

First Cases of Amygdaloid Cyst in Adults in Djibouti: Case Report of Two Patients

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Abstract

Amygdaloid cysts are rare cystic benign tumors due to congenital malformations resulting from an anomaly of embryonic development of the upper laterocervical region, originating from the second branchial cleft. They represent approximately 2% of all laterocervical tumors and 6% to 85% of anomalies of the second branchial cleft. This anomaly of the second branchial cleft is a frequent reason for consultation in the pediatric population but is relatively rare in adults. We report the cases of two patients aged 23 and 34 years with no particular pathological history. They were presenting a laterocervical swelling, one right and the other left, painless, evolving for two years for the first and for 10 years for the second, gradually increasing in volume without any other associated signs the diagnosis of which after radiological exploration (ultrasound and CT scan) was that of an amygdaloid cyst. A cervicotomy with anatomopathological examination of the surgical specimen confirmed the diagnosis of the amygdaloid cyst. The objective is to analyze the anatomic-clinical and therapeutic particularities of this pathology and to compare it with data from the literature.

Keywords

Amygdaloid Cyst, Second Branchial Cleft, Surgery, Djibouti

1. Introduction

Amygdaloid cysts 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. These are

malformations resulting from an anomaly during embryonic development. The cervicofacial region arises from the evolution of the branchial arches. Knowledge of embryology allows understanding of their topography. These cystic malformations represent a frequent reason for consultation in pediatrics but they can also be found in adults. The amygdaloid cyst is one of the most frequent branchial anomalies, in particular up to 85% of those anomalies originate from the second branchial arch [1] [2]. The circumstances of discovery are of two kinds, either discovery of a cervical swelling and/or a fistulous orifice chronic skin disease, or during an inaugural complication, with discovery of a cervical cystic swelling after an acute pharyngeal infectious episode. We report the cases of two observations of patients who were treated for a laterocervical amygdaloid cyst in adults in our department to analyze the anatomo-clinical and therapeutic particularities of this pathology and compare it with data from the literature.

2. Cases Reports

Case 1

- **Patient information:** Mrs. M.D. is a 23-year-old patient with no particular pathological history received in our department for a right laterocervical swelling evolving for 2 years and increasing in volume gradually without other associated signs for which she consulted a pulmonologist one month before her consultation in our department. She benefited from an evacuating puncture. And the puncture fluid was examined with the Genexpert. In front of the negative result and the resumption of the swelling, she was sent to us for a better exploration of the ENT sphere.
- **Clinical outcomes:** The general examination at the consultation found a patient in good general condition. Well colored integuments and conjunctive. General parameters were: temperature: 37 degrees Celsius; pulse: 76 BPM; BP: 10/07; SaO₂: 99% in ambient air; On examination of the cervico-facial skin, there was a right laterocervical swelling jugulo-carotid high under digastric, painless on palpation, mobile, of soft consistency, estimated about 4 cm long axis with healthy facing skin. (Figure 1)

Examination of the oral cavity and oropharynx was normal. Otoscopy and anterior rhinoscopy was normal. Examination of lymph node areas was also normal. Elsewhere the remainder of the patient's physical examination was unremarkable.

- **Diagnostic approach:** at the end of the clinical examination, the diagnosis of a cervical cyst was suspected. Cervical ultrasound showed an anechoic formation with a high right posterior laterocervical reinforcement, with regular contours and a volume of 22.8 ml. (Figure 2)

The cervical computed tomography (CT) performed revealed a heterogeneous cystic formation exerting a mass effect on the vascular axis of the neck in favor of a second branchial cleft cyst. (Figure 3, Figure 4)

A preoperative blood tests including complete blood count (CBC), prothrom-

bin level (TP) and rhesus blood group. The results of these assessments were normal: CBC: HB: 12 g/dl, WBC: 4000/mm³ and platelets at 350,000/mm³. The PT was 98% and the blood group was group a rhesus positive.



Figure 1. Photo of patient showing right laterocervical mass.



Figure 2. Ultrasound image showing anechoic cystic formation.

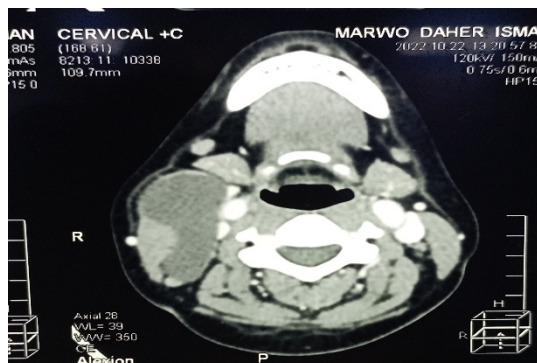


Figure 3. Cervical CT in axial section: well-limited cystic formation with contrast uptake in the periphery.

The patient was scheduled for surgery after having benefited from a preoperative assessment and a pre-anesthesia consultation.

Therapeutic intervention and follow-up: The diagnosis of a cervical cyst was retained and the patient underwent right cervicotomy with complete excision of the cyst. The postoperative course was simple. Histological examination confirmed the diagnosis of an amygdaloid cyst without signs of malignancy. (Figures 5-7)

Postoperative evolution: With a follow-up of 05 months after the surgical operation, we detected no recurrence.

Case 2:

- **Patient information:** Mrs. M.M.A. is a 34-year-old patient without particular pathological history received in our department for a left laterocervical swelling evolving for 10 years and increasing in volume gradually without other associated signs.
- **Clinical outcomes:** The general examination at the consultation found a patient in good general condition. Well colored integuments and conjunctivae.

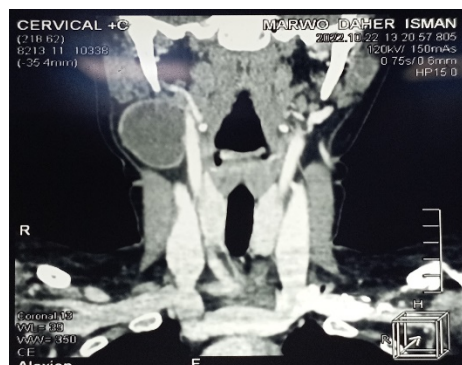


Figure 4. Cervical CT in coronal section: well-defined cystic formation in close contact with the vascular structures of the neck.

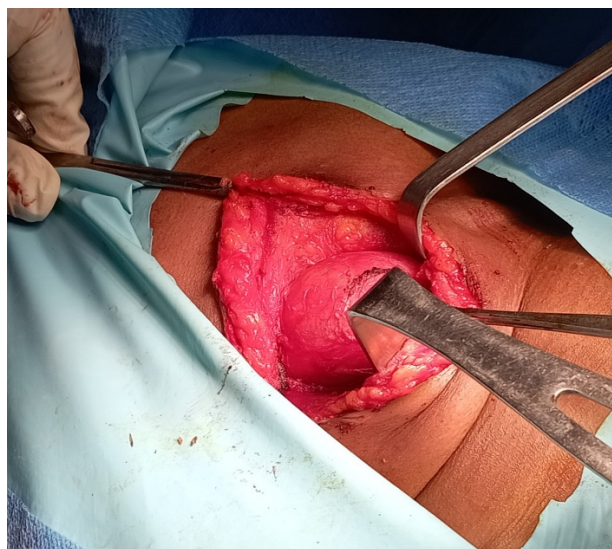


Figure 5. Photo showing the cyst during intraoperative exposure.



Figure 6. Photo of the surgical specimen after excision.

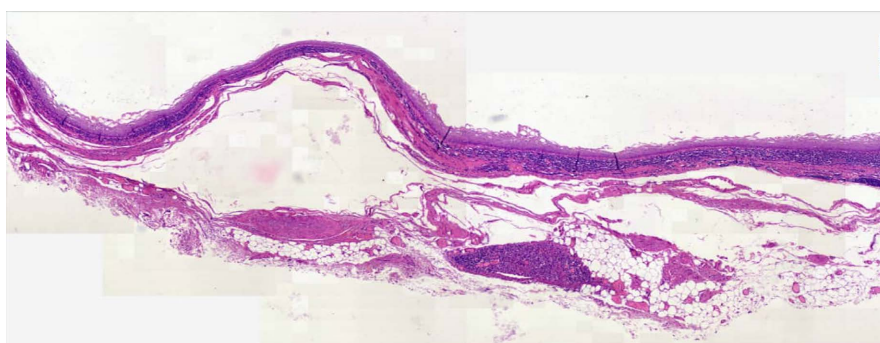


Figure 7. Histological appearance of the second branchial cleft cyst. (10×) Hematoxylin and eosin.

General parameters were: temperature: 36.8 degree Celsius; pulse: 80 BPM; BP: 12/07; SaO₂: 99% in ambient air;

Examination of the cervicofacial skin found a left laterocervical swelling in the upper jugulo-carotid sub-digastric region, painless on palpation, mobile, of soft consistency, measuring approximately 6 cm on the long axis with healthy facing skin.

Examination of the oral cavity and oropharynx was normal. Otoscopy and anterior rhinoscopy were normal. Examination of lymph node areas was also normal. Elsewhere the remainder of the patient's physical examination was unremarkable.

- **Diagnostic approach:** At the end of the clinical examination, the diagnosis of a cervical cyst was suspected. The cervical computed tomography (CT) performed revealed a left cervical cystic formation in favor of a second branchial cleft cyst. (**Figure 8**)

A preoperative blood tests including complete blood count (CBC), prothrombin level (TP) and rhesus blood group. The results of these assessments were normal: CBC: HB: 11 g/dl, WBC: 7000/mm³ and platelets at 288,000/mm³. The PT was 87% and the blood group was group AB rhesus positive.

However, the patient was scheduled for surgery after having benefited from a

preoperative assessment and a pre-anesthesia consultation.

- **Therapeutic intervention and follow-up:** The diagnosis of a cervical cyst was retained and the patient underwent left cervicotomy with complete excision of the cyst. The postoperative course was simple. Histological examination confirmed the diagnosis of an amygdaloid cyst without signs of malignancy. (**Figure 9, Figure 10**)

Postoperative evolution: with a follow-up of 01 year after the surgical operation, we detected no recurrence.

Patients' point of view: Since their first days of consultation in our service and throughout the duration of the management of their pathology, the patients were satisfied with the care they received and were optimistic about the evolution of

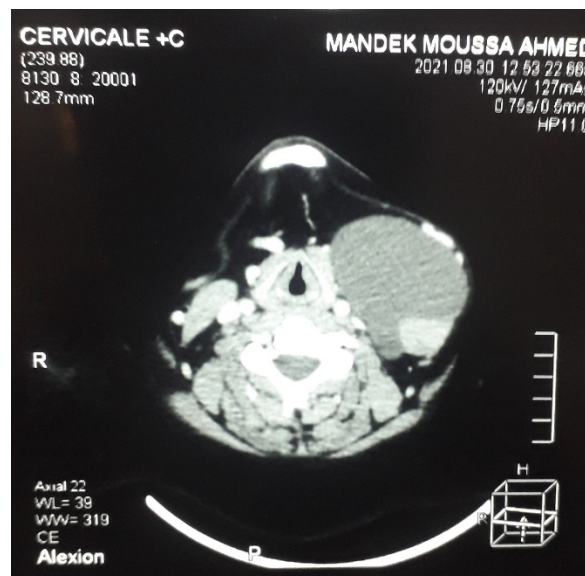


Figure 8. Axial CT of the neck: well-limited cystic formation on the left neck.



Figure 9. Photo showing the cyst after exposure.



Figure 10. Photo of the operative part after its complete excision. The cyst measures approximately 05 cm from axis macroscopically.

their condition.

Patient information and informed consent: Patients were informed of the authors' interest in publishing their clinical cases. They voluntarily agreed that we can use their photos for these cases reports.

3. Discussion

Cysts and fistulas of the neck are congenital malformations resulting from abnormal embryonic development in the cervical region. They are common in children but can occur in adults. Anomalies of the second cleft most often sit next to the anterior edge of the sternocleidomastoid muscle at the level of its middle third at the height of the hyoid bone. Amygdaloid cysts are among the most frequent anomalies of the second branchial arch, up to 85% of cases [1] [2]. Although it is a congenital anomaly, these cysts of the second branchial arch in adults are discovered late during the 2nd and 3rd decades of life without any sexual predominance. The circumstances of discovery are made during an episode of superinfection or on the occasion of signs of compression linked to the increase in the volume of the cyst [3] [4]. Clinically, the cyst presents as a renitent laterocervical swelling, tense, not very sensitive to palpation apart from an episode of superinfection with healthy-facing skin. This cyst can sometimes communicate internally at the level of the tonsillar region or abut directly with the skin, thus becoming an external cervical fistula. The fistula passes through the carotid bifurcation between the internal carotid and the external carotid [5]. These cysts were classified in 1929 by BAILEY into four subtypes [6]:

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, adja-

cent 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.

Nevertheless, the observation of a fistula of the second branchial cleft, especially when it is bilateral, should lead to a search for the branchio-oto-renal syndrome. The major challenge in the management of this cervical cyst is to think about eliminating the differential diagnoses that can evoke all the causes of chronic cervical masses in adults. These differential diagnoses include a lymphadenopathy which may reveal cancer of the upper aerodigestive tract, tuberculous adenitis, cryptic metastasis of tonsillar carcinoma, or cervical lymphoma. These multitudes of differential diagnoses make the indication of anatomopathological examination essential [2] [5] [7]. On the radiological level, ultrasound remains the first additional examination to be requested in the face of a cervical mass because it is a non-invasive examination, easy to perform, inexpensive and above all does not expose the patient to ionizing radiation. It allows to appreciate the echostructure of the palpated mass, to specify the existence or not of the vascularization within the mass by its Doppler mode. It also makes it possible to specify the anatomical location of the mass in relation to the vascular structures of the neck (carotid, jugular) or in relation to the muscular structures (sternocleidomastoid muscle) [8]. However, when a mass is large enough and the delimitation cannot be done by ultrasound, the use of cross-sectional imaging, in particular CT and Magnetic resonance imaging (MRI), is necessary to better study the mass. On computed tomography the cysts generally appear well circumscribed, uniformly hypodense with thin walls. However, the thickness of the wall may increase after superinfection of the cyst. As for MRI, it gives a better representation of the cyst and its extent and thus gives a better idea and precise in preoperative. Cyst contents vary from hypo to isointense (relative to muscle) in T1 sequences and hyperintense in T2 sequences. In case of presence of fistula, fistulography is necessary and it allows to clearly visualize the location and extent of the fistulous path [2] [9]. After clinical and radiological suspicion, the diagnosis of amygdaloid cysts is confirmed by histology. Fine-needle aspiration biopsy can be an important complement to clinical diagnosis in preoperative. The cytological criteria for bronchial cysts are [10]:

- The presence of a thick, yellow liquid resembling pus;
- The presence of anuclear keratinizing cells;
- The presence of squamous epithelial cells of variable maturity;
- The presence of a bottom of amorphous debris.

Histologically, the walls of cervical tonsilloid cysts are lined by a stratified squamous epithelium, but which can sometimes be pseudostratified, ciliated. Lymphoid tissues with germinal centers are often contained in the subepithelial connective tissue and constitute the most important morphological characteris-

tics [11]. Although rare, malignant intracystic degeneration exists with an incidence estimated at 4% to 22% according to studies. However, the diagnosis of primary carcinomatous degeneration of the cyst and cervical intracystic metastasis are still controversial and the greatest challenge remains to make their distinctions [12]. Therapeutically, the treatment of amygdaloid cysts is based on surgery. This surgical treatment consists in carrying out the complete excision of the cystic pocket but also the excision of the external cutaneous orifice of the fistula as well as its path if the latter exists. This so-called conventional surgical approach to tonsilloid cysts consists of making a skin incision 5 to 7 cm in length, along the lower mandible 2 to 2.5 cm below. Then the dissection is made plan by plan through the subcutaneous tissue, the platysma, the superficial cervical fascia to expose the fascia of the cyst. After the exposure of the cyst, the excision is done gradually while respecting the surrounding anatomical structures, hence the anatomical knowledge of the surrounding vessels and nerves is necessary for a safe and complete resection. If the fistulous path is located, the dissection continues in contact with the fistulous path. However, some authors recommend making a vertical incision along the anterior border of the sternocleidomastoid muscle [13].

Over the past two decades, several operative techniques have been developed for the surgical excision of second branchial cleft cysts and thus become an alternative to the so-called conventional surgical approach, in particular robot-assisted endoscopic surgery [13] [14]. The postoperative evolution is generally good, especially when the surgery is performed in a regulated program. However, some authors have reported complications 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 [9]. Recurrence rates after primary surgery are estimated at 3% - 4%. We did not observe any recurrence in our patients [3]. The degeneration of the amygdaloid cyst is exceptional and estimated at 4% - 22% according to studies. Its diagnosis must be retained after eliminating a metastasis within the cyst or a cystic lymph node metastasis from a distant primary carcinoma. Its treatment is based on complete excision of the cyst associated with cervical lymph node dissection followed by adjuvant radiotherapy [3] [12].

4. Conclusion

Amygdaloid cysts are relatively rare malformations in adults. They are manifested by a laterocervical swelling and their diagnosis is based on a range of clinical and radiological arguments, and confirmed by anatomopathological examination. Treatment is based on complete surgical excision. The difficulty of diagnosing this pathology in Djibouti lies in its presentation, which can be suggestive of other pathologies like tubercular lymph node because Djibouti remains a country where tuberculosis is endemic. This is the case of our patient No. 1 who was first treated for tuberculosis lymphadenopathy. The fear is also to miss the

diagnosis of an intracystic metastasis or a cystic lymph node metastasis from a distant primary carcinoma.

Authors Contributions

- Data collection: Abdallah Wittti ; Kamil Ahmed Kamil.
- Data analysis and interpretation: Abdallah Wittti, Awaleh Ahmed Awaleh, Kamil Ahmed Kamil.
- Writing of the article: Abdallah Wittti.
- Revision of the article: Abdallah Wittti, Awaleh Ahmed Awaleh, Kamil Ahmed Kamil.
- All authors have read and approved the final handwritten version.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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