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BRANHIJALNA CISTA VRATA: ŠTA JE ZAISTA VAŽNO? – PRIKAZ SLUČAJA

SAŽETAK: **Uvod:** Smatra se da se anomalije branhijalnog raseca razvijaju iz branhijalnog aparata koji nije u potpunosti obliterisao tokom embriogeneze glave i vrata. Ove anomalije predstavljaju značajan izazov u pogledu hirurškog lečenja, posebno praćene pogrešnim dijagnozama zbog svoje retkosti. Cilj ovog prikaza slučaja je da se predstavi pregled literature i lečenje retke anomalije kao što je cista porekla drugog branhijalnog raseca, sa posebnim osvrtom na kliničke, kao i histopatološke aspekte.

Prikaz slučaja: Žena, starosne dobi 24 godine, primljena je na naše odeljenje sa nalazom bezbolnog otoka desne strane vrata koji se javio tri meseca pre prijema, a nakon prethodno sanirane infekcije gornjih disajnih puteva. Pacijentkinji je učinjena potpuna ekscizija cistične promene, sa odličnim kozmetičkim rezultatima i bez znakova recidiva nakon jednogodišnjeg praćenja.

Zaključak: Imajući u vidu postignute rezultate, hirurški tretman predstavlja zlatni standard lečenja. Kod pacijenata starijih od 40 godina moraju se uzeti u obzir cistične metastaze iz okultnog primarnog karcinoma glave i vrata.

Ključne reči: cista branhijalnog raseca, cervikalna cista, branhijalna anomalija, tumefakt na vratu

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Uvod

Smatra se da se anomalije branhijalnog rasepa razvijaju iz ostataka branhijalnog aparata koji ima fundamentalnu ulogu u razvoju struktura glave i vrata tokom ranog embrionalnog razvoja. Tokom embriogeneze, drugi škržni luk se spaja sa trećim i četvrtim lukovima i formira cervikalni sinus i nakon toga nestaje. Stoga je duboko razumevanje razvojnih abnormalnosti od suštinskog značaja za prepoznavanje entiteta kao što su ciste, sinusi i fistule, kao i različite kombinacije prethodno navedenih entiteta [1]. Cista drugog branhijalnog rasepa je najčešći tip anomalije branhijalne anomalije i druga najčešća kongenitalna cistična anomalija vrata posle ciste tiroglosalnog kanala. Ašerson je bio prvi koji je opisao branhijalne fistule 1832. godine kao rezultat nepotpune obliteracije branhijalnih kesica (sakulusa) [2, 3]. Tačna godišnja incidencija u opštoj populaciji nije poznata, a većina slučajeva se dijagnostikuje kod pacijenata starosti između 20 i 40 godina [4, 5]. Ne postoje čvrsti dokazi za naslednu predispoziciju, kao ni za pol ili etničku pripadnost, iako su opisani slučajevi porodičnog nasleđivanja [4, 6, 7]. Obično postaju očigledni u mladosti kao bezbolna tumefakcija na vratu koja se nalazi duž prednje granice i gornje trećine sternokleidomastoidnog mišića (SCM). Ciste branhijalnog rasepa su histopatološki benigne, ali sa potencijalom za značajan morbiditet u slučajevima infekcije, kompresije okolnih struktura zbog svoje veličine, kozmetičkog izgleda i potencijalnih hirurških komplikacija. Stoga, ove retke anomalije predstavljaju složen klinički izazov u dijagnostici i lečenju, uključujući ispravnu preoperativnu dijagnostiku, lečenje moguće infekcije i potpunu hiruršku eksciziju. Osnovni vid lečenja uključuje pažljivo uklanjanje ciste i praćenje nakon operacije. Cilj ovog prikaza slučaja bio je da se predstavi slučaj retke anomalije kao što je cista drugog branhijalnog rasepa.

Prikaz slučaja

Pacijentkinja starosne dobi 24 godine primljena je na naše odeljenje sa kliničkim nalazom bezbolnog otoka na desnoj strani vrata koji se javio tri meseca pre prijema, a nakon prethodno sanirane infekcije gornjih disajnih puteva. Njena anamneza je bila neupadljiva, bez podataka o konzumiranju duvana ili alkohola. Kliničkim pregledom otkrivena je meka, jajolika, pokretna i dobro definisana tumefakcija na desnoj strani vrata, neposredno ispred gornje trećine sternokleidomastoidnog mišića, veličine 3,5 cm, sa intaktnom kožom iznad promene (Slika A). Urađena je fiberendoskopija gornjeg aerodigestivnog trakta, koja je bila normalna, i ultrasonografija vrata, koja je pokazala hipoehogenu cističnu masu tankih zidova. Na osnovu ovih nalaza, pacijent je uveden u operacionu salu i cistična masa je potpuno ekscidirana u opštoj anesteziji. Cista je u celosti prikazana i potom odstranjena kroz desnu bočnu cervikotomiju (Slike B i C). Nakon korektne hemostaze, rana je slojevito zatvorena i plasiran je dren spojen

na aktivnu aspiraciju. Histopatološkim pregledom uzorka potvrđena je dijagnoza branhijalne ciste. Mikroskopski, cistična šupljina je bila okružena skvamoznim epitelom sa gustim limfoidnim infiltratom, koji je formirao germinativne centre (Slika D). Postoperativni period je protekao bez komplikacija i nije bilo znakova recidiva nakon jednogodišnjeg praćenja.

Diskusija

Branhijalne ciste su kongenitalne inkapsulirane, epitelno obložene šupljine koje nastaju usled nepotpune obliteracije branhijalnog aparata tokom embriogeneze u bočnom delu vrata [8]. Postoji nekoliko teorija o poreklu branhijalnih cista. Najprihvaćenija je teorija o neuspehu involucije cervikalnog sinusa, koji se formira od drugog, trećeg i četvrtog branhijalnog luka, i perzistiranja ektodermalnih epitelnih ćelija unutar ovih rudimentarnih struktura. Džepovi ovih perzistentnih ektodermalnih ćelija mogu kasnije u životu da formiraju branhijalne ciste [9]. Ciste drugog branhijalnog rasepa najčešće se nalaze duž prednje granice u gornjoj trećini sternokleidomastoidnog mišića, iako su opisane različite atipične lokalizacije na vratu, uključujući parotidnu žlezdu [10]. Prisustvo limfoidnog tkiva u zidu ciste sugerise inkluzionu teoriju, po kojoj branhijalne ciste nastaju kao posledica epitelnih posuvraćanja unutar cervikalnog limfnog čvora [11]. Štaviše, ova teorija objašnjava zašto većina branhijalnih cista nema unutrašnji otvor, skoro su nepoznate kod novorođenčadi, a najviša incidenca javljanja je mnogo kasnije u životu nego kod drugih urođenih lezija vrata. Odgovor limfoidnog tkiva u sluznici ciste može objasniti iznenadno uvećanje tokom epizode infekcije gornjih disajnih puteva.

Tipičan klinički nalaz je mekana, bezbolna i kompresibilna masa na vratu, smeštena na prednjoj ivici sternokleidomastoidnog mišića, između mandibularnog ugla i klavikule. Pacijent može prijaviti progresivno oticanje navedene regije sa periodima smanjenja otoka ili čak nestajanja. Akutno povećanje veličine može se javiti tokom infekcije gornjih disajnih puteva, dok se mogu javiti i infekcija ciste i zapaljenje kože iznad promene [12]. Diferencijalna dijagnoza uključuje limfangiom, cistu tireoglosalnog kanala, dermoidnu cistu, cistični higrom, laringocelu, limfadenitis i razne mezenhimalne tumore. Posebnu pažnju treba posvetiti pacijentima starijim od 40 godina, kod kojih se moraju uzeti u obzir cistične metastaze iz okultnog primarnog karcinoma regije glave i vrata [13, 14].

Dijagnoza se prvenstveno zasniva na anamnezi i kliničkom pregledu. Ultrasonografija kao inicijalna metoda vizualizacije obično će pokazati anehogenu cistu sa tankim zidovima, dok se kompjuterska tomografija može koristiti u ekstenzivnijim lezijama, posebno za procenu odnosa ciste sa glavnim nervnim i vaskularnim strukturama na vratu [15]. Tretman izbora za branijalnu cistu je potpuna hirurška ekscizija [16]. Disekcija može biti relativno laka u pravom sloju kod prethodno

neinficiranih cista, a hirurg mora u potpunosti da isprepariše ležište ciste da bi isključio bilo kakvu pridruženu fistulu ili komunikaciju sa okolnim strukturama [17]. Ako se identifikuje komunikaciju treba pratiti, preparirati i ekscidirati „en bloc“ zajedno sa cistom [18]. Temeljno poznavanje anatomije vrata je obavezno da bi se izbegle povrede okolnih struktura [9]. Širi rez na koži olakšava uklanjanje ciste i identifikaciju glavnih neurovaskularnih struktura vrata, ali će na kraju ostaviti veći ožiljak. Zbog toga hirurg mora pažljivo razmotriti vizuelno prihvatljiv kozmetički efekat operacije, ali bez ugrožavanja hirurške radikalnosti, pošto su većina pacijenata mlade odrasle osobe.

Stulner i saradnici su primetili da su odgovarajuća ispitivanja i vođenje slučaja od strane tima iskusnih hirurga za glavu i vrat neophodni ako se želi postići zadovoljavajući ishod [19]. Pored toga, cistične metastaze iz okultnih primarnih tumora glave i vrata, posebno karcinom skvamoznih ćelija (SCC) nepčanog krajnika, mogu klinički i radiografski da oponašaju anomalije branhijalnog rasepa [13, 14, 20]. Lekari moraju biti oprezni u prikupljanju podataka o prekomernom konzumiranju duvana ili alkohola, posebno kod muškaraca starije životne dobi, jer su to dobro poznati faktori rizika za malignitete gornjih disajnih puteva. Pored toga, značajan deo cističnih metastaza na vratu su metastaze karcinoma orofaringealne regije povezanih sa humanim papiloma virusom koji se, za razliku od tradicionalnog SCC, javlja kod mlade populacije koji su uglavnom nepušači i ne konzumiraju velike količine alkohola [21]. Zbog toga svi pacijenti stariji od 40 godina treba da budu detaljno pregledani od strane specijaliste za uho, grlo i nos, uključujući endoskopiju gornjeg aerodigestivnog trakta u opštoj anesteziji. Ako je endoskopski nalaz nejasan preporučuje se bilateralna tonzilektomija [22].

Zaključak

Poznavanje embrionalnog razvoja glave i vrata, rana dijagnoza, kao i odgovarajući hirurški tretman, ključni su koraci za zadovoljavajući ishod u zbrinjavanju branhijalnih cista. Potpuno uklanjanje ciste pažljivom hirurškom disekcijom je zlatni standard u praksi i mora ga izvesti iskusni hirurg za regiju glave i vrata. Kod pacijenata starijih od 40 godina treba isključiti mogućnost cističnih metastaza u limfnim čvorovima vrata iz okultnog primarnog karcinoma glave i vrata.

SLIKE:



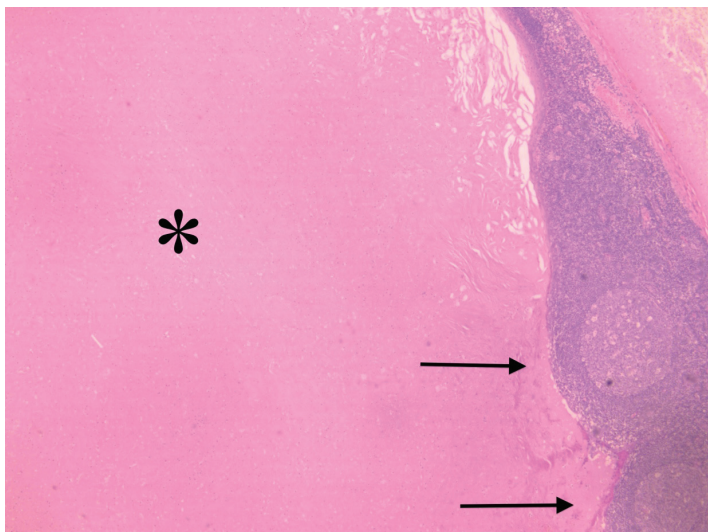
SLIKA A. Kliničkim pregledom uočena je meka, jajolika, pokretna i dobro definisana tumefakcija na vratu neposredno ispred gornje trećine desnog sternokleidomastoidnog mišića, koja je bila veličine 3,5 cm sa intaktnom kožom.



SLIKA B. Cista otkrivena i ispreparisana kroz rez na naboru kože na desnoj strani vrata.



SLIKA C. Makroskopski izgled preparata.



SLIKA D. Histološki, cistična šupljina (zvezdica) je okružena skvamoznim epitelom sa gustim limfoidnim infiltratom, koji formira germinalne centre (strelice; bojenje hematoxilinom i eozinom).

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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

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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 fiberoendoscopy 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.

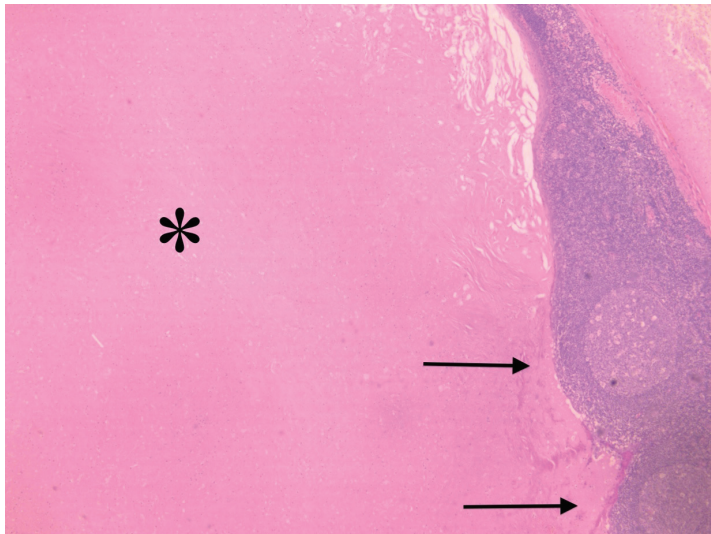


FIGURE D. Histologically, the cystic cavity (asterisk) is surrounded by squamous epithelium with dense lymphoid infiltrate, which forms germinal centers (arrows; hematoxylin and eosin staining).

Reference:

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