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# Cervical lymphoepithelial cyst: Case report and literature review

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#### ABSTRACT

*Introduction:* The cervical lymphoepithelial or branchial cleft cyst are benign dysembryologic cystic tumors developing in the anterolateral region of the neck. They are relatively uncommon anomalies.

The aims of this study are to analyze the anatomoclinical features and to discuss the modalities of care for the management of this disease.

*Presentation of case*: We report a case of a 70 years-old woman who was admitted to our department with a complaint of painless mass in the right supraclavicular region. Clinical examination and radiological investigations found a mass compatible with cervical cyst. Treatment consisted of the complete resection of the cyst. histopathological examination found a Cervical lymphoepithelial cyst.

*Conclusion:* The cervical lymphoepithelial can be easily misdiagnosed. It is imperative that clinicians make an accurate diagnosis for appropriate treatment (that is, surgical excision).

### 1. Introduction

Cysts presented in the lateral aspect of the neck were first described by Hunczovsky. Since then, a variety of names has been used for these cysts: branchial cyst, tumour of the branchial cleft, lateral lymphoepithelial cyst and benign cystic lymph nodes ... [1].

They are due to the persistence of the cervical sinus during the differentiation of the branchial apparatus. 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 supraclavicular region [2]. We report this rare observation of a lymphoepithelial cyst located in the right supraclavicular region in a 70-year-old woman. This work is reported by following the surgical case report (SCARE) guidelines [3].

### 1.1. Presentation of case

A 70-year-old woman was admitted to the department of ENT 20 august hospital, with the main complaints of a right laterocervical swelling.

The medical history was unremarkable, no pharmacological allergies, no psychosocial problems, smoking and no family genetic disease.

The mass had first appeared one years ago and had increased in size slowly without any previous signs of infections within the head and neck region.

On the clinical review she was apyretic, hemodynamic and respiratory stable. The External examination revealed a fluctuant, mobile mass within the right supraclavicular region that was approximately 4–5 cm in diameter.

There was no clinical evidence of a sinus or fistulous tract.

On cervical ultrasound examination we found a right latero-cervical cystic formation which may correspond to a branchial cyst of late discovery or cystic lymphangioma. Cervical CT scan showed a well-defined cystic lesion that was confined to the right supraclavicular region (Fig. 1).

The patient underwent a right cervicotomy with complete resection of the cyst, performed by professor ENT. The dissection of the mass was step by step using the bipolar forceps respecting the vascular axis of the neck. During the operation, no tract or cord connecting the cyst to the pharynx was noted (Fig. 2). 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 shows a cystic wall covering of squamous type, often abraded. the underlying chorion is fibrous punctuated

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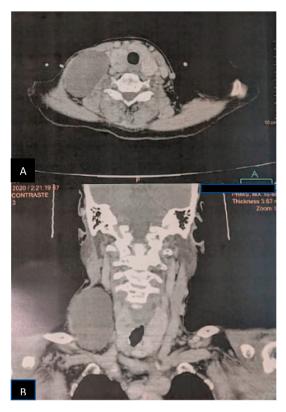


Fig. 1. A+B Cervical CT scan (axial and coronal view) demonstrated a well-limited cystic formation in the right supraclavicular region.



Fig. 2. Photo showing surgical removal of the cyst.

by lymphocytes grouping together in a follicle suggesting a lymphoepithelial cyst.

The patient was followed in ENT consultation every two weeks for the first month and then once every three months for the first year. The follow-up was essentially clinical without questionnaire or preestablished scale.

The patient, 1 years later still remains healthy.

#### 2. Discussion

Amygdaloid or lymphoepithelial cysts or cervical sinus cysts are rare benign dysembryologic cystic tumors which correspond to resorption defects of the second branchial arch [4,5].

Despite the fact that branchial anomalies, such as sinuses and fistulae, are usually diagnosed during infancy, the lateral cervical cysts are identified most commonly between the second and fourth decades of life, when they grow bigger because of infection or other causes [6]. In our case the lateral cervical cyst was identified at the age of seventy.

The swelling typically develops 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 [7]. Chandler and Mitchell [8] described the location as being between the tragus and clavicle.

The cyst sometimes communicates with the skin or pharynx, spontaneously or following infection. Communication with the outside is through a narrow canal called an external cervical fistula, the external opening of which is often located at the meeting of the middle third and lower third of the anterior border of the sternocleidomastoid muscle. While communication with the pharynx, rarer than the previous one, is via an internal fistula that opens into an embryonic derivative of the second branchial pouch, the palatal amygdala [4], and sometimes Depending on the size and the anatomical extension of the mass, local symptoms, such as dysphagia, dysphonia, dyspnea, and stridor, may occur [6].

The diagnosis of branchial cleft cyst is made primarily by medical history, clinical manifestations and exclusion. Preoperative ancillary diagnostic procedures include computed tomography, or CT, MRI, sonography and fine-needle aspiration, or FNA.

CT or MRI may be particularly useful not only to visualize the full extent of the lesion, but also to delineate its association with adjacent structures and to differentiate the lesion from other parapharyngeal tumors: a hemangioma, lymphangioma or dermoid cyst, metastatic lymphadenopathy [4,7].

The sonomorphologic findings typically yield a rounded mass that has a uniform low echogenicity lacking internal septation, with no acoustic enhancement or motion [9] as in our case.

FNA can be an important adjunct to clinical diagnosis of lateral neck lesions, especially when attempting to categorize the swelling as benign or malignant [7].

The criteria [10] for FNA 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.

The procedure is quick and findings typically are available in a matter of hours. However, FNA is not a substitute for thorough, microscopic examination of the lesion.20 Branchial cleft cysts may have the ability to become malignant. 21–23 To date, though, there has been no report of such an occurrence. Because of their lymphoid nature, these cysts can be confused with a metastatic lymph node or a primary malignancy from the thyroid gland.21-23.

Histologically, it is well known that cervical lymphoepithelial cyst walls are usually covered with squamous epithelium and/or in some instances with columnar or cuboidal cells. Lymphoid tissue with or without germinal centers in the subepithelial connective tissue are the most prominent morphologic characteristics [11].

Regarding our case the histological examination shows a cystic wall covering of squamous type, often abraded. the underlying chorion is fibrous punctuated by lymphocytes grouping together in a follicle.

Complete surgical resection, through a transverse cervicotomy under general anesthesia is the treatment of choice and results in a good prognosis. Identification, during operation, of the external and internal carotid arteries and the vagus, hypoglossal, glossopharyngeal and superior laryngeal nerves will avoid injury of these structures(6).

Approximately 80% of branchial sinuses will open to the skin, and fewer will open to the pharynx. These sinuses initially may suggest a cyst, but on surgical exploration prove to be sinuses with a tract leading medially. Clinicians must take care to remove the entire tract to decrease the chance of recurrence [12].

Complications of surgical treatment include recurrence, formation of a persistent fistula, and damage to the cranial nerves [6]. Patients with infected cysts receive a full course of antibiotics before surgery to decrease the risk of recurrence and persistent fistula. Alternative treatments, such as percutaneous sclerotherapy, remain unproven [13].

#### 3. Conclusion

The branchial cleft cyst, or cervical lymphoepithelial cyst, is a pathological entity whose etiology has yet to be delineated. It shares a clinical presentation with other pathological entities of the neck, making diagnosis difficult at times. The therapeutic management is always surgical, it must be carried out as soon as possible to limit the risk of inflammatory changes linked to the infectious episodes, it will then be advisable to operate only after complete cooling of the infections by an adapted antibiotic therapy. The recurrence is unlikely if all remnants are removed.

## Ethical approval

I certify that this kind of manuscript does not require ethical approval by the Ethical Committee of our institution.

## **Author contribution**

- Z. Najib: conception and design of the study.
- O. Berrada: conception and design of the study.
- M. Lahjaouj: acquisition of data.
- Y. Oukessou: drafting the article.
- S. Rouadi: drafting the article.
- R. Abada: revising the article.
- M. Roubal: revising the article.
- M. Mahtar: final approval of the version to be submitted.

#### Registration of research studies

This is a Case report that does not require a research registry.

# Guarantor

Zouhair Najib.

#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### **Declaration of competing interest**

The authors declare that they have no competing interests.

# Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2020.12.041.

#### References

- J. Golledge, H. Ellis, The aetiology of lateral cervical (branchial) cysts: past and present theories, J Laryngol Otol. août 108 (8) (1994) 653–659.
- [2] R. Ahmed, H. Bouchaib, E. Noureddine, B. Fouad, Enorme kyste amygdaloïde cervical: à propos d'un cas, The Pan African Medical Journal 31 (2018).
- [3] The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines | the EQUATOR network [Internet]. [cité 23 nov 2020]. Disponible sur, https://www.equator-network.org/reporting-guidelines/the-scare-statement-consensus-based-surgical-case-report-guidelines/.
- [4] A. Charfi, W. Abid, N. Romdhane, A. Madiouni, H. Chahed, R. Zainine, et al., Les kystes amygdaloides: à propos de 31 cas, Journal Tunisien d'ORL et de Chirurgie Cervico-Faciale 31 (2014) 24–26.
- [5] 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.
- [6] V. Thomaidis, K. Seretis, D. Tamiolakis, N. Papadopoulos, I. Tsamis, Branchial cysts. A report of 4 cases, Acta Dermatovenerol. Alpina Pannonica Adriatica 15 (2) (2006) 85–89.
- [7] J.W. Glosser, C.A.S. Pires, S.E. Feinberg, Branchial cleft or cervical lymphoepithelial cysts: etiology and management, J. Am. Dent. Assoc. 134 (1) (2003) 81–86.
- [8] J.R. Chandler, Branchial Cleft Cysts, Sinuses, and Fistulas, 1981.
- [9] M.J. Warren, K. Spencer, M. Nayagam, Sonographic appearance of branchial cysts, Clin. Radiol. 49 (5) (1994) 359–360.
- [10] S. Regauer, M. Gogg-Kamerer, H. Braun, A. Beham, 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) (1997) 623–630.
- [11] J. Hirota, Y. Maeda, E. Ueta, T. Osaki, Immunohistochemical and histologic study of cervical lymphoepithelial cysts, J. Oral Pathol. Med. 18 (4) (1989) 202–205.
- [12] H. Conway, A.P. Jerome, The surgical treatment of branchial cysts and fistulas, Surg. Gynecol. Obstet. 101 (5) (1955) 621.
- [13] J.O. Donegan, Congenital neck masses. Cummings CU, Frederick JM, Harker LA, et al Otolaryngology Head and Neck Sugery 2 (1993) 1554–1565.