Miscellaneous

Clinical-morphological aspects in the cyst of branchial arc II in children

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Abstract

Aspecte clinico-morfologice în chistul de arc II branchial la copil

Cazul prezentat ilucidează principiile de diagnostic și tratament chirurgical în chistul de arc II branchial la copii, autorii descriind și modificările morfopatologice caracteristice în această formațiune malformativă.
Raritatea cazului prezentat are ca scop de a atenționa necesitatea unui indice de suspiciune sporit în cazurile unor formațiuni cervicale laterale pentru un diagnostic adecvat și un tratament corespunzător, examenul histologic al piesei de rezecție fiind obligator.

Cuvinte-cheie: arc branchial, cleft branchial, chist cervical lateral

Abstract

The case illustrates the principles of diagnosis and treatment of branchial second arch cyst in children, the authors also described the morphopathological changes found in this malformative formation. The rarity of the presented case aims to warn of the need for an increased index of suspicion in cases of lateral cervical formations for proper diagnosis and treatment, the histological examination of the resection piece being mandatory.

Keywords: branchial arch, branchial cleft, lateral cervical cyst

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Introduction

Branchial arch abnormalities are a heterogeneous group of rare benign malformations, located along the sides of the face and neck in the form of sinuses, fistulas, cysts or cartilaginous reminiscences, resulting from involuntary errors of the branchial apparatus during embryonic periods. [14, 23]. These malformations constitute about 17 - 30% of the congenital formations of the head and neck in children [19, 21, 28], of these the most frequent (90-95% of cases) being diagnosed with abnormalities derived from the second branchial arch. The lesions derived from the first branchial arch constitute about 1-8%, and the reminiscences from arches 3 and 4 are much less common [25, 28, 29]. Branchial abnormalities are usually diagnosed in children up to 5 years of age, but can also be seen in adolescents or adults, showing an increase in volume or signs of infection [15].

Abnormalities of the second branchial arch, first described by Bailey in 1929 [10], are more commonly present in the submandibular space, and are found anywhere along the second tract of the branchial arch, which extends from the skin covering the supraclavicular fossa, between the internal and external carotid arteries to enter the pharynx at the level of the amygdala fossa [1]. We present a case of cervical brachial cyst, which after localization corresponds to the origin of the second branchial arch.

Case description

Patient R.E., 14 years old, was sent by the family doctor for a cervical volume formation on the right, being presumed cervical lymphadenitis. The examination found the presence of a formation located below the earlobe, along the sterno-cleido-mastoid muscle, partially mobile, elastic consistency, tense, painful to the touch, hyperemic regional skin. At this age, the formation was not observed. In the last month, the volume formation has obviously increased in size, becoming painful and causing discomfort at swallowing.

The data of the laboratory examination did not show any pathological deviations.

The ultrasound examination of the cervical region found the presence of a clear contour formation with dimensions 56x29 mm, with liquid and solid content.

Angiographic computed tomography revealed an ovoid, encapsulated, clearly contoured cystic formation, with protein liquid content (average density + 29UH), with dimensions (vert. X transv. X anterior) - 5.4cm x 3.1cm x 4.6cm. Post-contrast - moderate amplification at the level of the capsule up to + 100UH, without pathological amplification of the internal content, without intrastromal septa. Mass effect exerted by compression and cranial displacement of the left submendibular gland. Relationships with other anatomical structures: anterior and medial - platysma, subcutaneous and cutaneous tissue, medial - laryngeal cartilage, posterior - carotid space (including neurovascular bundle) and sternocleidomastoid muscle, superior - submendibular gland. Conclusion: CT imaging data suggestive for cystic formation of the soft tissue region of the right neck, possibly infected type 2 gill cyst (fig. 1).

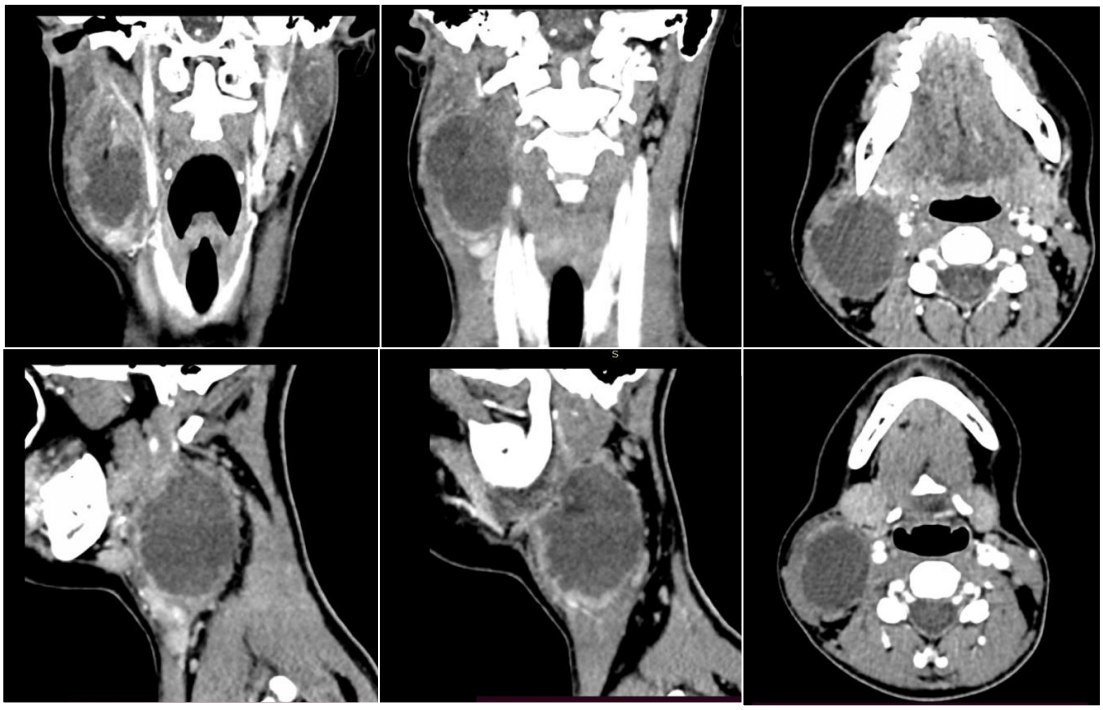

Fig. 1. Patient R.E., 14 years old. Preoperative CT: imaging data suggestive of a cystic formation of the soft tissues of the neck, possibly an infected type 2 gill cyst (explanations in the text).

Under general anesthesia with oro-tracheal intubation, surgery was performed, using the incision approach along the anterior edge of the sternocleidomastoid muscle, taking into account the large size of the formation. After incision of the skin and the superficial cervical fascia, plateau dissection, fascia colli propria dissection and lateral displacement of the sternocleidomastoid muscle with subsequent mobilization of the cystic formation were used, and its careful detachment from the adjacent neuro-vascular package was necessary. these anatomical structures, achieving complete excision of the cyst (fig. 2). We mention that intraoperatively a thin sinus tract was observed, which had a deep path that crossed the upper part of the carotid artery bifurcation to the pharynx, which, after mobilization, was ligated and excised. The operation ended with the restoration of the anatomical layers and the application of a drain. When the cyst was opened, gray liquid content was removed. On microbiological test of liquid content there was not detected the microbial flora, but the test from nasopharynx revealed streptococus oralis. The postoperative period was without events.

Histological examination revealed that the cystic formation was presented from fibrotic connective tissue, endowed with vascular network, lined with untradified multilayered squamous epithelium at 90% of the surface (fig. 3A). The subepithelial area was frequently presented by the presence of the dispersed lymphocyte cell component or of various intensity, in some areas, the cyst wall being presented by the parenchyma of the lymph node, characterized by the cortical area with follicular structures and small centers with insignificant reactive aspects. with lymphocyte content. The area of the ganglion hilum was represented as an integral part of the cystic wall, the respective area being designated adjacent to the epithelial envelope by the presence of a network of small-caliber lymphatic sinusoidal-tubular vessels lined with lymphocytes. Outside, the presence of the conjunctival capsule of the ganglion could be observed (fig. 3C). More frequently, the lymphatic tissue is attested in stratified strips in the capsule area (fig. 3B) or in the subepithelial area where the follicular structures were missing. In some areas, the lymphatic tissue was much thinner in the subepithelial area, with predilection in areas with a lower thickness of the cystic wall (fig. 3A, B). The ganglion, intimately attached to the cystic wall, presented the cortical area where there was an insignificant hyperplasia due to the germinal centers (fig. 3D).

In the samples taken from the adjacent area at the level of the sinus tract, which represents a tubular cord, the presence of the squamous-cellular epithelial covering, frequently detached, was attested. The lymphatic tissue had a dispersed or micro-nodular cellular manifestation associated with plasma cells and fibrillar-conjunctive reaction with sclerogenic changes (fig. 4A). Analogous changes were also found in the distal areas of the sinus tract with aspects of desquamation and partial detachment (fig. 4B).

We note that the tissue and ganglion structures involved in the structure of the cystic wall as well as the adherent ganglion were made of matured tissue, without atypia or proliferation, including the lack of metastatic neoplasms. Also, no active polymorphic-cellular inflammatory processes or the presence of PMN granulocytes were detected. No other types of tissue were found.

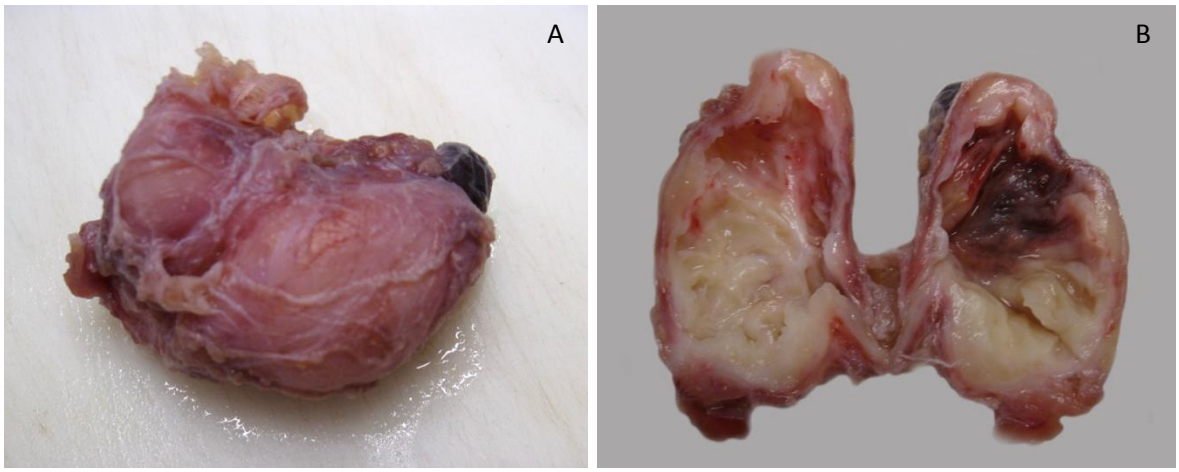

Fig. 2. External appearance of the cystic formation (A) to which a lymph node adheres and in section (B) where the internal folded-lacunar surface of the cyst is observed and communication with the sinus tract (arrow)

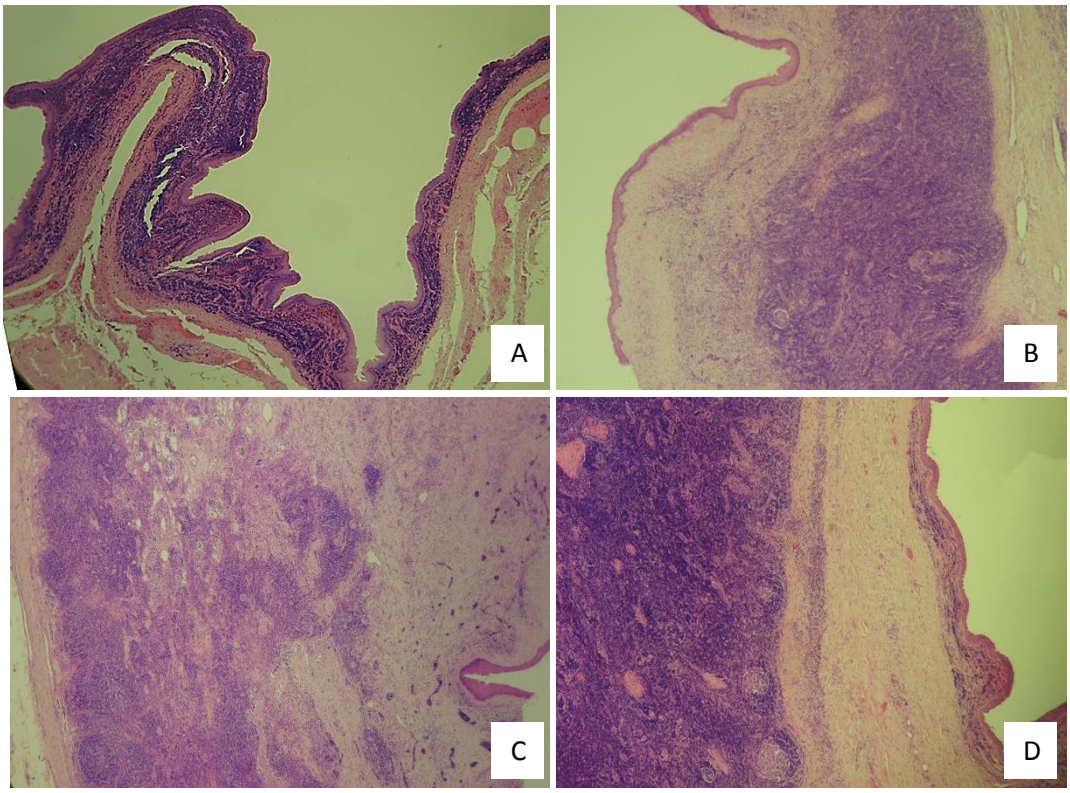

Fig. 3. Cystic wall structure: A - Folded appearance of the inner surface with strips of dense lymphatic tissue in the subepithelial area; B - Strips of lymphatic tissue in the area of the cystic wall; C - Lymph node integrated part of the cystic wall; D - Lymph node closely adhering to the cystic wall, follicular hyperplasia of the cortical follicles.

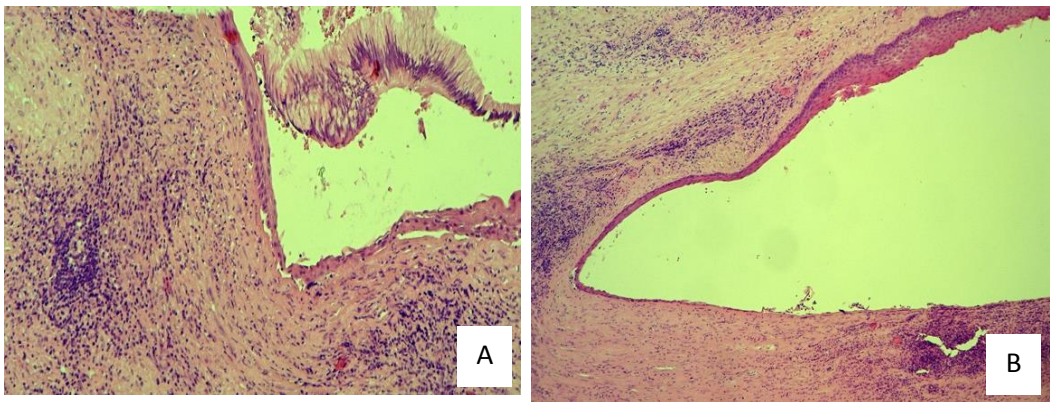

Fig. 4. The structure of the wall at the level of the tubular cord in the communication area. A - Fragments of multilayered prismatic epithelium; B - The blind area of the tubular cord partially lined with squamous cell epithelium with desquamation and detachment aspects..

Discussions

The first description of the branchial apparatus was attributed to Von Baer (1827). In 1828, Rathke described the development of pharyngeal arches in the human fetus, and in 1832, Acherson first described branchial fistula and proposed the notion of branchial cyst (Prasad S.C. et al., 2014).

The appearance of the branchial apparatus can be seen starting with the 4th week of embryonic development, being made up of six pairs of mesodermal arches separated inside the endoderm, which form the four pharyngeal sacs and outside ectoderm, which form the four branchial clefts (splits) .

The first two arcades proliferate, and the last two become rudimentary [3, 6]. Mesodermal tissue contains the artery, nerve, cartilage, and muscle for each branchial arch [12, 22].

The origin of branchial anomalies is a contested one, being proposed various theories:

- Congenital theory, which claims that branchial cysts develop from remnants of the embryonic branchial apparatus;
- Lymph node theories, according to which King (1949) concluded that the cyst comes from cystic changes of the parotid epithelium that are trapped in the upper cervical lymph nodes during the embryonic period;
- Branchial theory.

Bailey H. (1929) classified the anomalies of the second branchial cleft into four subtypes, taking into account the topography [1, 25, 27]:

Type I - the most superficial (located at the level of the superficial fascia), it is located along the anterior surface of the sternocleidomastoid muscle deep to platys, but has no contact with the carotid sheath;

Type II - the most common subtype in which the branchial cyst (located at the level of the deep fascia of the neck, in relation to the large vessels), is located anteriorly by the sternocleidomastoid muscle, posteriorly by the submandibular gland, adjacent and laterally by the carotid thorax;

Type III - branchial cyst (developed in the lodge of large cervical vessels) extends medially between the bifurcation of internal and external carotid arteries, lateral to the pharyngeal wall;

Type IV - the small cystic formation, which is located deep in the carotid sheath in the space of the pharyngeal mucosa and opens into the pharynx.

Some authors refer to the classification proposed by Proctor B. (1955) [26].

Clinically, cysts derived from the second branchial arch are manifested by the presence of a painless swelling located anterior to the sternocleidomastoid muscle between the mandibular angle and the clavicle. During an infection of the upper respiratory tract, an acute increase in size can be observed. Depending on the size, they can be manifested by compressive symptoms with dyspnea, stridor, dysphagia or dyssonia. Bilateral localization of these formations has been reported, and in some cases their presence is part of the branchio-oto-renal syndrome, an autosomal dominant disorder [16, 17].

Branchial abnormalities should be suspected in cases of unexplained recurrent throat infections or cervical volume formations and drainage in the area of the anterior edge of the sternocleidomastoid muscle. Although the complete anamnesis and objective examination may be adequate for diagnosis, preoperative imaging examination (fistulography, ultrasonography, computed tomography, and contrast magnetic resonance imaging) is helpful in confirming the clinical diagnosis [25].

Preoperative differential diagnosis includes thyroglossal duct cyst, cervical lymphadenitis, cervical abscess, toxoplasmosis, tuberculosis, dermoid cyst, dermal inclusion cyst, hydatid cyst, lymphangioma and malignant neoplasm [19].

From a histological point of view, branchial cysts are lined with stratified squamous epithelium (90% of cases), noting that in some cases pseudostratified columnar epithelium can be detected (8%), and in 2% their combination is found. Abundant lymphoid tissue is present in the connective tissue pears, which have germination centers separate from the epithelial cells of a thin basement membrane [2, 16, 30]. Some authors consider that branchial cysts are more frequently lined with squamous epithelium, while sinuses and fistulas tend to be lined with ciliated, columnar epithelium [7]. The increase in size of branchial cysts in short periods of time may be due to reactive hyperplasia of the lymphoid tissue [31]. Similar changes have been described by us. Cases describe cases of carcinoma that developed from the branchial cyst [18].

The microbiological profile of infected branchial abnormalities is not related to age and is different from that of throat infections, with monobacterial infections predominating. Empirical antibiotic treatment should cover Streptococcus species, including penicillin-resistant species, as well as clindamycin-resistant anaerobes. Possible empirical options are second- or third-generation cephalosporins (cefuroxime, ceftriaxone) with the addition of metronidazole against b-lactam-resistant anaerobes [9].

The treatment of choice for branchial abnormalities is radical surgical excision, the timing of surgery being controversial [5, 13]. In order to facilitate radical resection, some authors propose the intraoperative injection of fistulas and branchial cysts with a mixture of fibrin adhesive and methylene blue [20]. Some studies have demonstrated the efficacy and safety of ethanol ablation or sclerotherapy with dilute doxycycline or OK-432 as alternative treatment methods for patients with branchial abnormalities who refuse or are eligible for

surgical treatment [4, 8, 11]. In the specialty literature, a postoperative recurrence rate is reported that varies between 3% and 22%, the causes of recurrences being attributed to preoperative infection and incomplete excision [24].

Therefore, the case presented that branchial cysts in children may remain asymptomatic for long periods of time, but may reach large sizes in short periods of time, causing pain and compression of adjacent tissues. The rarity of the presented case aims to warn of the need for an increased index of suspicion in cases of lateral cervical formations for proper diagnosis and treatment, the histological examination of the resection piece being mandatory

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