UNUSUAL MASSIVE SUBMENTAL EXTENSION OF AN AMYGDALOID CYST

VOLUMINEUSE EXTENSION SOUS MENTALE INHABITUELLE D'UN KYSTE AMYGDALOÏDE

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ABSTRACT

Introduction: Second branchial cleft cysts are rare congenital cervical lesions, accounting for approximately 2% of neck tumors. Submental extension is exceptionally rare and may pose a diagnostic challenge.
Case Report: A 55-year-old man presented with a painless submental swelling that had progressively enlarged over five years. Imaging revealed a well-defined cystic lesion arising from the left submandibular region with marked submental extension. Complete surgical excision was performed through a cervicotomy. Histopathological examination confirmed a second branchial cleft cyst.
Conclusion: This rare presentation should be considered in the differential diagnosis of submandibular cystic masses. Complete surgical excision is both diagnostic and curative.
Keywords: Second branchial cleft cyst, Stypical location, Submental extension, Cervicotomy.

RÉSUMÉ

Introduction: Les kystes de la deuxième fente branchiale sont des lésions cervicales congénitales rares, représentant environ 2 % des tumeurs cervicales. Leur extension sous-mentonnière est exceptionnelle et peut poser un défi diagnostique.
Observation: Il s'agit d'un homme âgé de 55 ans qui a consulté pour une tuméfaction sous-mentonnière indolore, augmentant progressivement de volume depuis cinq ans. L'imagerie a objectivé une lésion kystique bien limitée, développée à partir de la région sous-mandibulaire gauche, avec une importante extension sous-mentonnière. Une exérèse chirurgicale complète a été réalisée par cervicotomie. L'examen anatomopathologique a confirmé le diagnostic de kyste de la deuxième fente branchiale.
Conclusion: Cette présentation rare doit être prise en compte dans le diagnostic différentiel des masses kystiques sous-mandibulaires. L'exérèse chirurgicale complète est à la fois diagnostique et curative.
Mots-clés: Kyste de la deuxième fente branchiale, Localisation atypique, Extension sous-mentonnière, Cervicotomie.

INTRODUCTION

The amygdaloid cyst, also known as a second branchial cleft cyst, is a benign congenital malformation presenting as a cystic mass in the anterolateral region of the neck. Although it is a rare congenital anomaly, it accounts for up to 85% of second branchial cleft anomalies[1]. The amygdaloid cyst typically develops along the middle third of the anterior border of the sternocleidomastoid muscle. However, they may occur anywhere from the middle pharyngeal constrictor muscle to the supraclavicular region[2]. Certain atypical locations can complicate the diagnosis and mimic other cystic, tumor, or infectious conditions.

We present a case of an amygdaloid cyst with a massive submental extension to emphasize the clinical and radiological characteristics of this unusual presentation, as well as its therapeutic management.

Case Presentation

A 55-year-old man, with no significant past medical or surgical history, presented with a progressively enlarging submental swelling that had evolved over a period of five years without associated pain or functional symptoms. Physical examination revealed a submental swelling extending to the left submandibular region, measuring 10 cm in its largest diameter. The tumefaction was painless, slightly mobile, non-pulsatile, of renitent consistency, and covered with normal skin. The oropharynx, nasopharynx and pharyngolarynx were examined and found to be normal.

Cervical ultrasound demonstrated a deep cystic lesion with hyperechoic content in the submental region that mesured 10 × 5 cm. Contrast-enhanced cervicofacial computed tomography showed a well-defined cystic lesion that occupies the left submandibular and submental regions, located between the genioglossus and mylohyoid muscles. The lesion measured 86 × 62 × 61 mm and exerted a mild mass effect on the upper airway (Figure 1).

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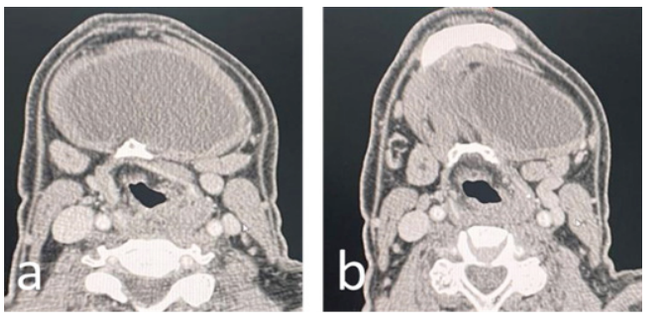


Figure 1: Axial contrast-enhanced CT images showing a large submental cystic mass (a) with extension to the left submandibular region (b).

Given its predominantly midline location, a dermoid cyst and a thyroglossal duct cyst were initially considered in the differential diagnosis. The patient underwent cervicotomy under general anesthesia. Intraoperative exploration revealed a large cystic lesion located medial to the mylohyoid muscle, with its posterior wall densely adherent to the body of the hyoid bone. As this intimate attachment prevented safe dissection from the hyoid without compromising the completeness of excision, en bloc resection of the cyst together with the body of the hyoid bone was performed to ensure complete surgical clearance (Figure 2). The postoperative course was uneventful.

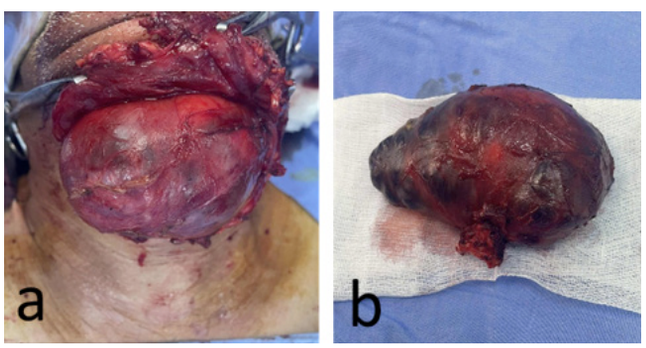


Figure 2: (a) Intraoperative view showing the partially dissected cyst; (b) completely excised cyst.

Histopathological examination confirmed the diagnosis of an amygdaloid cyst without evidence of malignancy. One-year postoperative follow-up did not show evidence of local recurrence.

DISCUSSION

The second branchial cleft cysts are the result of abnormal branchial apparatus development occurring between the fourth and seventh weeks of embryonic life [2,3]. They arise secondary to incomplete involution of the cervical sinus of His [1]. The amygdaloid cyst usually develops in the anterolateral region of the neck. Communication with the pharynx or skin may occur spontaneously or following superinfection. The external opening is generally located at the junction between the middle and lower thirds of the anterior border of the sternocleidomastoid muscle. Communication with the pharynx, less common than external fistulization, occurs through an internal fistula opening into the tonsillar region [1,4].

In 1929, Bailey classified second branchial cleft cysts into four types [3]:

- Type I: superficial cyst located under the superficial cervical fascia, anterior to the sternocleidomastoid muscle.
- Type II: the most common type, located under the middle cervical fascia, prevascular, anterior and lateral to the great vessels.
- Type III: intervascular cyst located between the internal and external carotid arteries, extending to the lateral pharyngeal wall.
- Type IV: deep cyst located medial to the carotid sheath within the pharyngeal mucosal space, opening into the pharynx.

Amygdaloid cysts may be present at birth, but are most commonly diagnosed during the third or fourth decade of life, usually after an infectious episode leading to enlargement of the lesion [3].

Clinically, second branchial cleft cysts typically present as a painless, slowly enlarging lateral cervical mass in the jugulocarotid region. They are usually unilateral [3–5].

However, atypical locations, particularly parapharyngeal or tonsillar localizations, may occur and make the diagnosis more difficult [6,7]. In our case, the patient presented an unusual massive anterior submental extension. Such atypical locations may be explained by variations in the embryological development of the cervical sinus and the tract of the second branchial arch [7,8].

Imaging is useful in diagnosis. Cervical ultrasound is the first-line imaging modality and helps confirm the cystic nature of the lesion. Computed tomography provides an accurate assessment of the lesion's location and its anatomical relationships, especially with the vascular axis. Magnetic resonance imaging is particularly useful in atypical forms and allows better evaluation of deep tissue extension [2,4,5,9].

The main differential diagnoses include metastatic lymphadenopathy, lymphangioma, and dermoid cysts. In adults, any lateral cystic neck mass should initially raise suspicion of metastatic lymphadenopathy. Clinical examination must systematically look for suspicious lesions within the upper aerodigestive tract, particularly in the oropharynx and nasopharynx [2,10,11]. Skalias et al. reported a case of papillary carcinoma discovered after metastasis within a branchial cleft cyst [12]. Due to its predominantly midline location, our case also raised suspicion of a thyroglossal duct cyst.

Treatment is based on complete surgical excision. The procedure consists of the total removal of the cyst. When a fistulous tract is present, it may be identified by catheterization and methylene blue injection. The dissection is then continued along the fistulous tract. It is rapidly terminated in the cases of blind external fistula. If the fistula continues upwards, a second incision will be required to follow its course [5,6,9,13]. Tonsillectomy is not routinely indicated [13,14].

In cases of infection, prior antibiotic therapy is recommended and surgery should be delayed until the acute inflammatory episode is resolved to facilitate complete excision and reduce the risk of recurrence [9]. The prognosis is generally favorable, with a low recurrence rate after complete excision (0–6%) [6,9,13].

CONCLUSION

The amygdaloid cyst should be considered in the differential diagnosis of a submandibular cystic mass with marked submental extension. Complete surgical excision is both diagnostic and curative. Histopathological examination remains essential for a definitive diagnosis.

Statements and declarations:

Ethical Consideration:

This study is exempted from ethical approval at our institution.

Consent:

Written informed consent for publication of their clinical details and images was obtained from the patient

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REFERENCES:

1. Shen LF, Zhou SH, Chen Q qiong, Yu Q. Second branchial cleft anomalies in children: a literature review. Pediatr Surg Int. déc 2018;34(12):1251-6. doi:10.1007/s00383-018-4348-8
2. Adams A, Mankad K, Offiah C, Childs L. Branchial cleft anomalies: a pictorial review of embryological development and spectrum of imaging findings. Insights Imaging. 10 déc 2015;7(1):69-76. doi:10.1007/s13244-015-0454-5 PubMed PMID: 26661849; PubMed Central PMCID: PMC4729717.
3. Prescher H, Nathan SL, Bauer BS, Reid RR. Branchial Cleft Anomalies: Embryogenesis, Clinical Features, Diagnosis, and Management. FACE. mars 2022;3(1):128-37. doi:10.1177/27325016211072292
4. Bagchi A, Hira P, Mittal K, Priyamvara A, Dey AK. Branchial cleft cysts: a pictorial review. Pol J Radiol. 11 mai 2018;83:204-9. doi:10.5114/pjr.2018.76278
5. Alshihmani SHA. A second branchial cleft cyst, a case report. International Journal of Surgery Case Reports. juill 2023;108:108429. doi:10.1016/j.ijscr.2023.108429
6. Charfi A, Abid W, Romdhane N, Madiouni A, Chahed H, Zainine R, et al. LES KYSTES AMYGDALOIDES : A PROPOS DE 31 CAS SECOND BRANCHIAL CLEFT CYST : ABOUT 31 CASES. 2014.
7. Choo MJ, Kim YJ, Jin HR. A Case of Second Branchial Cleft Cyst with Oropharyngeal Presentation. J Korean Med Sci. 2002;17(4):564. doi:10.3346/jkms.2002.17.4.564
8. Howlett J, Horwich P, Bullock MJ, Taylor SM, Hart R, Trites J, et al. An unusual presentation of a branchial cleft cyst in a 70-year-old. Otolaryngology Case Reports. mars 2018;6:16-8. doi:10.1016/j.xocr.2017.11.005
9. Bellakhdhar M, Haouas J. Les kystes amygdaloïdes: a propos de 34 cas Second branchial cleft cyst: about 34 cases. LA TUNISIE MEDICALE. 2018;96.
10. Liao YC, Adel M, Lee LY, Chang KP. Branchial cleft cyst: An unusual site for the cervical metastasis of nasopharyngeal carcinoma. Auris Nasus Larynx. avr 2018;45(2):328-31. doi:10.1016/j.anl.2017.03.021
11. Liang HH, Chen CY, Chen WY, Chen TM, Chan WP. Solitary cystic metastatic lymph node of occult human papillomavirus-related oropharyngeal cancer mimicking second branchial cleft cyst: A case report. Medicine. nov 2019;98(44):e17800. doi:10.1097/MD.0000000000017800
12. Skalias A, Garefis K, Tsetsos N, Goudakos J. Papillary Thyroid Carcinoma within a Second Branchial Cleft Cyst. A Diagnostic and Treatment Challenge. Maedica. 15 sept 2024;19(3). doi:10.26574/maedica.2024.19.3.648
13. Reddy A, Valika T, Maddalozzo J. Definitive surgical management for second branchial cleft fistula: A case series. Journal of Otolaryngology - Head & Neck Surgery. janv 2020;49(1):55. doi:10.1186/s40463-020-00453-2
14. Cheng J, Elden L. Management of pediatric second branchial fistulae: Is tonsillectomy necessary? International Journal of Pediatric Otorhinolaryngology. nov 2012;76(11):1601-3. doi:10.1016/j.ijporl.2012.07.024