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Case report

Persistent brachial cleft as an infrequent cause of infraglottic stridor and airway obstruction in a 24 year old woman. A case report

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ABSTRACT

Background: We present the case of a 24-year-old woman with respiratory distress associated with a cyst of the fourth branchial cleft that displaced and compressed the upper airway, so the cervical mass was surgically resected, the patient recovered completely.

Case presentation: We present the case of a 24 year old female no other pertinent medical history who presents to the emergency department of way outpatient to respiratory distress associated with a cervical mass. A computed tomography (CT) scan shows a right cervical cystic mass that was displacing and compressing the upper airway. A total resection of the cystic mass was performed, after which the patient recovered completely. The histopathological analysis indicated a branchial cleft cyst which, due to its location, was thought to be the fourth branchial cleft, a rare congenital anomaly.

Conclusions: Fourth branchial cleft cysts are rare malformations. They should be taken into consideration in the differential diagnosis of cervical masses in young adults, especially in situations of potentially life threatening airway compromise where an emergent procedure should be performed to guarantee the patient's life, the diagnosis is based on an adequate history and physical examination, with the support of imaging studies, with CT scan imaging being preferred as it provides information for surgical planning. Treatment is based on complete resection of the cystic mass, which relieves the symptoms of mass effect and decreases the risk of recurrence.

1. Introduction

The branchial apparatus is composed of six arches separated by clefts and pharyngeal pouches. Due to its complex development, abnormalities such as sinuses, fistulas, or cysts, and alterations of the thymus or parathyroid gland may occur [1]. A branchial cleft cyst presents as a cervical mass and its location depends on the responsible cleft. The diagnosis is clinical-radiological and requires surgical treatment [1,2].

We present the case of a 24-year-old female with no other pertinent medical history who presented to the emergency department of way outpatient due to respiratory distress associated with a cervical mass. On CT scan, a right cervical cystic mass was observed that displaced and compressed the upper airway. A total resection of the cystic mass was performed, after which the patient recovered completely. The histopathological analysis indicated a branchial cleft cyst which, due to its location, was thought to be the fourth branchial cleft, a rare congenital anomaly.

2. Case report

The patient is a 24-year-old female no other pertinent medical history, who presented a cervical mass that increased in size 5 days before her evaluation. The mass caused shortness of breath and pain in the cervical region.

On physical examination, a right lateral cervical mass of 8 cm in diameter was observed, at cervical Level III-IV. The mass was soft in consistency and mobile with no cervical adenopathy palpable. There was audible stridor (Figs. 1-2).

Ultrasound of the neck showed decreased thyroid size without the presence of nodules or cysts, with evidence of an anechoic space-

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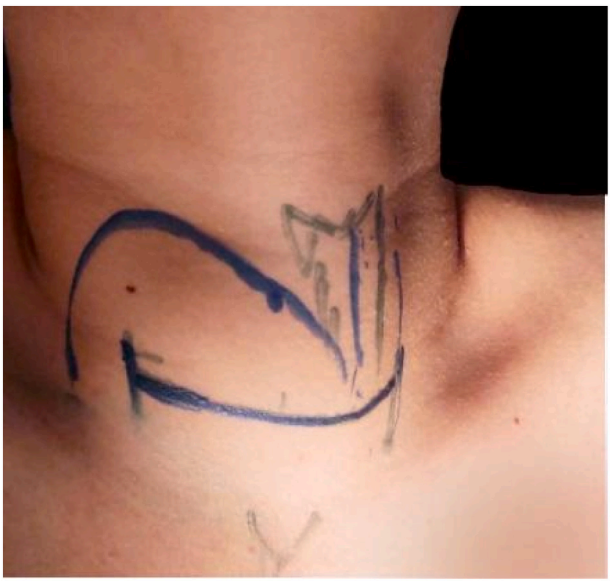

Fig. 1. Right lateral cervical mass.

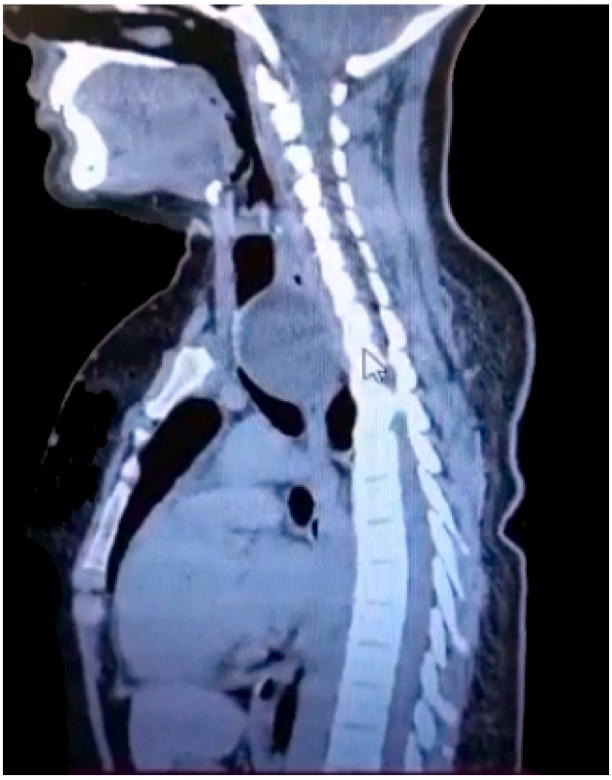

Fig. 2. CT scan of the neck - sagittal view: Homogeneous unilocular mass with a well-defined wall of cystic characteristics located in a cervical region with extension to mediastinum which compresses the trachea.

occupying lesion that displaces the right lobe of the thyroid and trachea.

A CT scan showed a homogeneous unilocular large mass with a well-defined wall of cystic characteristics located in the right lateral cervical region compressing the trachea without invading the airway (Figs. 3-4).

No inflammatory response was evident in laboratory studies. However, the thyroid profile indicated hypothyroidism, and arterial blood gas reflected respiratory acidosis. Due to her respiratory acidosis, an emergent surgical intervention was indicated. The patient was intubated using video laryngoscopy that visualized extrinsic compression of the

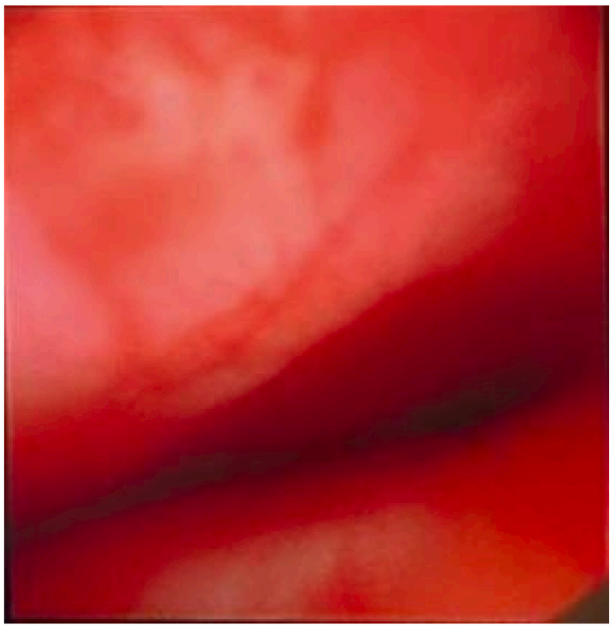

Fig. 3. Video laryngoscopy: Extrinsic compression of the trachea.

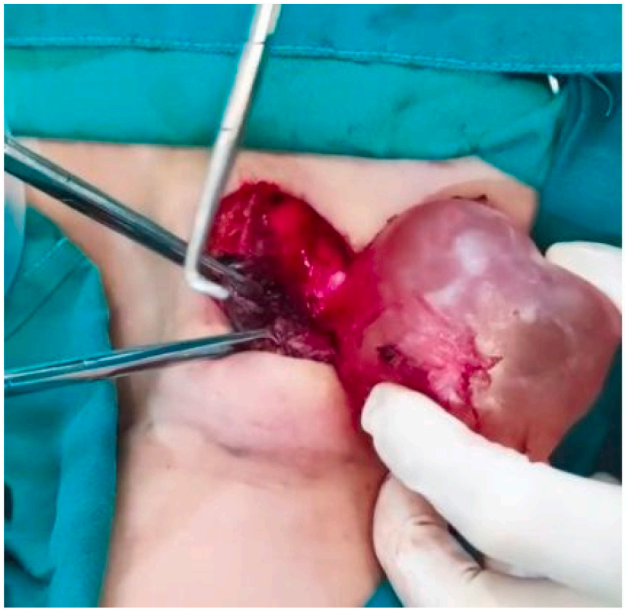

Fig. 4. Cystic-looking tumor on the right side of the cervical central compartment with pedicle that comes out from the piriform sinus through the cricopharyngeal musculature.

trachea (Fig. 5).

Excision of the mass was performed. The laryngotracheal complex was deviated to the left and the trachea was extrinsically compressed by a cystic-looking tumor on the right side of the central compartment. The pedicle-like tumor emerged from the pyriform sinus through the cricopharyngeal musculature, and inferior to the superior laryngeal nerve. The size of the mass was approximately 8 × 6X6 centimeters with retrothyroid location in the tracheoesophageal groove and extension of approximately 40 % of its volume into the superior mediastinum with adhesions to the right thyroid lobe. The procedure was successful without any complications (Figs. 6).

Her postoperative course was uneventful with no airway compromise. She tolerated diet well. Vital signs were stable, and she was

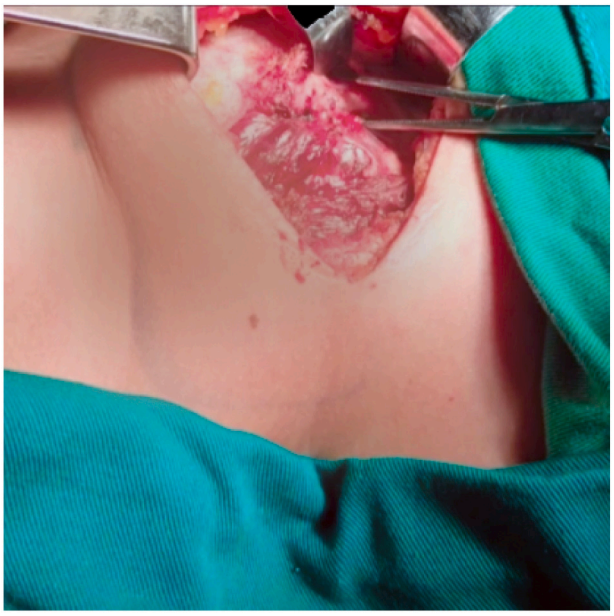

Fig. 5. Surgical bed with left-deviated laryngotracheal complex, trachea tapered by extrinsic compression.

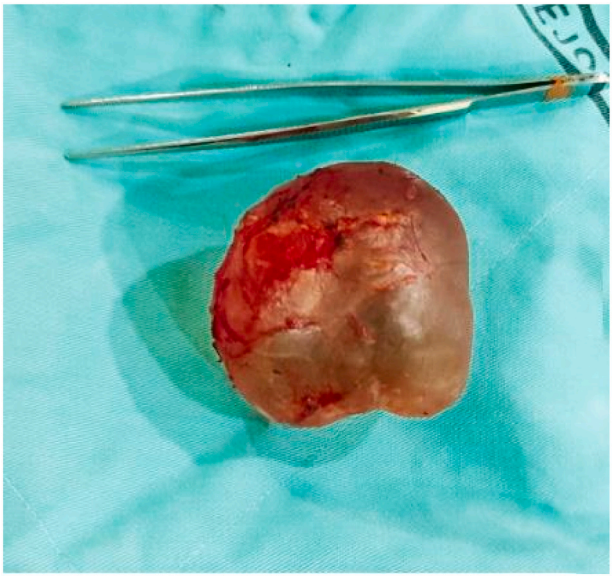

Fig. 6. 8 × 6 × cystic mass with an integrated external surface, whitish, translucent transparent liquid content, the parietal thickness of 0.1 cm, unilocular cavity, smooth whitish external surface.

discharged 48 h after surgery. The pathology report revealed a cystic formation with an integrated whitish external surface and translucent content of transparent liquid. The parietal thickness was 0.1 cm with a smooth unilocular cavity favoring branchial cleft.

At her 30 day follow-up, evaluating the patient on an outpatient basis, had no complications or recurrence with a favorable post-operation recovery.

3. Discussion

The branchial apparatus is composed of six mesodermal brachial arches separated by ectodermal clefts and endodermal pharyngeal pouches in the internal part. They appear between the fourth and fifth week of intrauterine growth. A developmental abnormality may occur causing the development of sinuses, fistulas, or cysts [1]. Branchial cleft cysts account for 20 % of pediatric cervical masses, usually diagnosed in late childhood, and only a small percentage present for the first time in adulthood [3].

Statistical data on the presentation of branchial cysts are variable with a prevalence of 8–10 % being first branchial cleft cyst, 67–97 % prevalence being second branchial cleft cysts, 2–8 % being third branchial cleft cyst. Fourth branchial cleft cyst are extremely rare, representing 1–4 % of all branchial apparatus pathologies [3–5].

Cystic malformations of the fourth brachial cleft originate at the apex of the pyriform sinus and extend to the thyroid region, passing below the superior laryngeal nerve. They are characterized by soft masses along the anterior border of the sternocleidomastoid muscle. At cervical level III, they can evolve to retropharyngeal abscesses, acute thyroiditis or they can cause compressive symptoms due to mass effect producing stridor, respiratory distress, odynophagia, and dysphagia if the cyst mass reaches large dimensions. In our case, the large size of the cystic mass was causing significant symptoms of respiratory distress and stridor. This required us to perform an excision of the cyst in an emergent manner. The origin of the cyst pedicle was observed in the piriform sinus extending through the cricopharyngeal musculature below the superior laryngeal nerve to the right thyroid lobe [3,4,6].

The presumptive diagnosis was or can be made through an adequate history and physical examination, using different imaging tests. Using an ultrasound, the mass can be observed like an image anaechogenic to hyperechoic. This technique is low cost and easily accessible, however, it does not provide enough information for surgical planning. CT scan allows for precise location of the lesion and provides unique details for resection of the cyst. It will show a thin regular wall structure without enhancement after contrast administration. In the case of infection the wall would show enhancement, increased thickness, and edema in surrounding tissues.

The CT scan in our patient allowed us to localize the cystic mass and plan the surgical intervention by observing the airway compression. A video laryngoscope was used to improve visibility and reduce the risk of failed endotracheal intubation [3,7].

The treatment of choice is complete surgical resection. Intervention is recommended in a non-inflammatory state, considering that previous infection or drainage makes dissection difficult. It is advisable to perform the cervicectomy in the parallel lines that coincide with skin folds avoiding incising the cyst [6].

The main risk of complications is injuries to the cranial nerves IX, X, XI, and XII, the superior laryngeal nerve, or great vessels. Recurrence after resection occurs in 3 % of patients after the first surgical intervention, increasing up to 14 % in the case of cystic mass infection and 21 % in cases of reoperations. In our case, the complete resection of the cyst was carried out without presenting complications. The patient was discharged in the immediate postoperative period and, to date, has not had recurrence [2].

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

The authors declare that we obtained permission from the ethics committee in our institution.

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Author contribution

León Sanguano Daysi: Conceptualization, Data curation, Formal analysis.

Jara Santamaria Christian: Conceptualization, Data curation, Formal analysis.

Jara Santamaria Bryan: Conceptualization, References.

Burbano Piñuela Liseth: Data curation.

Vasquéz Bracho Diego: Formal analysis.

Palacios Molina Antonio: Conceptualization, Data curation, Formal analysis.

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Registration of research studies

The authors declare that the patient gave her consent to publish this case, and, as this is a case report.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

The authors declare that there is no conflict of interest regarding the publication of this article.

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