Ognjen Čukić¹, Vladimir Milenković², Anđela Dimkić Milenković³, Dejan Moskovljević², Igor Lazić⁴, Marija Milenković⁴, Stojan Marić⁵, Radomir Vešović^{2,6}

BRANCHIAL CLEFT CYST: WHAT REALLY MATTERS? – CASE REPORT

ABSTRACT: Introduction: Branchial cleft anomalies are considered to develop from the branchial apparatus that did not completely obliterate during the embryogenesis of the head and neck. These anomalies pose a significant challenge in terms of surgical management, particularly followed by misdiagnosis due to its rarity. The aim of this case report was to present the review of literature and treatment of a rare anomaly such as a second branchial cleft cyst with a particular focus on clinical as well as histopathological aspects.

Case report: A 24-year-old woman was admitted to the otolaryngology department with a three-month history of painless right-sided neck swelling, following a previously resolved upper respiratory infection. The patient underwent complete excision of the cystic mass, with excellent cosmetic results and no signs of recurrence after a one-year follow-up.

Conclusion: Therefore, surgery should always be the gold standard of treatment. In patients aged over 40 years, cystic metastasis from the occult head and neck primary carcinoma must be considered.

Keywords: Branchial cleft cyst, cervical cyst, branchial anomaly, neck mass

¹ Ognjen Čukić, Clinic for Otorhinolaryngology, Military Medical Academy, Belgrade, Serbia.

² Vladimir Milenkovic, Clinic for Thoracic Surgery, University Clinical Center of Serbia, Belgrade, Serbia, vlada1309@gmail.com

³ Clinic for Burns, Plastic and Reconstructive Surgery, University Clinical Center of Serbia, Belgrade, Serbia.

⁴ Center for Anesthesiology, University Clinical Center of Serbia, Belgrade, Serbia.

⁵ Bežanijska Kosa University Hospital Medical Center, Department for General Surgery, Belgrade, Serbia.

⁶ University of Belgrade, Faculty of Medicine, Belgrade, Serbia.

Introduction

Branchial cleft anomalies are considered to develop from remnants of branchial apparatus which has a fundamental role in the development of head and neck structures during early embryonic life. During embryogenesis, the second branchial arch fuses with the third and fourth arches to form the cervical sinus and disappears afterward. Therefore, a profound understanding of the developmental abnormalities is essential to recognize entities such as cysts, sinuses, and fistulas as well as various combinations of these [1]. Second branchial cleft cyst is the most common type of branchial anomaly and the second most common congenital cystic anomaly of the neck after the thyroglossal duct cyst. Ascherson was the first one to describe the branchial fistulas in 1832 as a result of the incomplete obliteration of branchial pouches [2, 3]. The exact annual incidence in general population is unknown, and most cases are diagnosed in patients between the age of 20 to 40 years [4, 5]. There is no strong evidence for hereditary predisposition, as well as gender or ethnic predilection, although cases of familial clustering have been described [4, 6, 7]. They usually become apparent in young adulthood as a painless neck mass located alongside the anterior border and the upper third of the sternocleidomastoid muscle (SCM). Branchial cleft cysts are histopathologically benign, but with the potential for significant morbidity in cases of infection, mass effect, cosmetic appearance, and potential surgical complications. Therefore, these rare anomalies pose various clinical challenges, including correct preoperative diagnostics, treatment of the possible infection, and complete surgical excision. The main treatment for this disease includes meticulous cyst removal and follow-ups after surgery. The aim of this report was to present a case of a rare anomaly such as a second branchial cleft cyst.

Case report

A 24-year-old female patient was admitted to our department with a three-month history of painless right-sided neck swelling, following a previously resolved upper respiratory infection. Her medical history was unremarkable, and there was no history of smoking or alcohol consumption. Clinical examination revealed a soft, ovoid, mobile, and well-defined neck mass just anterior to the upper third of the right sternocleidomastoid muscle, which was measuring 3.5cm, with the intact overlying skin (Figure A). She underwent fiberendoscopy of the upper aerodigestive tract which was normal and neck ultrasonography that showed a hypoechoic thin-walled cystic mass. Based on these findings, the patient was taken to the operative room and the mass was completely excised under general anesthesia. The cyst was exposed and subsequently dissected through the right-sided lateral cervicotomy (Figures B and C). Following complete hemostasis, the wound was closed in layers and an active suction drain was

inserted. The histopathological examination of the specimen confirmed the diagnosis of a branchial cyst. Microscopically, the cystic cavity was surrounded by squamous epithelium with dense lymphoid infiltrate, which formed germinal centers (Figure D). The postoperative period was uneventful and there were no signs of recurrence after one-year follow-up.

Discussion

Branchial cysts are congenital encapsulated, epithelial-lined cavities resulting from incomplete obliteration of the branchial apparatus during embryogenesis in the lateral part of the neck [8]. There are several theories about the origin of branchial cysts. The most accepted one is the theory of failure of involution of the cervical sinus, which is formed by the second, third and fourth branchial arches, and persisting of ectodermal epithelial cells within these rudimentary structures. Pockets of these persisting ectodermal cells may form branchial cysts later in life [9]. Second branchial cleft cysts are most commonly located along the anterior border in the upper third of the sternocleidomastoid muscle, although various atypical localizations in the neck have been described, including the parotid gland [10]. The presence of the lymphoid tissue in the cyst wall suggests the inclusion theory, which postulates that branchial cysts result from epithelial inclusions within a cervical lymph node [11]. Furthermore, this theory explains why most branchial cysts have no internal opening, are almost unknown in neonates, and the peak age of presentation is much later in life than in other congenital neck lesions. The lymphoid tissue response in the cyst lining may explain the sudden enlargement of the cyst during an episode of the upper airway infection.

A typical clinical finding is of a soft, painless and compressible neck mass, situated at the anterior border of the sternocleidomastoid muscle, between the mandibular angle and the clavicle. The patient may report swelling of long duration with periods of waxing and waning. An acute increase in size can occur during an upper respiratory tract infection, while cyst infection and overlying skin inflammation may also occur [12]. Differential diagnoses include lymphangioma, thyroglossal duct cyst, dermoid cyst, cystic hygroma, laryngocele, lymphadenitis, and various mesenchymal tumors. Special attention should be given to the patients aged over 40 years, in whom cystic metastasis from the occult head and neck primary carcinoma must be considered [13, 14].

The diagnosis is primarily based on the history and the clinical examination. Ultrasonography as an initial imaging method will usually demonstrate the anechoic, thin-walled cyst, while computed tomography may be used in more extensive lesions, specifically to assess the relationship of the cyst to the major neural and vascular structures in the neck [15]. The treatment of choice for branchial cyst is complete surgical excision [16]. The dissection can be relatively straight-forward in previously

non-infected cysts, and the surgeon must dissect completely around the cyst bed to exclude any associated fistula or tract [17]. If identified, the tract should be followed, dissected and excised in 'en bloc' manner with the cyst [18]. Thorough knowledge of the neck anatomy is mandatory to avoid injury to the surrounding neck structures [9]. Wider skin incision facilitates cyst removal and identification of the major cervical neurovascular structures, but will eventually leave a larger scar. Therefore the surgeon must carefully consider the visually pleasing cosmetic outcome but without compromising the surgical radicality, as the majority of the patients are young adults.

Stulner et al. have noted that appropriate investigation and management by a team of experienced head and neck surgeons are necessary if a satisfactory outcome is to be achieved [19]. Additionally, cystic metastases from occult head and neck primaries, especially squamous cell carcinoma (SCC) of the palatine tonsil, can clinically and radiographically mimic branchial cleft anomalies [13, 14, 20]. Physicians must remain vigilant in obtaining data on excessive smoking or alcohol consumption, especially in older male patients, as these are all known risk factors for upper airway malignancy. Additionally, a substantial proportion of cystic neck metastases arise from human papilloma virus-related oropharyngeal carcinoma which, unlike traditional SCC, occurs in a younger population who are frequently non-smokers and not heavy drinkers [21]. Therefore all patients aged 40 years and over should be thoroughly examined by an ear, nose and throat specialist, including endoscopy of the upper aerodigestive tract in general anesthesia. If the endoscopy proves non-diagnostic, bilateral tonsillectomy is recommended [22].

Conclusion

Knowledge of the head and neck embryonic development, early diagnosis, as well as appropriate surgical treatment, are the key steps for a satisfactory outcome in the management of the branchial cysts. Complete cyst removal by careful surgical dissection is the current practice standard and must be performed by a skilled neck surgeon. In patients over 40 years, the possibility of cystic lymph node metastasis from the occult head and neck primary carcinoma should be excluded. FIGURES:



FIGURE A. Clinical examination revealed a soft, ovoid, mobile, and well-defined neck mass just anterior to the upper third of the right sternocleidomastoid muscle, which was measuring 3.5-cm with the intact overlying skin.



FIGURE B. Cyst exposed and dissected through the overlying skin crease incision of the right side of the neck.



FIGURE C. Macroscopic appearance of the specimen.



FIGURE D. Histologically, the cystic cavity (asterisk) is surrounded by squamous epithelium with dense lymphoid infiltrate, which forms germinal centers (arrows; hematoxylin and eosin staining).

Reference:

- Papadogeorgakis N., Petsinis V., Parara E., Papaspyrou K., Goutzanis L., Alexandridis C. Branchial cleft cysts in adults. Diagnostic procedures and treatment in a series of 18 cases. Oral Maxillofac Surg. 2009 Jun; 13(2): 79–85.
- 2. Ascherson GM. De fistulis colli congenitis adjecta fissurarum branchialium in mammalibus avibusquehistoria succincta. C. H. Jonas, Berolini 1832: 1–21.
- 3. Golledge J., Ellis H. The aetiology of lateral cervical (branchial) cysts: past and present theories. J Laryngol Otol. 1994 Aug; 108(8): 653–9.
- 4. Coste AH, Lofgren DH, Shermetaro C. Branchial cleft cyst. [Updated 2022 Apr 14]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2022 Jan. Available from: https://www.ncbi.nlm.nih.gov/books/NBK499914/
- 5. Panchbhai AS, Choudhary MS. Branchial cleft cyst at an unusual location: a rare case with a brief review. Dentomaxillofac Radiol. 2012 Dec; 41(8): 696–702.
- 6. Thomaidis V., Seretis K., Tamiolakis D., Papadopoulos N., Tsamis I. Branchial cysts. A report of 4 cases. Acta Dermatovenerol Alp Panonica Adriat. 2006 Jun; 15(2): 85–9.
- 7. Anand TS, Anand CS, Chaurasia BD. Seven cases of branchial cyst and sinuses in four generations. Hum Hered. 1979; 29(4): 213–6.
- Saussez S., De Maesschalk T., Mahillon V., Filleul O., Louryan S. Second branchia; l cyst in the parapharyngeal space: a case report. Auris Nasus Larynx. 2009 Jun; 36(3): 376–9.
- 9. Chavan S., Deshmukh R., Karande P., Ingale Y. Branchial cleft cyst: A case report and review of literature. J Oral Maxillofac Pathol. 2014 Jan; 18(1): 150.
- Nicollas R., Guelfucci B., Roman S., Triglia JM. Congenital cysts and fistulas of the neck. Int J Pediatr Otorhinolaryngol. 2000 Sep; 55(2): 117–24.
- Simo R., Jeannon JP, Ofo E. Benign neck disease. In: Watkinson JC, Clarke RW, editors. Scott-Brown's Otorhinolaryngology Head and Neck Surgery. 8th ed. Boca Raton: CRC Press; 2019; p. 611–2.
- Little JW, Rickles NH. The histogenesis of the branchial cyst. Am J Pathol. 1967 Mar; 50: 533-47.
- Micheau C., Klijanienko J., Luboinski B., Richard J. So-called branchiogenic carcinoma is actually cystic metastases in the neck from a tonsillar primary. Laryngoscope. 1990 Aug; 100(8): 878–83.
- Yehuda M., Schechter ME, Abu-Ghanem N., Golan G., Horowitz G., Fliss DM, Abu-Ghanem S. The incidence of malignancy in clinically benign cystic lesions of the lateral neck: our experience and proposed diagnostic algorithm. Eur Arch Otorhinolaryngol. 2018 Mar; 275(3): 767–73.
- Janicke S., Kettner R., Kuffner HD. A possible inflammatory reaction a lateral neck cyst (branchial cyst) because of odontogenic infection. Int J Oral Maxillofac Surg .1994 Dec; 23(6 Pt 1): 369–71.
- Cunningham M.J. The management of congenital neck masses. Am J Otolaryngol. 1992; 13: 78–92.

- 17. Choi SS, Zalzal GH. Branchial anomalies: a review of 52 cases. Laryngoscope. 1995 Sep; 105(9 Pt 1): 909–13.
- Bellakhdar M., Haouas J., El Abed W., Ghammem M., Kermani W., Abdelkefi M. Second branchial cleft cyst: about 34 cases. Tunis Med. 2018 Dec; 96(12): 888–92.
- 19. Stulner C., Chambers PA, Telfer MR, Corrigan AM. Management of first branchial cleft anomalies: report of two cases. Br J Oral Maxillofac Surg. 2001 Feb; 39(1): 30–3.
- 20. Briggs RD, Pou AM, Schnadig VJ. Cystic metastasis versus branchial cleft carcinoma: a diagnostic challenge. Laryngoscope. 2002 Jun; 112(6): 1010–4.
- 21. Wratten C., Anne S., Tieu MT, Kumar B., Eisenberg R. The dangers of diagnosing cystic neck masses as benign in the era of HPV-associated oropharyngeal cancer. Med J Aust. 2015; 203(9): 371–2.
- 22. McQuone SJ, Eisele DW, Lee DJ, Westra WH, Koch WM. Occult tonsillar carcinoma in the unknown primary. Laryngoscope. 1998 Nov; 108(11 Pt 1): 1605–10.