Case Report

Giant cyst of the second branchial arch in an adult: case report and literature review

Cisto gigante de segundo arco branquial em adulto: Relato de caso e revisão de literatura

LUIS GUSTAVO RAMOS RAUPP PEREIRA[1]
MARIA EDUARDA TOMASETTO[1]
RENAN MATHIAS FERREIRA SALTIÉL[1]
IGOR FAGUNDES SARTORI[1]
BRUNO LUIZ AMARAL[1]
EDUARDO MADALOSSO ZANIN[1]*

■ **ABSTRACT**

Introduction: The branchial arches are the embryological precursors of the face, neck, and pharynx. Branchial arch anomalies are the second most common congenital head and neck lesions in children. Among these anomalies are branchial arch cysts (BCC), which arise due to incorrect obliteration of the branchial slits, still in the embryonic period. BCCs may be asymptomatic, only noticed incidentally, and not manifest until adulthood. **Results:** Anomalies of the second branchial arch should be considered as one of the possible differential diagnoses of neck masses, especially those that manifest as a bulge in the lateral region of the neck. BCCs are epithelial lining formations without external openings. After diagnosis, treatment is surgical, usually through a transverse cervical incision and careful dissection of the structures, with the aim of extirpating the entire lesion. **Conclusion:** The method described of excision of the lesion through a transverse incision in the cervical region, tissue dissection in planes, and resection of the cystic mass is an option for the treatment of this deformity, with adequate aesthetic results and good reproducibility.

Keywords: Branchial region; Plastic surgery procedures; Epidermal cysts; Genetics; Diagnosis, differential.

■ **RESUMO**

Introdução: Os arcos branquiais são os precursores embriológicos da face, pescoço e faringe. As anomalias dos arcos branquiais são a segunda lesão congênita mais comum de cabeça e pescoço em crianças. Entre essas anomalias, estão os cistos de arcos branquiais (BCC), que surgem devido a uma incorreta obliteração das fendas branquiais, ainda no período embrionário. Os BCC podem ser assintomáticos, apenas percebidos incidentalmente, e não se manifestar até a idade adulta. **Resultados:** Anomalias do segundo arco branquial devem ser consideradas como um dos possíveis diagnósticos diferenciais de massas cervicais, especialmente as que se manifestam como um abaulamento em região lateral do pescoço. Os BCC são formações de revestimento epitelial, sem aberturas externas. Após seu diagnóstico, o tratamento é cirúrgico, usualmente por meio de uma incisão cervical transversa e cuidadosa dissecação das estruturas, com o objetivo de extirpar toda a lesão. **Conclusão:** O método descrito, de excisão da lesão, por meio de incisão transversa em região cervical, dissecção tecidual por planos e ressecção de massa cística, é uma opção para o tratamento dessa deformidade, com adequado resultado estético e boa reprodutibilidade.

Descritores: Região branquial; Procedimentos de cirurgia plástica; Cisto epidérmico; Genética; Diagnóstico diferencial.

Institution: Escola de Medicina, Universidade de Passo Fundo, Passo Fundo, RS, Brazil.

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[1] Universidade de Passo Fundo, Passo Fundo, RS, Brazil.

INTRODUCTION

The branchial arches are the embryological precursors of the face, neck, and pharynx, and the anomalies that affect them are the second most common congenital lesion of the head and neck in children, being divided into 4 groups: those of the first, second, third and fourth branchial arch, depending on your location[1]. Anomalies of the second branchial arch are the most prevalent and correspond to 95% of the changes found in the branchial apparatus.

Deformities of the branchial arches can present as cysts, sinus tracts, fistulas, or cartilaginous remnants[2]. In the case of branchial arch cysts (BCC), they usually occur in older children and young adults, while fistulas are present in infants or younger children. In the case described, the patient presented with the cyst at the age of 24.

BCCs can be asymptomatic, only noticed incidentally, and not present until adulthood, or manifest through non-specific symptoms, including edema in the neck region or recurrent infections1. The diagnosis is normally made with clinical examination and imaging. Treatment is surgical, with complete excision of the lesion, usually through a transverse cervical incision and careful dissection of the structures, with the aim of removing the entire lesion[3].

OBJECTIVE

In this report, we present the case of a 24-year-old female patient diagnosed with a second branchial arch cyst. The objective of this report is to correlate the case described with the knowledge available in the literature. Because second arch BCC is one of the differential diagnoses of masses in the cervical region and is very frequently underdiagnosed, the surgeon must know how to identify it in order to avoid incorrect diagnoses and promote adequate patient management.

CASE REPORT

Female patient, 24 years old, primiparous, sought outpatient care at the University of Passo Fundo-RS in December 2022 due to a nodule in the anterior cervical region, 7cm in diameter, on the right (Figures 1A, 1C). She reports the evolution of the cervical mass with slow and progressive growth and denies a previous medical history of chronic diseases, smoking, and continuous use of medications.

On physical examination, a mobile mass was observed, painless on palpation, without adherence to deep planes, without indications of invasion into adjacent tissues, and complications due to phlogistic signs. The ultrasound examination of the cervical region carried out prior to the consultation revealed the presence of an echogenic nodulation in the right cervical region, measuring 6.69cm.

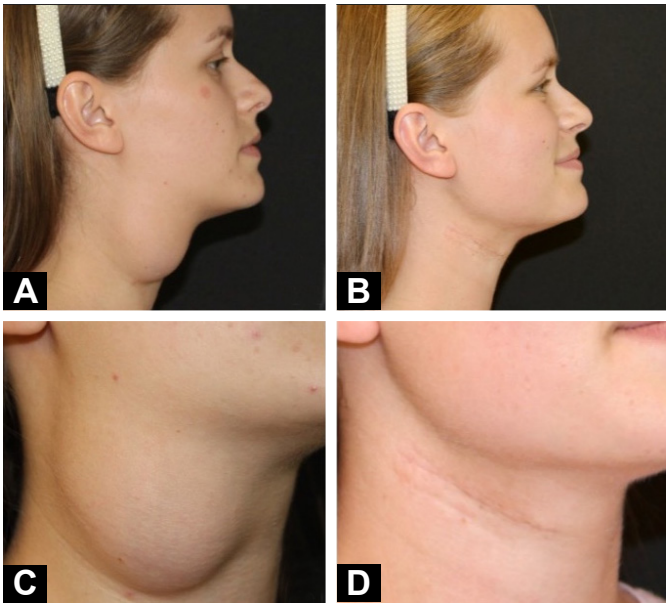

Figure 1. A, **B**, **C** e **D:** Pre and post-operative images.

A surgical procedure was performed to excise the lesion through a transverse incision in the cervical region, measuring approximately 5 cm, according to the orientation of Langer's lines. We continued with tissue dissection in layers (Figure 2) and resection of a cystic cervical mass with a cysto-serous appearance. The mandibular branch of the facial nerve was identified and preserved during the intervention and finished with surgical synthesis, in plans, and placement of a number 2 Penrose drain at the surgical site, with an exit external to the surgical wound.

The patient evolved well clinically and with progressive recovery. The drain was removed five days postoperatively. After the procedure, the patient developed paraparesis in the right corner of her mouth. The depressor anguli oris muscle on the right with reduced strength on the contralateral side. Symmetrization was carried out with the application of 2 units at 1 point of botulinum toxin in the contralateral muscle and complemented with motor physiotherapy.

The patient was monitored on the 5^{th}, 10^{th}, 15^{th}, 30^{th}, 45^{th}, 90^{th} days and 6 months postoperatively. She evolved with progressive improvement in paraparesis and local edema (Figures 1B, 1D).

The result of the anatomopathological examination was compatible with descriptions of a

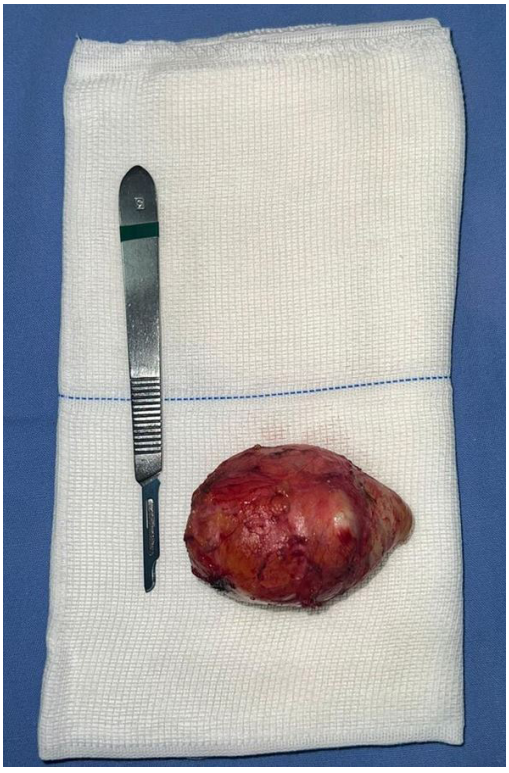

Figure 2. Intraoperative image of cyst excision from the second branchial arch via cervical incision.

cyst of the second branchial arch - cystic structure, measuring 6.0 x 5.5 x 4.5cm, which on section presents yellowish and serous content.

DISCUSSION

Branchial arch anomalies are usually reported in childhood or adolescence but can be first diagnosed at any age. Sinus tracts or fistulas tend to be diagnosed earlier due to skin contact and possible drainage or infection. The average age of fistula and sinus tract diagnosis is 2.6 and 3.6 years, respectively. The average age of diagnosis for branchial cysts is 4.1 years[4,5]. The patient in the aforementioned case was 24 years old at the time of diagnosis and total surgical removal of the lesion.

It is estimated that up to 95% of cases of malformations of the branchial apparatus are derived from the second branchial arch, which can occur in any area between the anterior third of the sternocleidomastoid muscle and the tonsillar fossa. Diagnosis is through clinical analysis and can be aided by radiological evaluation to exclude possible differential diagnoses[3,6]. In the case highlighted, the patient presented significant bulging in the right lateral portion of the neck - a condition compatible with malformations of the branchial apparatus - and an ultrasound examination that showed nodulation in the cervical region without signs of local complications or compromise of vascular and nervous structures.

In general, they do not present an important predominance of sex or side of occurrence[7]. The pathophysiological mechanism of BCC formation generally results from incomplete obliteration of the branchial slits, with subsequent formation of cysts and fistulas[2]. Cysts have an epithelial lining without external openings, while branchial cleft fistulas are true communications that connect the pharynx or larynx to the external skin and can drain mucous secretions[7,8]. In the report above, the present clinical picture corresponded to the cystic presentation of BCC without obvious complications and drainage of mucous contents.

Ultrasound examination usually shows a well-circumscribed cyst. However, there is variability in the ultrasound appearance of second arch BCC when there is a secondary infection or when septa or cellular debris are present within the cyst, resulting in a pseudosolid or heterogeneous appearance[9]. The cervical ultrasound performed in the case revealed a well-defined echogenic nodulation in the cervical region on the right, with no evidence of the presence of infection associated with the cyst.

Computed tomography helps in the diagnosis and topographic study of the lesion and its relationships with important vascular and nervous cervical structures, but it is not essentia[13]. In the reported context, a tomographic examination was not performed, as the clinical and ultrasonographic presentation were not compatible with warning signs of malignancy and involvement of important structures, evidenced by the lack of adherence to deep planes and infiltration of tissues adjacent to the lesion.

Anomalies of the second branchial arch should be considered as one of the possible differential diagnoses of neck masses, especially those that manifest as bulging in the lateral region of the neck. Furthermore, the high rate of mistaken clinical diagnoses is highlighted, especially in relation to branchial cysts and fistulas, which makes it clear that these changes are constantly neglected in relation to differential diagnoses[4,9].

Furthermore, it is noteworthy that the treatment is surgical, with complete excision of the lesion, usually through a transverse cervical incision and careful dissection of the structures, with the aim of extirpating the entire lesion[3]. Therefore, it was the therapeutic approach of choice in the case report, which was followed by an anatomopathological analysis of the excised cyst, which confirmed compatibility with a cyst of the branchial apparatus. Furthermore, an

endoscope-assisted transcervical approach can also be performed, which has a smaller incision size[10]. Studies on less invasive procedures for various anomalies are promising, including sclerotherapy and endoscopic excision of the second arch BCC[11].

Finally, second arch BCC is one of the differential diagnoses of masses in the cervical region and is very often underdiagnosed, in addition to the fact that, in the adult population, branchial cysts are a challenge due to the possibility of cystic metastasis from occult carcinoma[1,11]. Therefore, the surgeon must know how to identify it in order to avoid incorrect diagnoses and to promote adequate patient management.

COLLABORATIONS

LGRRP Analysis and/or data interpretation, Conception and design study, Conceptualization, Data Curation, Final manuscript approval, Formal Analysis, Funding Acquisition, Investigation, Methodology, Project Administration, Realization of operations and/or trials, Resources, Software, Supervision, Validation, Visualization, Writing - Original Draft Preparation, Writing - Review & Editing.

MET Analysis and/or data interpretation, Conception and design study, Conceptualization, Data Curation, Final manuscript approval, Formal Analysis, Funding Acquisition, Investigation, Methodology, Project Administration, Realization of operations and/or trials, Resources, Software, Supervision, Validation, Visualization, Writing - Original Draft Preparation, Writing - Review & Editing.

RMFS Analysis and/or data interpretation, Conception and design study, Conceptualization, Data Curation, Final manuscript approval, Formal Analysis, Funding Acquisition, Investigation, Methodology, Project Administration, Realization of operations and/or trials, Resources, Software, Supervision, Validation, Visualization, Writing - Original Draft Preparation, Writing - Review & Editing.

IFS Analysis and/or data interpretation, Conception and design study, Conceptualization, Data Curation, Final manuscript approval, Formal Analysis, Funding Acquisition, Investigation, Methodology, Project Administration, Realization of operations and/or trials, Resources, Software, Supervision, Validation, Visualization, Writing - Original Draft Preparation, Writing - Review & Editing.

BLA Analysis and/or data interpretation, Conception and design study, Conceptualization, Data Curation, Final manuscript approval, Formal Analysis, Funding Acquisition, Investigation, Methodology, Project Administration, Realization of operations and/or trials, Resources, Software, Supervision, Validation, Visualization, Writing - Original Draft Preparation, Writing - Review & Editing.

EMZ Analysis and/or data interpretation, Conception and design study, Conceptualization, Data Curation, Final manuscript approval, Formal Analysis, Funding Acquisition, Investigation, Methodology, Project Administration, Realization of operations and/or trials, Resources, Software, Supervision, Validation, Visualization, Writing - Original Draft Preparation, Writing - Review & Editing.

CONCLUSION

As noted, the branchial cyst is the most prevalent subtype of malformations of the second branchial arch, being an important differential diagnosis of masses in the cervical region. The treatment of choice is surgical excision, which is an option for the extirpation of this deformity as it provides an adequate aesthetic result with high-resolution rates, presenting low rates of both recurrences and complications.

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***Corresponding author:** **Eduardo Madalosso Zanin**
Rua Uruguai, 1932, 3º andar, Passo Fundo, RS, Brazil
Zip Code: 99010-112
E-mail: eduardo.zanin@gmail.com