Rare Clinical Presentation of Branchial Cyst: A Case Report.

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Case Report

Abstract
A case report of a big lateral branchial cyst growing for one year and presented as sixty three year old female is described. The diagnosis was based on ultrasonography with aspiration biopsy, computed tomography and histopathological findings. Successful surgical treatment was performed in Al-Karama teaching hospital. There is no recurrence after a period of follow up for about eighteen months postoperatively. Rarity of big size and strange presentation in the terms of old age and the site of the cyst is emphasized.

 소개한 계통성의 중추부 공기방어의 특이한 진단

Introduction
Branchial cysts are the commonest cysts to arise in the neck. In the early descriptions they were described as to arise anterior to the sternocleidomastoid muscle. However there have been a number of case reports describing cysts which were found in areas other than the classical position. Just as there have been various sites for the cysts, there have been a number of theories proposed as possible etiologies. This is a case report of huge branchial cyst found in the left posterior triangle of the neck of an elderly woman, with a review of the available literature.Lateral cysts of the neck were first described by Hunczovsky in 1785. [1] Since then these swellings have been described by various names and various etiological hypotheses have been proposed for them. These hypotheses can be conveniently classified as the "Congenital theories" and the "Lymph node theories". Rathke described pharyngeal pouches in 1828. Following this Ascherson proposed the "Branchial theory", suggesting the imperfect obliteration of the pharyngeal cleft as the cause of these cysts. But in 1886 he proposed his "Precervical Sinus Theory" saying that these cysts were related to the cervical sinus rather than the pharyngeal clefts or pouches. [1] Wenglowski in 1912 in his dissection of cadavers found that no pharyngeal cleft tissue was found below the level of the hyoid bone and thus pointed out the inability of the "branchial theory" in explaining the cysts in the lower neck. He proposed
the possibility of incomplete obliteration of the thymopharyngeal duct.[1]

Based on the findings of Lucke and Luschka, King proposed the "Lymph node theory". Bhasker and Bernier, after their histological study of cysts concluded that these cysts developed due to cystic transformation of cervical lymph nodes. Maran and Buchanan arrived at the same conclusion however they also pointed out that such a transformation was not known to occur in any other site. [1]

There was a lot of speculation about the possible origin of the epithelium within the lymph node that led to the cystic transformation of the node and eventually formed the lining of the cyst. Bhasker and Bernier suggested 3 possible sources, the branchial cleft, the pharyngeal pouch and the parotid gland. [1-4]

Wilde and Mischke [5] , [6] studied the keratin content of the epithelial lining of the cysts and found it homologous to the keratin content of the upper digestive tract epithelium, particularly that of the palatine tonsils. This was further confirmed by Stoll7 who injected blue dye into the tonsillar fossa of 3 cases preoperatively and found the distribution of stain within the capsule of the cyst during surgery. Hosemann after his study of the histopathological characteristics of the cyst epithelium, found it similar to the tonsillar epithelium. [8]

Although both the branchial cysts and fistulas were regarded to be of branchial origin now it is believed that "branchial cysts" have a non-branchial origin. [9-13]

The median age of presentation of these cysts is in the 3rd decade. They are found to occur more commonly in females. [1], [14] They have classically been described to occur anterior to the upper third of the sternomastoid. [15] However they have been reported to occur in the other areas of the neck, [1], [16] as well as in the oral cavity,17-19 within the salivary glands, [16], [20-23] the thyroid24 and even in the mediastinum 25 and within the pancreas. [26]

Within the neck they occur most commonly in the region anterior to the upper third of the sternomastoid, followed by the region to the middle and lower third of the sternomastoid. [15] It very rarely occurs in the posterior triangle of the neck. Most of the cysts occur deep to the investing layer of the deep cervical fascia and none of them have a cord or tract attached leading to the skin or the pharynx. [1], [11], [12], [14]

On microscopic examination these cysts are composed of a wall of lymphoid tissue lined with squamous or columnar cells. [1], [8], [14] Due to the variability of the position King suggested that any cyst arising outside the midline, with the histological features as above should be regarded as a lymphoepithelial or a branchial cyst. [1]
TABLE 1
Sites of branchial cysts in the neck

<table>
<thead>
<tr>
<th>Site</th>
<th>Titchener and Allison</th>
<th>Colledge and Ellis</th>
<th>Neel and Pemberton</th>
<th>King</th>
<th>Bhasker and Bernier</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ant. to Upper 1/3 of SCM</td>
<td>35</td>
<td>85</td>
<td>73</td>
<td>74</td>
<td>41</td>
</tr>
<tr>
<td>Ant. to Mid and Lower 1/3 of SCM</td>
<td>10</td>
<td>22</td>
<td>7</td>
<td>15</td>
<td></td>
</tr>
<tr>
<td>Submandibular Region</td>
<td>7</td>
<td>5</td>
<td>0</td>
<td>3</td>
<td>10</td>
</tr>
<tr>
<td>Parotid</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>3</td>
<td></td>
</tr>
<tr>
<td>Anterior Neck</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>15</td>
<td></td>
</tr>
<tr>
<td>Posterior Triangle</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>3</td>
<td></td>
</tr>
</tbody>
</table>

Langlois [27] has described pancreatic tissue in a lateral cervical cyst, while Gosain and Wildes[28] found gastric epithelium within a branchial cyst. There have also been reports of secondaries from papillary thyroid cancers [29] and tonsillar cancers masquerading as branchial cysts, [30] but such cases are rare. Most cases present with a painless, soft, cystic and brilliantly transluscent swelling in the neck. Rarely is a cyst painful. [14]

Although clinical diagnosis of a cyst in the classical position is relatively simple. They are seldom correctly diagnosed preoperatively at other sites. Titchener and Allison [14] could make a correct preoperative diagnosis in only 22 out of their 42 cases, and have stressed on the role of preoperative ultrasonography of the neck and FNAB for evaluation of such cases. Earl31 has also found USG to be very helpful in the diagnosis of cystic neck swellings. The role of FNAB and preoperative CT scan of the neck has also been discussed by Rossell. [32]

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Most patients are cured by complete surgical excision 9 of the cyst and usually never get a recurrence [11] and surgery remains the mainstay in the treatment of lateral cervical cysts. Unlike cystic hygromas, there are no reports of any use of sclerosing agents or other drugs like OK432 or bleomycin in this condition.

Case presentation:
A 63 year old lady presented to my private clinic with history of left –sided neck swelling of one year duration where the lesion became visible and palpable by the patient, and evolved over a year to larger size and associated with heaviness sensation and cause respiratory difficulty, especially when she was lying down in supine position.

Clinical examination of the patient:
An elderly lady, crippled due to poliomyelitis.
There was a left –sided neck swelling in the upper 2/3 of neck , occupying the posterior triangle and the lateral aspect of anterior triangle of neck, of about 8x6cm., soft, smooth surface,
neither hot nor tender, fluctuation test was positive, non pulsatile, non movable. There was no cervical lymph node enlargement. Scattered tattooing of the face and neck.

![Figure 1](image1.png) **Figure 1** anterior view of the patient preoperatively

![Figure 2](image2.png) **Figure 2** lateral view of the patient preoperatively showing the posterior triangle BC

**Investigations**

Heamatological were within normal x-ray of neck and chest were non specific US. of neck: showed a huge purely cystic, of thin wall lesion with no internal septation, no solid component, measuring 7.5 X 4.5 cm. seen in the left side of the neck, pushing the left carotid sheath medially, abutting but not arising from the left lobe of thyroid gland as shown in fig 3.
CT scan with contrast revealed well defined thin wall unilocular purely cystic lesion oval shaped measuring 5x7cm in dimensions occupying the left side of the neck displacing left thyroid lobe anteromedially, left carotid sheath medially and trachea to controlateral side the cyst is deeply seated forming indentation of oropharynx and abutting the posterior part of the trachea giving an impression of benign cystic lesion most likely branchial cyst.

FNAC: necrotic cells with abundant lymphoid and inflammatory cells. Doppler US.: indicate non vascular lesion.
On operation fig.4

a- intraoperative view of the cyst

b-cyst after removal

Total excision of a unilocular cyst with clear fluid deep to the investing fascia without stalk by transverse skin crease incision. Suction drain left inside the wound followed by closure in layers and dressing. Where the specimen submitted for histopathological examination. The patient run uneventful postoperative period. Drain tube removed in the first post operative day.

Histopathological examination of the specimen revealed abundant lymphoid tissues with germinal centers with focal fibrosis, concluding that the branchial cyst is the most probable diagnosis fig.5.
Fig.(5-a) Histopathological examination of the cyst.

Fig.5b HPE of the cyst.
Discussion

Branchial Cyst
The novel criteria in this case are:
1. the age of presentation
2. the site and the magnitude of the cyst

As per King’s criteria any cyst arising outside the midline of the neck and having lymphoepithelial characteristics should be regarded as a branchial cyst. Such cysts are found more commonly in females and usually occur in the 2nd or 3rd decade of life. Presentation of the cyst in the elderly female aged 63 year old with gradual increment in size over a year before hospital admission is extremely rare. They are most commonly found in the anterior triangle of the neck anterior to the upper third of the sternomastoid. A cyst occupying the posterior triangle is extremely rare. However these cysts have been reported to occur in all the regions of the neck, and even in the mediastinum and the abdomen. Hence they should be suspected in all the cystic swellings of the neck except the median ones. Ultrasonography, CTscan and FNAB definitely help in arriving at the diagnosis and is especially recommended for patients in the older age group to rule out cystic secondaries from head and neck malignancies. On operation a unilocular cyst with clear fluid, deep to the investing fascia and without a connecting stalk more or less makes the diagnosis certain. The histological picture is classical and confirmatory. The ‘Branchial theory’ has now fallen into disfavour and the most appropriate hypothesis explaining the aetiology of these cysts is the "Lymph node inclusion theory" with the palatine tonsils as the most likely source of the enclosed epithelium. Complete surgical removal remains the only acceptable form of treatment

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