# Branchial cysts.
## A report of 4 cases

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ABSTRACT

***Objective***. Cysts presented in the lateral aspect of the neck are relatively uncommon anomalies. Many theories have been proposed to explain the aetiology of these cysts, grouped in two main categories: the congenital and the cervical lymph nodes cystic transformation hypotheses.

***Settings***. In this paper we present 4 patient-cases documented in the department of Maxillofacial Surgery, and try to trace the profile of these lesions and investigate their origin.

***Results***. All lesions were well circumscribed by Computed Tomography, and prior to surgery they were assessed by means of fine needle aspiration cytology. Neutrophils, debris and mature squamous epithelial cells including degenerate forms, were the key-features in the cytological diagnosis. Histologic examination of the excised surgical specimen established the diagnosis in all cases.

***Conclusions***. Our findings, compatible with the congenital theory lead us to the conclusion that the branchial cysts are the result of imperfect obliteration of the branchial clefts, arches, and pouches.

KEY WORDS

**lateral cervical cysts, branchial cysts**

## Introduction

Cysts presented in the lateral aspect of the neck were first described by Hunczovsky (1). Since then, a variety of names has been ued for these cysts: dermoid cyst of the sheath of the internal jugular vein (2), deep-seated atheromatous tumour (3), congenital hydrocoele of the neck (4), hygroma colli (5), branchial cyst (6), tumour of the branchial cleft (7), lateral lympho-epithelial cyst (8) and benign cystic lymph nodes (9).

The most widely held belief is that cervical cysts are derived from the branchial apparatus (10). According to the branchial theory the lateral cervical cysts are the result of imperfect obliteration of the branchial clefts, arches, and pouches. Ascherson (6), in 1832, was the first investigator who described 11 cases of branchial fistulae, equating the development of lateral cervical cysts with that of branchial fistulae due to their location. He suggested that incomplete obliteration of branchial cleft mucosa, which remains dormant until stimulated to grow later in life, result in cyst formation. Since then, a number of investigators re-emphasized this link between the pharyngeal arches and cervical cysts (2, 11-14).

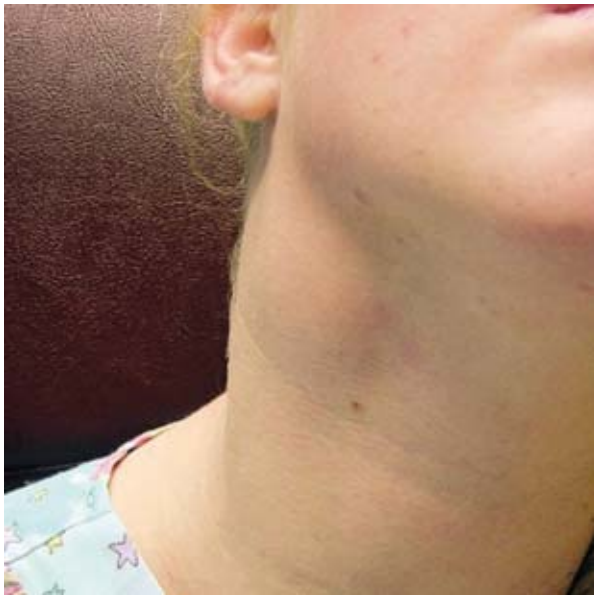

**Figure 1. Case 1: Clinical appearance of a right sided branchial cleft cyst.**

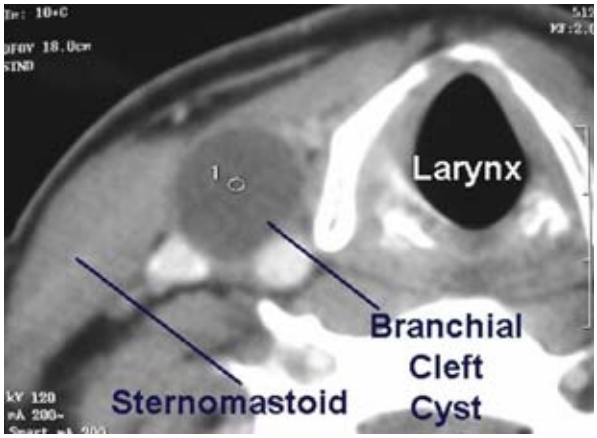


**Figure 2. Case 1: Typical appearance of the lesion demonstrated by a CT scan.**

A second theory was suggested by His (15) considering that branchial fistulae are vestiges of the cervical sinus rather than of the pharyngeal clefts or pouches. Parallel to the branchial theory, this precervical sinus theory was extended to include lateral cervical cysts.

Wenglowski (16) suggested an entirely different hypothesis based on human embryo and cadaver studies. He showed that pharyngeal cleft tissue was not represented in any adult structure inferior to the hyoid bone. Thus any cyst lying below this level could not be derived from a pharyngeal cleft. Wenglowski went on to describe the development of the thymus from the third pharyngeal pouch via the thymopharyngeal duct. He suggested that incomplete obliteration of the thymopharyngeal duct resulted in a lateral cervical cyst. However, a major problem with the thymopharyngeal duct theory is that more anomalies are to be found at the lower end of the embryonic thymopharyngeal duct, that is, at the lower end of the neck and mediastinum rather than cephalad near the hyoid bone.

An alternative theory suggests that lateral cervical cysts represent cystic lymph nodes. Lucke (17) noted that the external appearance of a lateral cervical cyst showed a great similarity to a hypertrophied lymphatic gland. Luschka (5) described a lymph node between the external and internal carotid arteries (ganglion caroticum). He suggested that cystic degeneration of cervical lymph nodes was the mechanism by which lateral cervical cysts were formed. However, it was not until the work of King (8) that the Lymph Node Theory received much support. King studied 76 cervical cysts and concluded that they had no direct relationship with any structure in the embryo. Instead he emphasized the close relationship between these cysts and lymphoid tissue. Further support for this theory was provided by Bhaskar and Bernier (18), who reviewed the histology of 468 cysts: 452 had a wall composed of lymphoid tissue. On the basis of this, they concluded that cervical cysts developed as a result of cystic transformation of cervical lymph nodes. Furthermore, they suggested that the cystic alteration of cervical lymph nodes is stimulated by trapped epithelium. This hypothesis has become known as the "Inclusion Theory". They originally suggested three possible sources for these epithelial inclusions: brachial cleft, pharyngeal pouch and parotid gland. More recent reviews (19, 20) revealed similar histological findings. However, it has been pointed out that cystic transformation of lymph nodes is not known to occur in other anatomical sites (19).

At present, to clarify the origin of these cysts, immunohistochemistry is being used to study, at the molecular level, the nature of the epithelial cells and their rela-

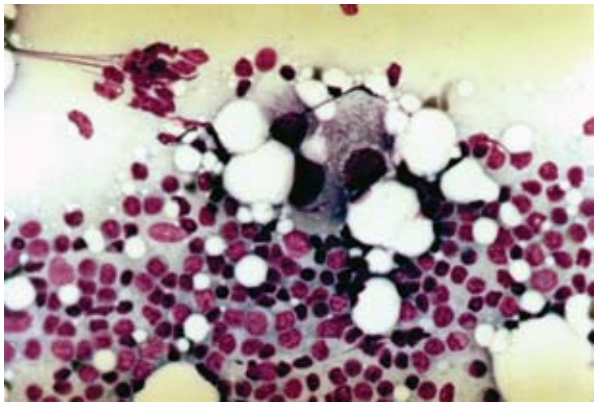

**Figure 3. Cytological appearance of the branchial cleft cyst ( Lymphoid tissue) - FNA ( May-Grunwald-Giemsa stain X 400).**

tionship to other types of epithelium elsewhere in the body.

## Case series

### Case 1

A 30-year-old woman was admitted to the department of Maxillofacial Surgery, with an asymptomatic mass on the right side of the neck (Figure 1). The mass had first appeared 2 years ago and had increased in size very slowly. On physical examination, a well circumscribed and mobile mass, located close to the anterior border of the sternocleidomastoid muscle was revealed. On computed tomographic (CT) examination a large cystic structure, was detected (Figure 2), while Fine Needle Aspiration (FNA) findings were consistent with branchial cyst. Upon the preoperative diagnosis of branchial cyst surgery was performed, and a cystic lesion, lying anterior to the sternocleidomastoid muscle and in contact with internal jugular vein was excised. During the operation no tract or cord connecting the cyst to the pharynx was noted.

The histological examination revealed a cystic wall lined with stratified squamous epithelium, while lymphoid follicles were present outside the epithelial lining. The patient, 5 years later still remains healthy.

### Case 2

A 34-year-old woman was referred to our department for investigation and treatment of a slowly enlarging mass on the left side of the neck. The swelling which appeared 3 years earlier progressed gradually to its current size. The examination revealed a mobile, non-tender mass, located close to the anterior border of the sternocleidomastoid muscle, and the CT-scan demonstrated a cystic lesion. FNA examination was indicative of branchial cyst. The lesion was excised through a wide, transverse cervicotomy under general anaesthesia, while at the same time no tract or cord was noted. Histopathologic evaluation documented a second branchial cleft cyst. The patient had an uneventful postoperative course.

### Case 3

A 27-year-old man was referred for assessment and treatment of a lump on the right side of the neck. The patient had noticed a slow growing mass at his neck for two years prior to his admission. Physical examination showed a 3x4 cm movable, painless mass of the carotid triangle. A CT-scan was obtained disclosing a well circumscribed cystic lesion, close to the great vessels, while FNA disclosed a branchial cyst. The mass was excised through a transverse cervicotomy. Histological examination disclosed a second branchial cleft cyst, with its wall lined with stratified squamous epithelium, while lymphoid follicles were present outside the epithelial lining. The patient, 3 years postoperatively, has an uneventful course.

### Case 4

A 51-year-old man was admitted to our department with an asymptomatic mass, located on the left side of his neck. The time period from the initial presentation to admission was 8 years. On examination an asymptomatic, movable mass at the anterior border of the sternocleidomastoid muscle, between its upper and middle third was present. The CT scan revealed a cystic lesion. The patient denied FNA. On the basis of these clinical and radiological findings, the preoperative diagnosis of a lateral cervical cyst was made. At the operation a cystic lesion was found under the platysma, anterior to the sternocleidomastoid, in contact with the internal jugular vein, as well as the extremely rare internal tract. The internal tract extended from the anterior pole of the cyst to the lateral pharyngeal wall, inferior to the posterior belly of the digastric muscle, and was excised at its superior point, with the cyst. Histopathology confirmed a second branchial cleft cyst.

## Discussion

Cysts on the lateral side of the neck are relatively common anomalies.

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 (21). Males and females are equally affected and there is occasionally a hereditary tendency (22). Cysts, may be bilateral, are slow-growing and have a duration of weeks to many years. The lesion, usually, is presented as an asymptomatic circumscribed movable mass, usually close to the anterior border of the sternocleidomastoid muscle. Depending on the size and the anatomical extension of the mass, local symptoms, such as dysphagia, dysphonia, dyspnea, and stridor, may occur.

Cysts may vary in size and may fluctuate. Infected cysts may develop into abscess, especially during periods of upper respiratory tract infections, due to the lymphoid tissue located beneath the epithelium. Spontaneous rupture of an abscessed branchial cleft cyst may occur, resulting in a purulent draining sinus to the skin or the pharynx.

Larger cysts may displace the sternocleidomastoid muscle posterolaterally, and the carotid and internal jugular vein medially. With any further increase in size of

the cyst, the deep cervical fascia prevents its expansion between the sternocleidomastoid muscle and the strap muscles of the larynx. As a result, the cyst takes the path of least resistance and extends posterior and medial to the sternocleidomastoid. The preoperative diagnosis is mainly based on clinical and radiological criteria, which also define the topographic relationship. The precise location and course of the cervical cysts depend on the particular branchial pouch or cleft from which they are derived.

The second branchial cleft cysts account for up to 90% of branchial anomalies. Most frequently, these cysts are identified along the anterior border of the upper third of the sternocleidomastoid muscle. However, these cysts may develop anywhere along the course of a second branchial fistula, extending from the skin to the lateral neck, between the internal and external carotid arteries, and into the palatine tonsil. Therefore, the second branchial cleft cyst is in the differential of a parapharyngeal mass. Proctor (23) classified the second cleft cysts

Complete surgical resection, through a wide, transverse cervicotomy under general anaesthesia is the treatment of choice and results in a good prognosis. Identification, during operation, of the internal and external carotid arteries and the vagus, hypoglossal, glossopharyngeal and superior laryngeal nerves will avoid injury of these structures. Complications of surgical treatment include recurrence, formation of a persistent fistula, and damage to the cranial nerves (24). 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 (25).

On histopathologic examination (26) most branchial cleft cysts are lined with stratified squamous epithelium and contain a brownish fluid with crystals of cholesterol. Occasionally the cyst is lined with respiratory (ciliated columnar) epithelium. The wall may contain lymphoid tissue arranged in a follicular pattern. In infected or ruptured lesions, inflammatory cells are seen within the cyst cavity or the surrounding stroma.

FNA is a very helpful tool in the preoperative diagnosis: The criteria (27) 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.

FNA smears sometimes show a large number of acute inflammatory cells. A component of multinucleate giant cells, representing a granulomatous reaction at the edge of the cyst, may be seen. Lymphoid tissue, although usually evident in tissue sections, is sometimes represented in smears (Figure 3).

To detect the origin of these cysts, immunohistochemistry, particularly the cytokeratin expression profile (28) is being used. At the molecular level the nature of epithelial cells and their relationship to other types of epithelium are also studied. It is assumed that the respiratory epithelium is the native epithelium of the cysts and squamous metaplasia results from inflammation induced stem cell hyperplasia in respiratory epithelium evidenced by co-expression of simple cytokeratins (CKs 7, 8, 18) and stratified-epithelial type cytokeratins (CKs 1, 10, 13).

Lateral cysts of the neck containing malignant epithelium represent a diagnostic and therapeutic challenge. Much attention in the literature has been focused on the frequent relationship between a solitary cystic cervical metastasis and an occult primary tumour in the tonsil or the tongue base. It has been suggested that metastatic deposits from primaries in these sites have a particular tendency to undergo cystic transformation. Despite rigorous investigation however a high proportion of primary sites remain occult (29). Branchial cleft cyst carcinoma is extremely rare, compared to the far more frequent cystic metastases arising from primary malignancies.

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