International Journal of Otorhinolaryngology
2022; 8(2): 33-36
http://www.sciencepublishinggroup.com/j/ijo
doi: 10.11648/j.ijo.20220802.11
ISSN: 2472-2405 (Print); ISSN: 2472-2413 (Online)

Extraparotid Locations of Pleomorphic Adenoma: Reflection on 21 Cases Collected in Bamako

Kassim Diarra[1], Nfaly Konate[1], Harouna Sanogo[2], Drissa Kaloga Bagayoko[3], Kalifa Coulibaly[1], Youssouf Sidibe[4], Fatogoma Issa Kone[1], Boubacary Guindo[1], Siaka Soumaoro[1], Doumbia Kadidiatou Singare[1], Mohamed Amadou Keita[1]

[1]Department of Surgery, University Hospital Gabriel Toure, Bamako, Mali

[2]Reference Health Center Kalanbancoro, Bamako, Mali

[3]Department of Anesthesia-resuscitation, University Hospital Mother-Child "Le Luxembourg", Bamako, Mali

[4]Department of Surgery, University Hospital Mother-Child "Le Luxembourg", Bamako, Mali

Email address:
diarrakassim84@yahoo.fr (K. Diarra)

To cite this article:
Kassim Diarra, Nfaly Konate, Harouna Sanogo, Drissa Kaloga Bagayoko, Kalifa Coulibaly, Youssouf Sidibe, Fatogoma Issa Kone, Boubacary Guindo, Siaka Soumaoro, Doumbia Kadidiatou Singare, Mohamed Amadou Keita. Extraparotid Locations of Pleomorphic Adenoma: Reflection on 21 Cases Collected in Bamako. *International Journal of Otorhinolaryngology*. Vol. 8, No. 2, 2022, pp. 33-36. doi: 10.11648/j.ijo.20220802.11

Received: June 14, 2022; **Accepted**: June 27, 2022; **Published**: July 5, 2022

Abstract: Objectives: To study the epidemiological, diagnostic and therapeutic characteristics of this rare histological entity and to discuss it with the literature. Materials and methods: This was a descriptive, retrospective and longitudinal study over a period of 7 years from January 2015 to December 2022. We made an exhaustive sampling of all patients seen and treated for pleomorphic adenoma (PA) salivary glands with the exception of parotid locations in the ENT department Surgery, University hospital Gabriel Tour. Results: During the study period, 63 patients were operated on in the department for pleomorphic adenoma of the salivary glands, 21 patients met our criteria, either 9 men or 12 women with an average age of 38 years with extremes ranging from 7 at 78 years old. The seat was the submandibular gland in 11 cases (52.3%), the hard and soft palate 6 cases (28.5%), the oropharynx 1 case (4.8%), lower lip 1 case (4. 8%), nasal fossa 1 case (4.8%), para-pharyngeal space 1 case (4.8%). The clinical examination found a firm, mobile, painless and well-defined swelling in the area of interest. Computed tomography (CT) showed a mass with a regular border, of homogeneous tissue density with moderate enhancement by the contrast product (PDC), with no bone lysis opposite. Cervicofacial ultrasound was performed for 100% of cases of tumors with submandibular location. Surgery was the treatment of choice for all patients. Conclusion: The pleomorphic adenoma raises a problem of differential diagnosis with other tumors of the salivary glands. Rigorous monitoring must be conducted finally to detect signs of malignancy and recurrence after surgery.

Keywords: Pleomorphic Adenoma, Salivary Glands, Surgery, Bamako

1. Introduction

Pleomorphic adenoma, formerly called mixed tumor because of its dual epithelial and mesenchymal component, is a benign tumor that grows slowly, quietly, over several years [1]. Salivary gland tumors are rare, they represent 3 to 5% of cervico-facial tumors and 75% of them are benign. These tumors can develop in the main or accessory salivary gland. In 20% they develop at the expense of accessory salivary glands [2]. The histological natures developed at the expense of the accessory salivary glands are very varied, the most frequent of which is the pleomorphic adenoma. The pleomorphic adenoma of the accessory salivary glands, also called mixed tumor, is a heterogeneous benign epithelial tumor [2, 3]. Their preferred seat is the palate, which contains more than 50% of the accessory salivary glands [3]. This tumor has the potential for malignant transformation into carcinoma after a long course and recurrence in the event of incomplete surgical excision or capsular rupture. It is essential to evoke a malignant transformation in the face of

certain signs, in particular a recent increase in size, ulceration, infiltration and spontaneous bleeding [4, 5]. In our context, there was no specific study on the subject. Thus we approached it in its entirety in order to have a database in the service and compared our results with data from the literature while setting ourselves the objectives of studying the epidemiological, diagnostic and therapeutic characteristics of this histological entity rare.

2. Materials and Methods

This was a descriptive, retrospective and longitudinal study over a period of 7 years from January 2015 to December 2022. We made an exhaustive sampling of all patients, men and women of all ages seen and treated for pleomorphic adenoma (AP) of the salivary glands with the exception of the parotid locations in the ENT department of the university hospital Gabriel TOURE.

Non-inclusion criteria:

Parotid localized adenomas, patients who have refused any therapeutic procedures, any patient treated outside the said interval and who does not present any histological evidence as well as all cancers of the salivary glands.

Sampling:

The sampling was exhaustive and we obtained 21 cases during the study period.

Variables studied:

Frequency, gender, age, socioeconomic status, consultation time, clinical signs, tumor site, computed tomography (CT), ultrasound, histology, treatment received and postoperative follow-up.

Average follow-up time was 3 years.

Data analysis:

The data was entered and analyzed on Word and Excel 2019 software which allowed us to obtain the following results.

Ethical and administrative considerations:

This is purely scientific work aimed at improving patient care. Anonymity was strictly respected. The consent of the patients or their relatives (accompanying) was obtained beforehand.

3. Results

During the study period, 63 patients were operated on in the service for pleomorphic adenoma of the salivary glands, 21 patients met our criteria, either 9 men and 12 women with an average age of 38 years with extremes ranging from 7 to 78 year. The seat was the submandibular gland in 11 cases (52.3%), the hard and soft palate 6 cases (28.5%), the oropharynx 2 cases (9.5%), lower lip 1 case (4. 8%), nasal fossa 1 case (4.8%) (Table 1). No notable pathological history was observed. Patients consulted after an average period of 3 years with extremes ranging from 6 months to 9 years. The reason for consultation was in 100% of cases an isolated swelling in the region concerned, slowly increasing in volume. Upper dysphagia associated with rhinolalia has been reported by patients with a tumor located in the palate and oropharynx. The clinical examination found a firm, mobile, painless and well-defined swelling in the area of interest. The palate tumors extended to the right and left retro molar region, pushing the uvula inwards and covering almost all of the oropharynx, preventing the proximal pole from being seen. The adjacent mucosa was healthy. They measured on average 4cm in transverse diameter, painless, of firm consistency.

Computed tomography (CT) performed in 4 cases regardless of the site of the tumor, it objectified a mass with regular limits, of homogeneous tissue density with moderate enhancement by the contrast product (PDC), without bone lysis opposite. Cervicofacial ultrasound was performed for 100% of cases of tumors with submandibular location. She most often identified a well-limited, hypoechoic, heterogeneous image with multiple fluid compartments.

The preoperative assessments were all normal, which led us to perform total excision of the tumors with curettage of the tumor bed. Surgery was the treatment of choice for all patients. Excision under local anesthesia was performed in the case with labial localization. A submandibulectomy was performed for the 11 cases with submandibular location. Tumors located on the palate were operated intraorally under general anesthesia. Tumors of the oropharynx were also operated by pure endobuccal approach and that of the nasal fossa by endonasal approach. In general, the masses were easily detachable, always presenting a cleavage plane whether by endobuccal, endonasal or laterocervical approach. The anatomopathological examination of the surgical specimens concluded to a pleomorphic adenoma. The average hospital stay was 7 days. With an average follow-up of 3 years, we did not note any recurrence or malignant transformation.

Figure 1. *A) Front view of a patient preoperatively: Bulky cervical swelling extending to the area basicervical. B) Bulging of the palate with mucous membrane normal looking. C) Externalized budding mass from the left nasal cavity, not bleeding on palpation.*

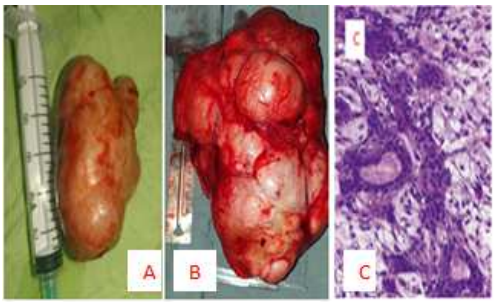

Figure 2. *A and B) Operating parts C) Histopathological image.*

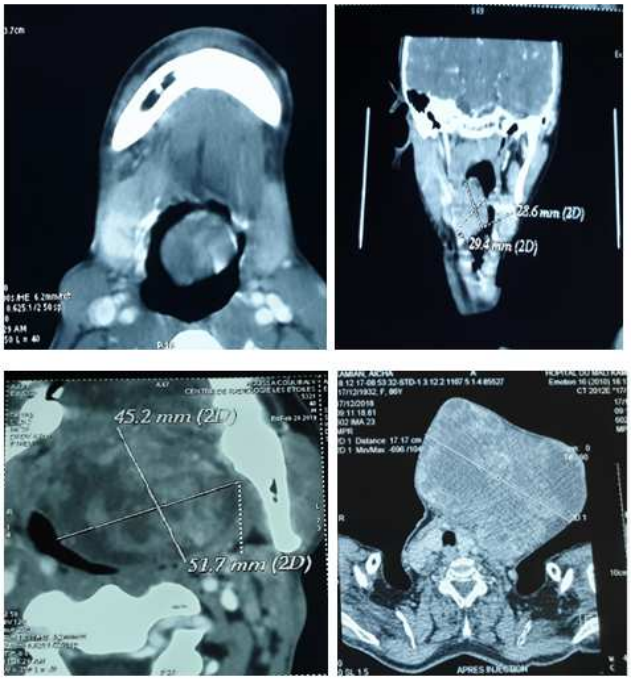

Figure 3. *A and B) Isodense mass slightly enhanced after injection of contrast product at the expense of the oropharynx between C2 and C4. C) CT image in axial section showing a density mass tissue, well limited to the soft palate. D) CT image in axial section showing tissue density formation heterogeneous polylobed with regular contours, occupying the entire region left anterolateral cervical spine measuring 16.5 cm in diameter.*

Table 1. *Distribution by site of pleomorphic adenomas.*

Tumor sites	Effective	Percentage
Submandibular gland	11	52,3
Palace	6	28,5
Oropharynx	2	9,5
Bottom lip	1	4,8
Nasal cavity	1	4,8
TOTAL	21	100

4. Discussion

We noted, in agreement with the literature that the extra parotid localization of the pleomorphic adenoma is a rare entity of tumors of the salivary glands. Of the 900 hundred anatomopathological results of the last ten years of service, it represents 2.33% of cases. The salivary glands are disseminated throughout the upper aerodigestive tract, they can be the seat of tumors especially in the mucous membrane of the mouth [1, 3]. These rare tumors occur on average between the third and fifth decade and affect women slightly more than men [3]. They are not the prerogative of the child [5]. In our series, there was only one child and the location was in the left nasal cavity. We found no significant difference between the two sexes and the average age of our patients was 38 years. These tumors can pose a diagnostic and therapeutic problem depending on their location and especially their size [6]. The period of evolution is long and very varied according to the literature; it is 10 years in the series of NOURI H [3]. The authors reported an average delay of 2 years with extremes ranging from 1 month to 20 years [2, 5]. The average time of evolution of our patients was 3 years with extremes ranging from 6 months to 9 years. The clinical spectrum depends on the size of the tumor, small tumors are sometimes asymptomatic and large tumors under the effect of their mass can cause obstructive signs, namely an endobuccal or pharyngeal gene, snoring, closed rhinolalia, or even inspiratory dyspnea, slow growth should be sought [6]. The endobuccal examination finds an oropharyngeal mass, firm, renitent, painless, covered by a healthy mucosa, mobile in general. The mucosa is respected for a long time, only appearing blown by the tumor formation. The main signs concerned rhinolalia, pharyngeal gene and ronchopathy, nasal obstruction in our patients whose adjacent mucosa was healthy. Computed tomography is essential in the preoperative assessment of pleomorphic adenoma. It makes it possible to characterize the tumor, to evaluate its extension to the neighboring tissues and to look for bone lysis. MRI is the most effective examination, it highlights the typical appearance of a pleomorphic adenoma. The characteristic appearance is a lobulated tumor, well limited, in T1 hyposignal and T2 hypersignal, which enhances homogeneously after injection of contrast product [7]. Imaging generally guides the choice of surgical approach and predicts operative difficulties [6]. The CT scan performed in our patients allowed us to rule out tumor extension or lysis. In our work, we did not use MRI since it is not available in our context. Diagnosis is mainly based on histological examination [6]. This examination macroscopically highlights a tumor of firm consistency, with a grey-whitish surface, in places translucent on cutting and histologically, there is a proliferation of epithelial and myoepithelial cells which produce a mucopolysaccharide matrix which may undergo chrondroid or bone metaplasia [6, 8]. The anatomopathological examination allowed us to conclude with the diagnosis of pleomorphic adenoma in all cases.

Pleomorphic adenomas pose the differential diagnosis with other tumors of the salivary glands such as polymorphic adenocarcinoma of low malignancy: the palate is its preferential location, it occurs in patients over 40 years old and is manifested by a mobile or fixed swelling, slow growing, painless with lymph node metastases. The mucosa can be ulcerative and exophytic [9]. Adenoid cystic carcinoma, or cylindroma, accounts for 30% of tumors of the accessory salivary glands, its preferred site being the palatine region. The symptomatology is noisy (pain, paresis, lymphadenopathy). The tumor is solid, riddled with cavities, poorly circumscribed [6]. Clear cell adenocarcinoma, verrucous carcinoma, acinar cell carcinoma, fibrosarcoma, hemangiopericytoma, hemangioendothelioma. The pains are earlier in the soft palate than in the hard palate. The invasion of the tumor from the veil is towards the anterior pillar and the ipsilateral infratemporal fossa [9, 10]. The treatment of pleomorphic adenoma is surgical from the outset according to the review of the literature [8]. The surgical approach depends on the size of the tumour. The approaches used are:

the trans-velar endobuccal route, the cervical route, the trans-parotid route associated with an endobuccal route in the event of a large tumor of the palate. Total excision of the tumor with curettage of the tumor bed must be performed [6, 11, 12]. Some authors have recommended wide tumor excision with a 5mm margin in healthy tissue (1). Any partial surgery or capsular ruptures are sources of recurrence [6, 8]. In the event of substantial loss of substance, secondary reconstruction by flap is necessary [9]. Our patients were all operated on either by intraoral route for tumors of the palate as well as those of the oropharynx. The submandibular tumors were operated by cervicotomy and that of the false nasal by endonasal way. In one patient, intubation was difficult due to the volume of the tumor in the palate, so we resorted to tracheostomy. All the patients benefited from a curettage of the tumor bed after the total excision of the tumor, one did not resort to the reconstruction by a flap. With an average follow-up of 3 years, we had no recurrence.

5. Conclusion

The pleomorphic adenoma raises a problem of differential diagnosis with other tumors of the salivary glands. Surgery occupies an undeniable place for treatment. According to the review of the literature, this mixed tumor raises a double problem: malignant transformation and recurrence. Its recurrence is a real challenge for the surgeon. Compliance with the principle of adenoma surgery and early management can overcome this dual problem. Rigorous monitoring must be conducted finally to detect signs of malignancy and recurrence.

Conflicts of Interest

All the authors do not have any possible conflicts of interest.

References

[1] Moukram K., El Bouhairi M., Ben Yahya I. Les localisations rares de l'adénome pléomorphe [Rare localizations of pleomorphic adenoma]. AOS 2017; n° 285: 1-7. https://doi.org/10.1051/aos/2017055

[2] Ben Brahim E, Ferchiou M, Khayat O, Zribi S Et Al. Les tumeurs des glandes salivaires: Etude anatomo-clinique et épidemiologique d'une série de 180 cas [Tumors of the salivary glands: Anatomo-clinical and epidemiological study of a series of 180 cases]. Tunisie médicale 2010; 88: 240-244.

[3] Nouri H, Raji A, Elhattab Y, Rochdi Y, Ait M'barek B. Adénome pléomorphe de la voûte palatine. A propos d'un cas [Pleomorphic adenoma of the palatal vault. About a case]. La Lettre d'Oto-rhino-laryngologie et de chirurgie cervico-faciale 2006; 302: 22-23.

[4] Hamama J., EL Khayati S., Arrob A., Khalfi L., Abouchadi A., Elkhatib M. K. Adénome pléomorphe des glandes salivaires accessoires [Pleomorphic adenoma of the accessory salivary glands]. AOS 2015; 271: 4-9 DOI: 10.1051/aos/2015107.

[5] Bouguacha L, Kermani W, Abdelkafi M, Hasni I, Et Al. Adénome pléomorphe des glandes salivaires accessoires [Pleomorphic adenoma of the accessory salivary glands]. J. Tun orl 2010; 24: 35-39.

[6] Gattia N, Kachouchia K V Kouassi N, Tanon M, Et Al. Tumeur mixte du palais: à propos de deux cas [Mixed tumor of the palate: about two cases]. Médecine d'afrique noire octobre 2011; 58: 479-481.

[7] Gassab E, Berkaoui A, Kedous S, Korbi A, Et Al. Adénome pléomorphe à localisation extra-parotidienne [Pleomorphic adenoma with extra-parotid localization]. J. Tun orl 2009; 22: 36-39.

[8] Bouaity B, Nadour K, Errami N, Chihani M, Et Al. Adénome pléomorphe de l'oropharynx [Pleomorphic adenoma of the oropharynx]. La Lettre d'ORL et de chirurgie cervico-faciale 2009; 319: 23-25.

[9] El Bakkouri W, Barry B. Les tumeurs du voile du palais [Soft palate tumors]. La lettre d'Oto-rhino-laryngologie et de chirurgie cervico-faciale 2003; 283: 16-17.

[10] Y. Yazibenea, N. Ait-Mesbaha, S. Kalafatea. Adénome pléomorphe dégénéré de la fosse nasale [Degenerated pleomorphic adenoma of the nasal cavity]. Annales françaises d'oto-rhino-laryngologie et de pathologie cervico-faciale 2011; 128: 41-45.

[11] S. Hirai et al. Pleomorphic adenoma in nasal cavity: immunohistochemical study of three cases. Auris Nasus Larynx 2002; 29: 291-295.

[12] Honghai Fu, Jun Wang, Lizhen Wang. Pleomorphic adenoma of the salivary glands in children and adolescents. Journal of Pediatric Surgery. 2012; 47: 715-719.