IJAE
Vol. 123, n. 3: 194-198, 2018
ITALIAN JOURNAL OF ANATOMY AND EMBRYOLOGY

Research Article - Basic and Applied Anatomy

Branchial cleft cysts: serie of 33 cases and review of the literature

Alicia González-Mourelle[1,*], Cristina Vicente-Fernández[2], María Pombo-Castro[1], Inés Vázquez-Mahía[1], José Luis López-Cedrún[1]

[1] Department of Maxillofacial Surgery of A Coruña University Hospital, A Coruña, Spain
[2] Department of Otorhinolaryngology of Pontevedra Hospital, Spain

Abstract

Branchial cleft cysts develop because an anomaly in the caudal growth of the second arch over the third and the fourth ones. They usually present as an asymptomatic circumscribed movable mass, close to the anterior border of the sternocleidomastoid muscle. The location depends on the branchial pouch or cleft they are derived from. We present a retrospective study including 30 cases analysing epidemiology, clinical presentation, diagnosis, treatment and complications. We obtained the following results: 15 of the 30 patients were females and 15 males. Age ranged from 19 to 81 years with an average of 40. All cysts had origin from the second branchial cleft. Twenty-three appeared as painless cervical masses, 5 were painful and 2 had an infection. Clinical suspicion of branchial cleft cyst formed in 23 cases. Computerized tomography and fine needle aspiration cytology was used in 18 cases, magnetic resonance alone in 1 and only ultrasound in 1. Branchial cleft cyst is a differential diagnosis of a lateral neck swelling mass and the most accurate diagnostic test is magnetic resonance, but computerized tomography is the most often performed in most hospitals. Treatment is surgical excision.

Key words

Branchial arch, branchial groove, branchial pouch, pharyngeal pouch, branchial fistula.

Introduction

Pharyngeal arches and clefts are the most important elements in the development of the head and neck. Branchial arch anomalies are associated with several craniofacial syndromes including branchio-oto-renal, Goldenhar, Treacher-Collins, Nager, Miller, Wildervanck and Bixler syndrome (Cunningham, 1992; Ozolek, 2013). Branchial cleft cysts develop when there is an anomaly in the caudal growth of the second arch over the third and the fourth ones. There are other theories: vestiges of cervical sinus, incomplete obliteration of thymopharyngeal duct, cystic degeneration of cervical lymph nodes, trapped epithelium from the parotid gland, pharyngeal pouch or branchial cleft (Inclusion Theory) (Daoud, 2005; Thomaidis et al., 2006). Reviewing the literature we don't find any difference between sexes. Branchial cysts constitute 20-80% of branchial anomalies (Daoud, 2005). They comprise 17% of pediatric cervical masses (Daoud, 2005) and 4% of the cases of neck lump (Cunningham, 1992). They usually present as an asymptomatic circumscribed movable mass, close to the

* Corresponding author. E-mail: campodafonte@hotmail.com

http://www.fupress.com/ijae
DOI: 10.13128/IJAE-25408

anterior border of the sternocleidomastoid muscle (Daoud, 2005). They use to gradually enlarge and do not become evident until the second or third decade of life. Their presentation in childhood is related to enlargement due to an upper respiratory tract infection. They have lymphoid tissue located beneath the epithelium that makes them as reactive as lymph nodes. Infected cysts may develop into abscesses (Cunningham, 1992; Thomaidis et al., 2006). The precise location and course of branchial cleft cysts depend on the particular branchial pouch or cleft they are derived from (Lore, 2005). First branchial cleft cyts are located inferiorly to the external auditory canal and above the level of the hyoid bone. The sinus tract can drain in the postauricular or preauricular regions or near the mandibular angle, just anteriorly to the sternocleidomastoid muscle (Gómez-Torres et al., 2013). Daoud (2005) reported an incidence 1-8%. Second branchial cleft cysts are identified along the anterior border of the upper third of the sternocleidomastoid muscle (Thomaidis et al., 2006). There may be a fistula inferiorly, above the anterior border of the sternocleidomastoid muscle, as well as a communication with the tonsillar fossa; Daoud (2005) reported an incidence of up to 95%. Third branchial cleft cysts are located near the largyngeal ventricle. A fistula enters the posterior portion of the thyrohyoid membrane and may communicate with the trachea or the larynx. They present as a cyst located anterior and under the sternocleidomastoid muscle or as a fistula at its posterior border. They represent less than the 1% of the branchial cleft anomalies (Batuecas et al., 2006). Fourth branchial cleft anomalies are have not yet been demonstrated.

Diagnosis is based on clinical and radiological criteria (Cunningham, 1992) and histological confirmation with fine needle aspiration cytology. Magnetic resonance gives better information on the benign or malignant character and the vascular appearance than computerized tomography (Howie and Proops, 1982; Batuecas et al., 2006). Fistulograms can help when an external sinus opening is present (Cunningham, 1992). The presence of cholesterol crystals and/or epithelial cells in the aspirate will suggest the diagnosis. Treatment is surgical excision (Batuecas et al., 2006) through a wide transverse cervicotomy under general anaesthesia. It's very important to avoid any type of drainage operation and resection in the presence of active infection. The entire duct or tract must be removed if it is patent (Lore, 2005; Múñoz-Fernández et al., 2011; Gómez-Torres et al., 2013). The internal opening of the sinus is crucial for defining its embryologic origin, removing these developmental defects and avoiding recurrence (Ozolek, 2009). Injections of sterile methylene blue, liquid paraffin or quick-hardening polimers, and the insertion of esophageal bougies and other blunt probes have been used to enhance the success of the surgical procedure. Potential benefits have been reported for a newer approach using trans-catheter balloon embolectomy over previously used methods (Cunningham, 1992). The most frequent complications reported in the literature are recurrence and nerve injuries. Recurrences are twice more frequent in the case of sinus or fistulas resection compared to cysts (Cunningham, 1992). Histopathology findings show a cyst and/or sinus tract lined by respiratory or squamous epithelium. Lymphoid tissue, sebaceous elements and salivary glands may be present in the wall of the cysts, making distinction from dermoid cysts challenging (Batuecas, 2006; Ozolek, 2009).

Materials and methods

We performed a retrospective study including 30 cases of branchial cleft cysts from 30 patients collected at the Departments of Otorhinolaryngology and Maxillofacial Surgery of A Coruña University Hospital from 2006 to 2016: they were 15 females and 15 males. Age ranged from 19 to 81 years with an average of 40 and a median of 30. The aim of the study was to analyse the epidemiology of this congenital anomaly, clinical presentation, diagnosis, treatment and complications.

Results

All the 30 cases were second branchial cleft cysts. Fourteen were located in the II cervical level, ten in the IB level, one in the III level, two in the II and III levels, one in the II, III and IV levels. Two had not been localized in the clinical history. There were no statistically significant differences between both sides of the neck. Twenty-three of the 30 cases appeared as painless cervical masses, 5 were painful and 2 had an infection process. Clinical suspicion was branchial cleft cyst in 23 of the 30 patients, inflammatory adenopathy in 3, metastatic adenopathy in 2, lipoma in 1 and submandibular gland inflammation in 1. Two of them had no clinical orientation. For the diagnosis, computerized tomography and fine needle aspiration was used in 18 cases. In 11 of them a concomitant ultrasound scan was performed. Computerized tomography alone was used in 5. In 3 of them an ultrasound scan was also acquired. Exclusively magnetic resonance was used in 1 case, ultrasound and fine needle aspiration in 4 cases, exclusively ultrasound in 1 case. A computerized tomography and a biopsy were performed in the case that was mistaken with a lipoma. Surgical excision was performed in all the patients except two who were lost before making the diagnosis sure. No complications or recurrences occurred.

Discussion

There are few recent series discussing branchial cysts separately from other branchial anomalies. It is one of the commonest cystic lesions in the neck. Most of them have their origin in the second branchial cleft or pouch. In this series of 30 cases all had this origin. Twenty-four of the 30 were localized in the classical site (upper neck, anterior to the sternocleidomastoid muscle). One of them was in the middle neck, anterior to the sternocleidomastoid muscle, and 3, because of their size, were in the upper and middle neck. As in previously reported series there was no difference here between the sides of the neck. In other series the cysts were more frequent in females than in males, that did not apply to the series we are presenting. The mean age of presentation in our series was higher than in previously published ones (Table 1). We demonstrated a high rate of diagnostic accuracy as in other series (Maran and Buchanan, 1978; Chandler and Mitchel, 1981; Kenealy et al., 1990). However Titchener and Allison (1989) reported a failure in clinical orientation in 52% and Ingoldby (1985) in 47.8% of a series of 23 cysts. In our study the branchial cleft cysts were mistaken with reactive adenopathies (2/30), metastatic adenopathies (3/30), lipoma (1/30) and

Table 1. Age, gender, side of presentation of branchial cysts compared with other series.

Series	N	Mean age (range)	Male - Female	Right side - Left side
Howie and Proops (1982)	50	30 (14-74)	2-3	26-25
Titchener and Allison (1989)	42	(2-62)	1-2	21-21
Golledge and Ellis (1994)	20	31 (14-50)	9-11	12-8
Agaton-Bonilla and Gay-Escoda (1996)	148	23.06	42-106	47-101
Daoud (2005)	34	19 (1-57)	12-22	17-17
González-Mourelle and Vicente-Fernández (2016)[this study]	30	40 (19-81)	15-15	10-17

submandibular gland inflammation (1/30). Computerized tomography combined with fine needle aspiration were the studies used in most of our patients (18). In the rest we performed computerized tomography (5), ultrasound (1), ultrasound (1) and ultrasound with fine needle aspiration (4). In previous studies fine needle aspiration and ultrasound were of minor importance: Agaton-Bonilla and Gay-Escoda (1996) used computerized tomography in 44% and ultrasound in 13% of their cases. Choi and Zalzal (1995) also used computerized tomography and ultrasound in 27% of the patients. As a conclusion, when we receive in our consultation a patient with any lateral neck swelling mass we must take into account the diagnosis of branchial cyst, regardless of whether the swelling is solid or cystic, painful or painless. The most accurate diagnostic test is ultrasound, but computerized tomography is the most often performed in most of the hospital environments because it is more easily achievable. It is also advisable to perform fine needle aspiration to accurately demonstrate the cystic nature. Treatment is surgical excision and uses to be curative. Recurrences and complications have a low frequency. The risk is higher if the cyst has been drained before.

Acknowledgement

The completion of this undertaking could not have been possible without the participation and assistance of so many people whose names may not all be enumerated. Their contributions are sincerely appreciated and gratefully acknowledged. The group would like to express deep appreciation and indebtedness particularly to M. Pombo, I. Váquez Mahía, F. Lorenzo, J.L. López-Cedrún, T. Pazos, R. Ramiro. To all relatives, friends and others who in one way or another shared their support, either morally or physically, thank you. There was no financial support nor is there any conflict of interest.

References

Agaton-Bonilla F.C., Gay-Escoda C. (1996) Diagnosis and treatment of branchial cleft cysts and fistulae. A retrospective study of 183 patients. Int. J. Oral Maxillofac. Surg. 25: 449-452.

Batuecas C.A., Blanco P.P., Santa C.R.S., Serradilla L.J.M., González S.M., Moreno J.D. (2006) Persistencia de tercer arco branquial. Acta Otorrinolaringol. Esp. 57: 193-195.

Chandler J.R., Mitchell B. (1981) Branchial cleft cysts, sinuses, and fistulas. Otolaryngol. Clin. North Am. 14: 175-186.

Choi S.S., Zalzal G.H. (1995). Branchial anomalies: a review of 52 cases. Laryngoscope 105: 909-913.

Cunningham M.J. (1992) The management of congenital neck masses. Am. J. Otolaryngol. 13: 78-92.

Daoud F.S. (2005) Branchial cyst: An often forgotten diagnosis. Asian J. Surg. 28: 174-178.

Golledge J., Ellis H. (1994) The aetiology of lateral cervical (branchial) cysts: past and present theories. J. Laryngol. Otol. 108: 653-659.

Gómez-Torres A., Tirado Z. I., Esteban O. F. (2013) Branchial cyst causing laryngeal dyspnoea in a newborn. Acta Otorrinolaringol. Esp. 64: 243.

Howie A.J., Proops D.W. (1982) The definition of branchial cysts, sinuses and fistulae. Clin. Otolaryngol. Allied Sci. 7: 51-57.

Ingoldby C.J.H. (1985) Unusual presentations of branchial cysts: a trap for the unwary. Ann. R. Coll. Surg. Engl. 67: 175-176.

Kenealy J.F., Torsiglieri A.J. Jr., Tom L.W. (1990) Branchial cleft anomalies: a five-year retrospective review. Trans. Pa. Acad. Ophthalmol. Otolaryngol. 42: 1022-1025.

Lore J.M. (2005) An Atlas of Head & Neck Surgery. Elsevier, Philadelphia.

Maran A.G., Buchanan D.R. (1978) Branchial cysts, sinuses and fistulae. Clin. Otolaryngol. Allied Sci. 3: 77-92.

Muñoz-Fernández N., Mallea-Cañizares I., Fernández-Julián E., De La Fuente-Arjona L., Marco-Algarra J. (2011) Doble anomalía de la segunda hendidura branquial. Acta Otorrinolaringol. Esp. 62: 68-70.

Ozolek J.A. (2013) Selective pathologies of the head and neck in children. A developmental perspective. Adv. Anat. Pathol. 16: 332-358.

Thomaidis V., Seretis K., Tamiolakis D., Papadopoulos N., Tsamis I. (2006) Branchial cysts. A report of 4 cases. Acta Dermatovenerol. Alp. Pannonica Adriat. 15: 85-89.

Titchener G.W., Allison R.S. (1989) Lateral cervical cysts: a review of 42 cases. N.Z. Med. J. 102 :536-537.