectomy allowing the mass to be removed in one piece (**Figure 2**).

The surgical specimen was sent to pathology. Following the operation, antibiotic prophylaxis based on injectable amoxicillin 1000 mg every 12 hours and an analgesic based on paracetamol 1000 mg combined with nefopam 20 mg injectable every 6 hours intravenously for 5 days were started.



(a) (b)

Figure 1. (a) CT image in sagittal section showing a pedunculated oropharyngeal mass extending towards the hypopharynx; (b) CT image in axial sections showing a mass reducing the Oropharyngeal lumen.

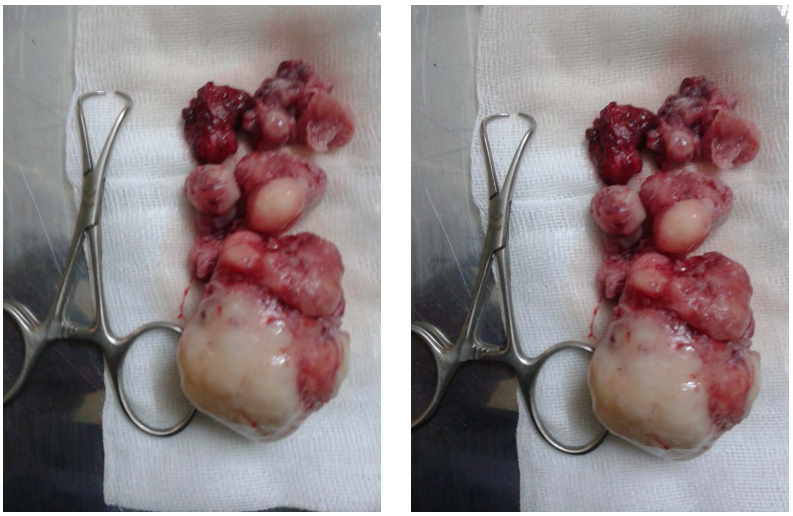


Figure 2. Post-operative image of pleomorphic adenoma.

The postoperative course was simple. Histological examination revealed a pleomorphic adenoma of the oropharynx. A control pharyngo-laryngeal CTscann 2 months postoperatively was normal (**Figure 3**). To date, there has been no recurrence.

2.2. Observation 2

This was a 72-year-old woman who consulted us on March 10, 2022 for swelling of the hard palate. The onset of symptoms dates back 10 months. It had a gradual onset and a permanent evolution associated with difficulty chewing and progressive weight loss. General condition was rated 1 according to the WHO activity index. The weight was 55 kg with the presence of signs of malnutrition.

Examination of the oral cavity showed a mass located on the hard palate. It was of a hard consistency, mobile in relation to the deep plane. It was rounded with a regular outline and covered with a healthy mucous membrane. The remainder of the physical examination was unremarkable. The CTscann showed a homogeneous hypodensity at the expense of the well-defined rounded bony palate measuring 41 × 31 mm in axial section, increasing after injection of the contrast product. It did not invade neighboring structures (Figure 4). A biopsy was performed and histological examination revealed a pleomorphic adenoma (Figure 5). We performed surgical excision (Figure 6) under general anesthesia. A nasogastric feeding tube was placed. The surgical specimen sent for histology concluded that there was a pleomorphic adenoma of the accessory salivary glands. Following the operation, antibiotic prophylaxis based on injectable amoxicillin 1000 mg every 12 hours and an analgesic based on paracetamol 1000 mg combined with nefopan 20 mg injectable every 6 hours intravenously for 5 days were started. The postoperative course was simple; the nasogastric tube was removed on day 05. To date, there has been no recurrence.

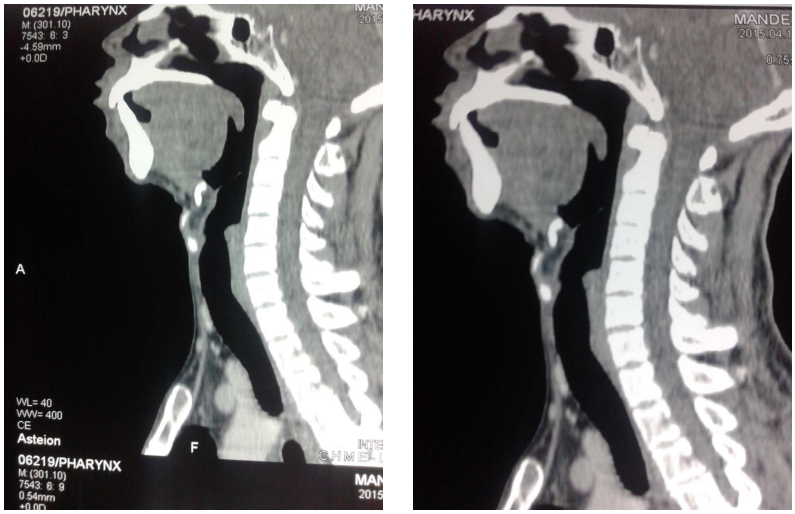


Figure 3. Control CT image after excision of the pleomorphic adenoma.

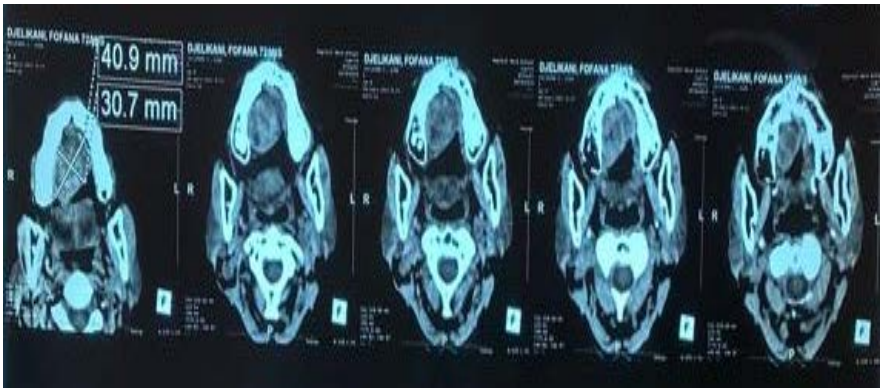


Figure 4. CT image in axial sections showing a well-defined palate mass.

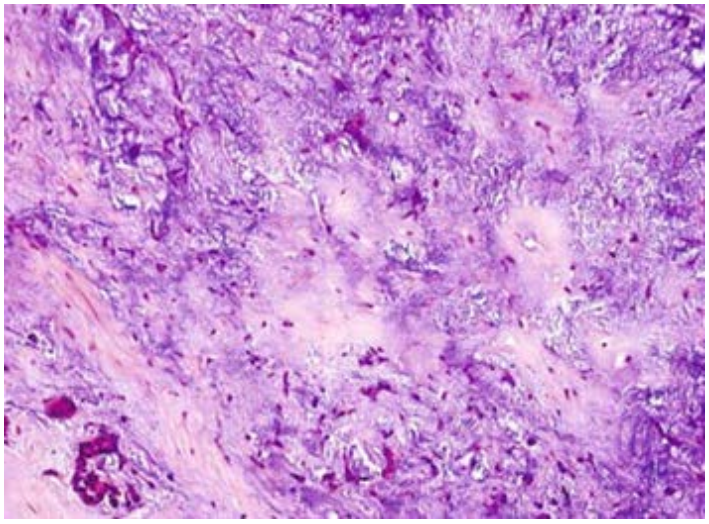


Figure 5. Histological image of pleomorphic adenoma of the palate Pleomorphic adenoma showing chondromyxoid stroma containing small friends of isolated epithelial and myoepithelial cells (HEx400).

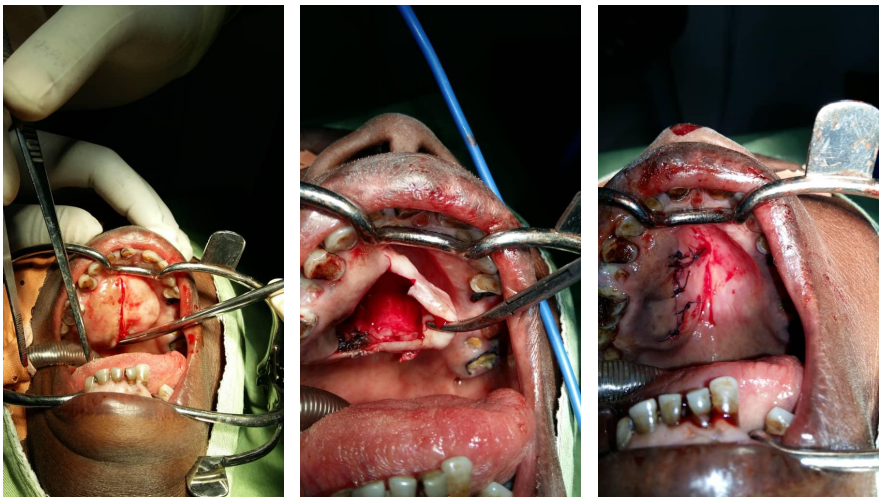


Figure 6. Intraoperative images of a pleomorphic adenoma of the palate.

3. Discussion

The tumor lesions that affect the GSA are the same as those that affect the main salivary glands but with less frequency [2]. Indeed, GSA tumors only represent 10% to 20% of all salivary tumors [2] [4]. On the other hand, and unlike those of the main glands, 50% to 60% of GSA tumors are malignant [2]. The accessory salivary glands, scattered throughout the upper aero-digestive tracts, can be the site of rare tumors and are reputed to be benign in the majority of cases, mainly in the mucous membrane of the mouth (roof or soft palate, floor of the mouth, cheeks, lips), more rarely of the larynx or nasal cavity. The tumor varieties are numerous; the most common is pleomorphic adenoma (formerly called mixed tumor) [5] [6]. Pleomorphic adenoma is the most common tumor of the parotid gland (65%). Its extra-parotid locations are rare. They are distinguished from the

parotid by their clinical, histological and therapeutic characteristics. They are distributed between the submandibular gland and the accessory salivary glands [1]. In the literature, both male and females are concerned [3] [5]. In our two patients, the tumor was located at the level of the palatal vault and the palatine tonsil. The palatal location of the pleomorphic adenoma is described by several authors [2] [5] [6]. Sixty percent of intraoral salivary tumors are found in the palatal region [2]. It is generally a firm, hard and well-defined submucosal tumor on palpation covered by a mucous membrane that is most often normal and can sometimes take on an erythematous appearance. It sits on the lateral part of the palate and, exceptionally, on the median line. This tumor is most often found on the hard palate and more rarely on the soft palate. As part of the differential diagnosis, cases of hypertrophy of the palatine salivary glands have been described with symptoms similar to those of pleomorphic adenoma. The predominant locations are the hard palate-soft palate junction. The etiology of these hypertrophies remains unknown [1] [2]. Oropharyngeal localization, particularly tonsillar, remains rare [1]. The presence of these benign tumors in the digestive tract often causes discomfort when eating. This discomfort is due to a progressive increase in tumor volume, making chewing and swallowing difficult [5] [6]. In our patients, a slow progression of 10 months and three years was noted. Indeed, in the literature, most authors agree on the slow growth of pleomorphic adenomas [1] [2]. The imaging performed in our patients made it possible to assess the tumor boundaries and the existence or not of locoregional lysis. Although we did not note signs of invasion in our patients, the aggressive form is described in the literature [2]. Performing a biopsy when the tumor is accessible allows a histopathological diagnosis confirming the benign nature of the tumor before a possible therapeutic decision is made. Therapeutic management is surgical and requires wide excision to avoid any recurrence linked to capsular dehiscences [2] [7]. In the event of a palatal location, surgical revision could then pose serious difficulties. To illustrate these points, in parotid locations, a parotidectomy is immediately performed and not a simple enucleation of the tumor and, despite this, recurrences remain frequent [1] [2]. Histologically, the tumor is generally round and well-defined and is surrounded by a fibrous capsule that is often discontinuous. This last point explains the possible recurrences if the excision is not wide enough. Its structure is polymorphic. It is composed of epithelial, myoepithelial cells and various mesenchymal substances [1] [2]. After a follow-up of nine and two years, no recurrence was noted in our patients.

4. Conclusion

Pleomorphic adenomas are relatively rare benign tumors of the accessory salivary glands. Their clinical expression remains swelling. They are mostly slow growing and are generally not aggressive. Therapeutic management is broad surgical in order to avoid recurrences and the diagnosis is confirmed by histopathological examination.

Funding

The authors declare that they have not received any specific funding for this work.

Conflicts of Interest

The authors declare that they have no conflicts of interest in relation to this article. Parental consent was taken.

References

[1] Bonfils, P. (2007) Tumeurs des glandes salivaires. *EMC-Oto-rhino-laryngologie*, **22**, 1-18. https://doi.org/10.1016/S0246-0351(07)46829-5

[2] Gleizal, A., Merrot, O. and Bouletreau, P. (2005) Affections vélopalatines. *EMC-Stomatologie*, **1**, 141-161. https://doi.org/10.1016/j.emcsto.2005.01.008

[3] Moukram, K., El Bouhairi, M. and BenYahya, I. (2017) les localisationsrares des adénomespléomorphes à propos de 4 cas. *AOS*, **285**, 1-7. https://doi.org/10.1051/aos/2017055

[4] Hamama, J., Khayati, S., Arrob, A. and Abouchadi, A. (2015) Adénomepléomorphe des glandes salivairesaccessoires. *AOS*, **271**, 4-9. https://doi.org/10.1051/aos/2015107

[5] Nouri, H., Raji, A., Elhattab, Y., Rochdi, Y. and AitM'barek, B. (2006) Adénomepléomorphe de la voûtepalatine: À propos d'un cas. *Rev d'Oto-rhinolaryngologie et de chirurgiecervico-faciale*, **302**, 22-23.

[6] Gassab, E., Berkaoui, A., Kedous, S., Korbi, A., Khairedine, N. and Harrathi, K. (2009) Adénomepléomorphe à localisation extra parotidienne. *Journal Tunisien d'ORL et de Chirurgie Cervico-Faciale*, **22**, 36-37. https://doi.org/10.4314/jtdorl.v22i1.64184

[7] Uro-Coste, E. (2007) Particularités des tumeurs des glandes salivairesaccessoires. *Annales de Pathologie*, **27**, 76-78. https://doi.org/10.1016/S0242-6498(07)92873-4