Acta Otorrinolaringol Esp. 2011;**62(1):**68–70

Acta Otorrinolaringológica Española

www.elsevier.es/otorrino

CLINICAL CASE

Double second branchial cleft anomaly

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Received April 22, 2009; accepted January 18, 2010

KEYWORDS
Branchial anomalies;
Cyst;
Second branchial arch

Abstract Second branchial cleft anomalies are the most common of this type of neck masses. They can be classified in four types (Bailey/Proctor classification) according to their location. Type II is the most common, and related to vital neck structures such as the carotid artery and jugular vein. Cysts are the most frequent among them. Management consists of surgical excision of the cyst and tract by cervicotomy to avoid recurrence. We present an extremely rare case of a 32-year-old male who presented a sudden appearance of a right lateral neck mass that was identified by an image study as a double branchial cleft cyst. A review of simultaneous branchial cleft cyst in the literature is also made.

PALABRAS CLAVE
Anomalías branquiales;
Quiste;
Segundo arco branquial

Doble anomalía de la segunda hendidura branquial

Resumen Las anomalías de la segunda hendidura branquial son las más comunes. Hay 4 tipos (clasificación de Bailey/Proctor) según su localización. El tipo II, que es el más común, está en contacto con los grandes vasos. Su forma de presentación es diversa, aunque la más frecuente es la quística. El tratamiento es quirúrgico con extirpación completa por cervicotomía para evitar recidivas. Presentamos el caso clínico extremadamente raro de un varón de 32 años con debut brusco de tumoración laterocervical derecha, con estudio de imagen que demuestra la presencia de doble quiste branquial. Revisamos en la literatura casos de simultaneidad de quistes branquiales.

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0001-6519/$ - see front matter

Clinical case

We report the case of a 32-year-old man who was referred to our clinic from the emergency department due to a large right cervical mass of various hours' evolution. He presented no fever, malaise or discomfort on swallowing; the only relevant history was having performed strenuous exercise some hours before. He reported no recent infections or toxic habits. The remaining pathological examinations and history contained no data of interest.

On examination, we noted a well-defined elastic laterocervical mass of 7×4 cm in areas II-III, adhered to deep planes, without fluctuations or acute inflammatory signs. We proceeded to complete the study.

The ultrasound scan showed a solid ovoid lesion with a homogeneous echostructure, slightly echogenic. The lesion had smooth, well-defined boundaries of 65mm in close contact with large vessels, without evidence of flow.

The CT scan revealed a well-marked bulky cystic lesion with no identified walls, medial to the sternocleidomastoid and posterior to the carotid space, 4.4x2.4x7.9 cm in size (AP, transverse and cephalocaudal). There was another 1.9x1.7x2.5 cm image with similar characteristics anterior to the carotid space and posterior to the submandibular gland (Figure 1). The internal jugular vein appeared compressed between the two lesions described, and the submandibular gland appeared displaced in the anterior plane (Figure 2). Cytology was consistent with a branchial cyst.

Faced with the possibility of two branchial cysts or one bilobed cyst, a cervicotomy was performed, which corroborated the first option.

The definitive histological examination revealed a double branchial cyst.

Figure 1 Computed tomography: axial section showing the 2 lesions described. It is possible to observe the displacement of surrounding structures.

Figure 2 Sagittal image from computed tomography. The compression of the internal jugular vein by cystic lesions is clearly identified.

Discussion

The term "branchial apparatus" refers to elements giving rise to the anterior and lateral neck regions, which are formed in the 3rd week of development. The 5 "branchial" arches originate from condensations of the mesoblast that are ventrally fused to the pharynx. Due to evolutionary reasons, the 5th arch (which is hidden) is often referred to as the "6th arch".[1] The 2nd, which develops caudally, covers the 3rd, 4th and 6th (or 5th) arches, creating a cavity. A kind of cyst covered by ectoblast that would be formed disappears subsequently; this is the cervical sinus.[2] Malformation occurs when an incomplete resorption takes place. A lateral cyst appears in front of and within the sternocleidomastoid. The cyst is round, renitent, mobile and implanted below the hyoid bone.

The way these anomalies form is such a complex issue that it is necessary to study their embryonic development to understand them. Consequently, the development of two anomalies depending on the same branchial arch is even more difficult to understand.

The development of the 2nd arch takes place on a more extended time period; the anomalies in this arch are therefore more common.[3] Bailey (1933) and Proctor (1955) described a classification for 2nd-arch cysts based on their location and anatomical relationships. Type I is superficial to the anterior edge of the sternocleidomastoid beneath the cervical fascia and platysma. Type II is the most frequent and appears adjacently to large vessels. Type III passes between the carotids, extending into the pharynx, and Type IV grows on the pharyngeal wall.[4]

At present, complete excision is recommended. Nobody contests the use of imaging and cytology tests for a diagnostic approach. The definitive diagnosis is always given by the anatomopathological examination of the piece.

In the published literature, we verified the scarce presence of multiple malformations. Approximately 2%

3% of abnormalities are bilateral, but we do not know the percentage of simultaneity. Gupta reported a case of simultaneous occurrence of an abnormality of the lst and 2nd branchial clefts.[5] Mehrzad et al published the coexistence of malformation and of the 3rd and 4th arches.[6] The series of Hu[7] reported two cases of multiple branchial cysts.

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