

Voluminous Amygdaloid Cyst: A Case Report

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Abstract

Case Report

Amygdaloid cysts also known as lymphoepithelial cysts or branchial cleft cysts are rare and benign dysembryological cystic tumors that develop in the antero-lateral part of the neck; they represent 2% of laterocervical tumors of the neck. Originating from the second branchial cleft, due to the persistence of the cervical sinus during the differentiation of the branchial apparatus. We report the case of a 34-year-old man who consulted for a huge right laterocervical swelling that had been gradually increasing in volume for 8 months without any other associated symptoms. A cervicotomy and a complete resection of the cyst was performed, histopathological examination of the surgical specimen confirmed the diagnosis of the amygdaloid cyst without signs of malignancy. The aims of this study are to analyze the anatomo-clinical features and to discuss the modalities of care and the therapeutic indications of this affection.

Keywords: Amygdaloid cysts, lymphoepithelial cysts, branchial cysts, case report.

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INTRODUCTION

A variety of names has been used for the amygdaloid cysts: Branchial cyst, tumor of the branchial cleft, lateral lympho-epithelial cyst and benign cystic lymph nodes ... [1].

They are rare benign cystic tumors due to congenital malformations resulting from abnormal embryonic development of the upper laterocervical region which represents approximately 2% of all laterocervical tumors and are one of the most common branchial anomalies, accounting for 6.1% to 85.2% of second cleft anomalies [2].

The usual site is the middle third of the anterior border of the sternocleidomastoid muscle, but they can be anywhere from the middle constrictor muscle of the pharynx to the Laterocervical region [3]. We report this rare observation of a lymphoepithelial cyst located in the right Laterocervical region in a 34-year-old man.

Treated in our department to analyze the anatomo-clinical and therapeutic particularities of this pathology and compare it with data from the literature.

CASE REPORT

We report the case of a 34-year-old male patient admitted to our department, with the main complaints of a right laterocervical swelling.

The medical history is marked by a chronic myeloid leukemia, diagnosed a year ago, the patient is currently under Tyrosine kinase inhibitor treatment: Imatinib, no pharmacological allergies, no psychosocial problems, or family genetic disease reported.

The mass had first appeared 8 months ago and had increased in size slowly without any previous signs of infections within the head and neck region. On the clinical review he was apyretic, hemodynamic and respiratory stable. The clinical examination revealed a fluctuant, mobile mass within the right cervical region that was approximately 5-6 cm in diameter. There was no clinical evidence of a sinus or fistulous tract (Figure 1). Cervical CT scan showed a well-defined cystic lesion exerting a mass effect on the jugulocarotid axis (Figure 2 & 3).

The patient underwent a right cervicotomy with complete resection of the cyst. The dissection of the mass was step by step respecting the vascular axis of the neck (Figure 4). During the operation, no tract or cord connecting the cyst to the pharynx was noted. postoperative follow-up was simple. The patient adhered

well to the treatment received with a good tolerance to the surgery and post-operative care including antibiotics, local care. The histological examination confirmed the

diagnosis of lymphoepithelial cyst. The follow-up was essentially clinical without questionnaire or pre-established scale.



Figure 1: Pre-operative photo of the patient showing right latero-cervical mass

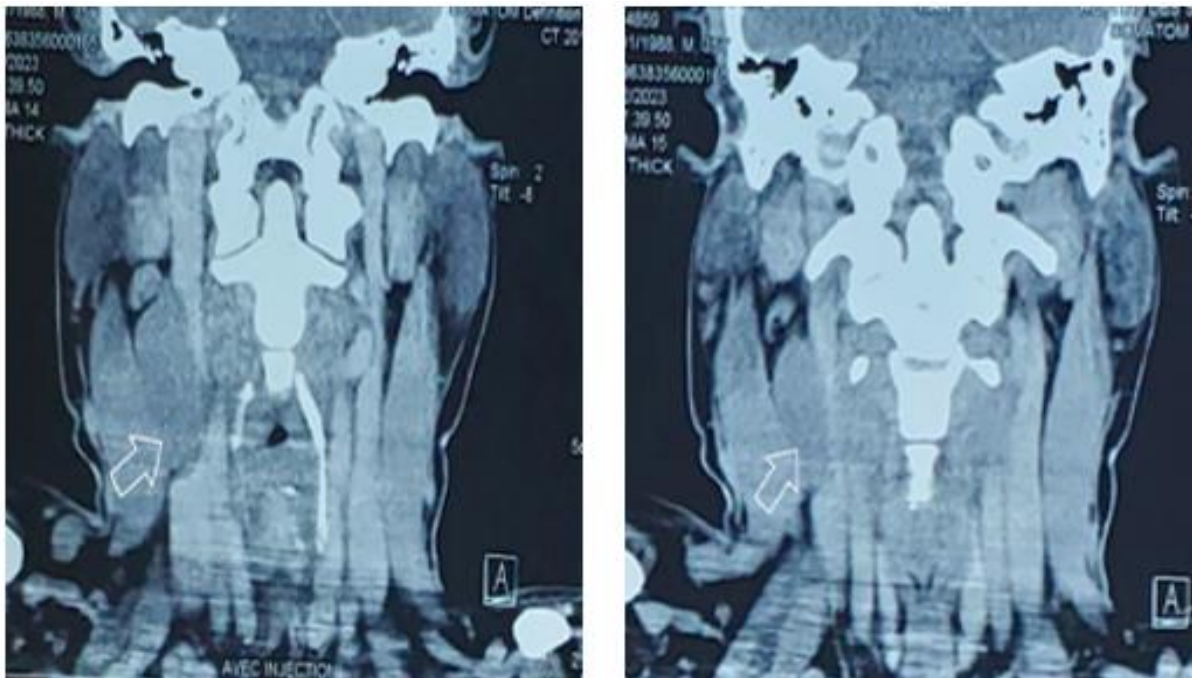


Figure 2: Cervicofacial CT in coronal section showing a well-limited cystic formation exerting a mass effect on the jugulocarotid axis

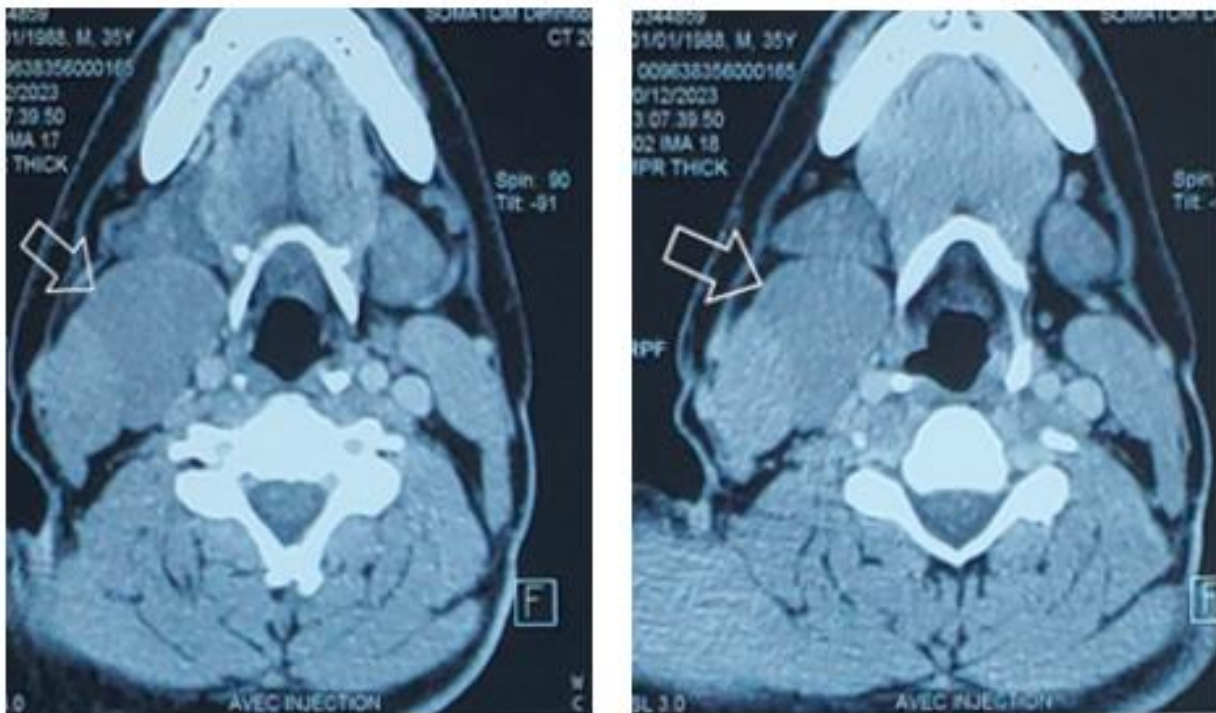


Figure 3: Cervicofacial CT in axial section showing well-limited cystic formation exerting a mass effect on the jugulocarotid axis

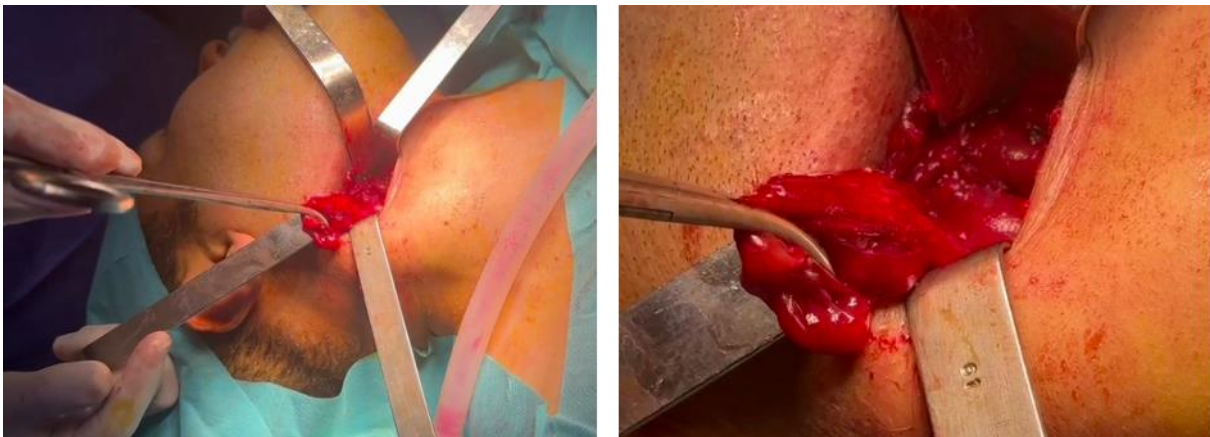


Figure 4: Per-operative photos showing the incision and surgical removal of the cyst

DISCUSSION

Congenital cysts and fistulas of the face and neck are infrequent and poorly understood malformations of embryological origin. Amygdaloid or lympho-epithelial cysts, or cervical sinus cysts, are rare benign cystic dysembryological tumors that correspond to resorption defects of the second branchial arch [2, 4] and develop in the anterolateral part of the neck. The frequency of amygdaloid cysts in relation to second branchial arch defects ranges from 6.1% to 85.2% [2]. The age of discovery is older than for other congenital anomalies, with two peaks in frequency, in children under 5 years of age and between the 2nd and 3rd decades, with no gender predominance. Clinically, a cervical sinus cyst appears as an oval, renitent swelling, mobile under the superficial planes, usually located near the carotid bifurcation in a subhyoid position [4, 5].

Although these lesions are congenital, they are usually identified between the second and fourth decades of life, when it increases in size or becomes symptomatic because of infection or the extension of the mass [2]. In our case the lateral cervical cyst was identified at the age of 34. The lesion may communicate externally through a narrow channel, forming a so-called external cervical fistula, the external orifice of which is located in the junction between the upper one-third and lower two-thirds of the anterior sternocleidomastoid muscle, but it can occur at any level from the hyoid to the suprasternal notch [6]. The cyst sometimes communicates with the skin or pharynx, spontaneously or following infection via an internal fistula that opens into an embryonic derivative of the second branchial pouch, the palatal amygdala [2], and sometimes. Depending on the size and the anatomical extension of the mass, local

symptoms, such as dysphagia, dysphonia, dyspnea, and stridor, may occur [7].

These cysts were classified in 1929 by BAILEY into four subtypes [8]:

Type I: the most superficial and lies along the anterior surface of the sternocleidomastoid to the platysma, but not in contact with the carotid sheath.

Type II: the most common type where the branchial cleft cyst is located in front of the sternocleidomastoid muscle, behind the submandibular gland, adjacent and lateral to the carotid sheath.

Type III: extends medially between the bifurcation of the internal and external carotid arteries, lateral to the pharyngeal wall.

Type IV: lies deep in the carotid sheath in the pharyngeal mucosal space and opens into the pharynx.

The diagnosis of branchial cleft cyst is made primarily by medical history, clinical manifestations and exclusion of the differential diagnoses. We can evoke all the causes of chronic cervical masses in adults, including a lymphadenopathy which may reveal cancer of the upper aerodigestive tract, cervical lymphoma, tuberculous adenitis in our context, cryptic metastasis of tonsillar carcinoma. Which make the indication of anatomopathological examination essential [2, 9, 10].

On the radiological level, ultrasound remains the first-line radiological examination to be requested because it is a non-invasive, inexpensive, easy to perform and anodyne examination. However, when a mass is voluminous, the use of computer tomography CT scan and Magnetic resonance imaging (MRI), is necessary to better study the extension of the mass. On CT scan the cysts generally appear well circumscribed, uniformly hypodense with thin walls, that may increase after superinfection of the cyst. As for MRI, it gives a better and more precise representation of the cyst. Hypo to isointense in T1 sequences and hyperintense in T2 sequences. CT or MRI are also useful to differentiate the lesion from other parapharyngeal tumors: a hemangioma, dermoid cyst, lymphangioma or metastatic lymphadenopathy [2, 6]. In case of presence of fistula, fistulography is necessary and it allows to clearly visualize the location and extent of the fistulous path [9, 11]. The diagnosis of amygdaloid cysts is confirmed by histology. Fine-needle aspiration biopsy for cytological criteria can be an important complement to clinical diagnosis in preoperative. The criteria for Fine-needle aspiration cytologic diagnosis of branchial cysts are: a) thick, yellow, pus-like fluid, b) anuclear, keratinising cells, c) squamous epithelial cells of variable maturity and d) a background of amorphous debris [12]. Histologically, the amygdaloid cyst is lined by an epithelium that is most often squamous [2], but it may also be a ciliated columnar epithelium of ectodermal origin. The presence of lymphoid tissue with or without

germinal centers in the subepithelial connective tissue are the most prominent morphologic characteristics [13].

Cervical sinus cysts can cause discomfort and bradycardia when they are located at the level of the carotid bulb, to improve the patient's symptoms a cyst puncture can be proposed. Malignant intracystic degeneration remains exceptional [14]. However infection remains the most frequent complication of this malformation.

Therapeutic management is based on surgery, and should be carried out as early as possible to avoid complications, This surgical treatment consists of a complete surgical resection, through a transverse cervicotomy under general anesthesia. Approximately 80% of branchial sinuses will open to the skin, and fewer will open to the pharynx. The fistulous path can be identified by catheterization with methylene blue. Clinicians must take care to remove the entire tract to decrease the chance of recurrence [15].

Dissection continues at the point of contact with the fistula path, stopping rapidly in the case of a blind external fistula. If the fistula continues upwards, a second incision is required to follow its course. Rupture of the fistula at the top is usually of no consequence, as the residual fistula drains into the oropharynx. Finally, there is no need for a systematic tonsillectomy [2].

The postoperative evolution is generally good. However, some complications were reported such as bleeding, surgical site infection, nerve damage involving the spinal accessory nerve, the mandibular branch of the facial nerve, the greater auricular nerve, the hypoglossal nerve and the superior laryngeal nerve [11]. Recurrence rates after primary surgery are estimated at 3% - 4% [16]. Alternative treatments, such as percutaneous sclerotherapy, remain unproven [17].

CONCLUSION

Amygdaloid cysts, branchial cleft cysts, or cervical lymphoepithelial cysts, are benign dysembryologic cystic tumors developing in the anterolateral portion of the neck. They share a clinical presentation with other pathological entities of the neck, making it easily misdiagnosed by clinicians. The fear is specially to miss the diagnosis of an intracystic metastasis or a cystic lymph node metastasis from a distant primary carcinoma. Therapeutic management is always surgical and should be performed as early as possible to limit the risk of inflammatory changes related to infectious episodes.

Conflicts of Interest: No

REFERENCES

- Golledge, J., & Ellis, H. (1994). The aetiology of lateral cervical (branchial) cysts: past and present

- theories. *The Journal of Laryngology & Otolaryngology*, 108(8), 653-659.
2. Charfi, A., Abid, W., Romdhane, N., Madiouni, A., Chahed, H., Zainine, R., ... & Besbes, G. (2014). Les kystes amygdaloïdes: à propos de 31 cas. *Journal Tunisien d'ORL et de Chirurgie Cervico-Faciale*, 31, 24-26.
 3. Rouihi, A., Hemmaoui, B., Errami, N., & Benariba, F. (2018). Enorme kyste amygdaloïde cervical à propos d'un cas. *Pan African Medical Journal*, 31(1).
 4. Madoz, A., & Frampas, E. Gayet-de la croix M, Liberge R, Dupas B. *Imagerie des formations kystiques congénitales cervicales. Service central de radiologie et imagerie médicale CHU de Nantes.*
 5. Coiffier, T. Garabedian. *Que faire devant les kystes et les fistules de la face et du cou.*
 6. Glosser, J. W., Pires, C. A. S., & Feinberg, S. E. (2003). Branchial cleft or cervical lymphoepithelial cysts: etiology and management. *The Journal of the American Dental Association*, 134(1), 81-86.
 7. Thomaidis, V., Seretis, K., Tamiolakis, D., Papadopoulos, N., & Tsamis, I. (2006). Branchial cysts. A report of 4 cases. *Acta Dermatovenerologica Alpina, Pannonica, et Adriatica*, 15(2), 85-89.
 8. Bailey, H. (1929). Branchial Cysts and Other Essays on Surgical Subjects in the Fascio-Cervical Region. H.K. Lewis & Company Ltd., London.
 9. de Régloix, S. B., Maurin, O., Crambert, A., Genestier, L., Bonfort, G., & Pons, Y. (2019). Kystes et fistules congénitales du cou chez l'adulte. *La Presse Médicale*, 48(1), 29-33. <https://doi.org/10.1016/j.lpm.2018.09.019>
 10. Najib, Z., Berrada, O., Lahjaouj, M., Oukessou, Y., Rouadi, S., Abada, R. A., ... & Mahtar, M. (2021). Cervical lymphoepithelial cyst: Case report and literature review. *Annals of Medicine and Surgery*, 61, 185-187. <https://doi.org/10.1016/j.amsu.2020.12.041>
 11. Pacheco-Ojeda, L., Ayala-Ochoa, A., & Salvador, K. (2020). Branchial cysts in Quito, Ecuador. *International Archives of Otorhinolaryngology*, 24, 347-350. <https://doi.org/10.1055/s-0039-1695023>.
 12. Regauer, S., Gogg-Kamerer, M. A. R. G. I. T., Braun, H., & Beham, A. (1997). Lateral neck cysts—the branchial theory revisited: A critical review and clinicopathological study of 97 cases with special emphasis on cytokeratin expression. *Apmis*, 105(7-12), 623-630.
 13. Hirota, J., Maeda, Y., Ueta, E., & Osaki, T. (1989). Immunohistochemical and histologic study of cervical lymphoepithelial cysts. *Journal of Oral Pathology & Medicine*, 18(4), 202-205.
 14. Abdenour, S., Allag, S., & Benhalima, H. (2014). Un cas de dégénérescence maligne de kyste amygdaloïde. *Revue de Stomatologie, de Chirurgie Maxillo-faciale et de Chirurgie Orale*, 115(6), 385-388.
 15. Conway, H., & Jerome, A. P. (1955). The surgical treatment of branchial cysts and fistulas. *Surgery, gynecology & obstetrics*, 101(5), 621-628.
 16. Bellakhdhar, M., Haouas, J., El Abed, W., Ghammem, M., Kermani, W., & Abdelkefi, M. (2018). Kystes amygdaloïdes: À propos de 34 cas. *La Tunisie Médicale*, 96, 888-892.
 17. Donegan, J. O. (1993). Congenital neck masses. *Cummings CU, Frederick JM, Harker LA, et al. Otolaryngology Head and Neck Surgery*, 2, 1554-1565.