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Atypical Ist branchial cleft fistula – exploring the problem based on a patient's case

Przetoka I łuku skrzelowego o nietypowym przebiegu – omówienie problemu na podstawie przypadku pacjenta

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Keywords:

Summary

Fistula or cyst of first branchial arch is not very common aberration comparing to other fistulas of head and neck. Presentation of that congenital abnormality can be lump or small hole on the skin in triangle between auricula, mentum and hyoid bone. Usually diagnosed after upper respiratory tract infection when the cyst enlarges and start to secretion of mucus. Additional radiological examination as Ultrasound, CT scan with contrast injected into fistula if possible or MRI could be helpful with diagnostic and planning the surgical treatment of the patient. The following step is only one. Treatment of this congenital abnormalities is surgery, but the scope of surgery depends on the localization, presence of the inflammation and size of the lesion. Problem with diagnostic, choice of proper radiological examination and next planning the right treatment we would like to present as a care report of 17 years old girl admitted to Clinical Department of Pediatric Otorhinolaryngology in University Clinical Center Medical University in Warsaw.

Słowa kluczowe:

Streszczenie

Przetoka lub torbiel pierwszego łuku skrzelowego nie jest zbyt częstą aberracją w porównaniu z innymi przetokami głowy i szyi. Objawem tej wrodzonej nieprawidłowości może być guzek lub mała dziura na skórze w trójkącie między małżowiną uszną, bródką i kością gnykową. Zwykle diagnozuje się ją po infekcji górnych dróg oddechowych, gdy torbiel powiększa się i zaczyna wydzielać śluz. Dodatkowe badanie radiologiczne, takie jak: USG, tomografia komputerowa z kontrastem wstrzykniętym do przetoki, jeśli to możliwe, lub rezonans magnetyczny mogą być pomocne w diagnostyce i planowaniu leczenia operacyjnego pacjenta. Następnym krokiem w postępowaniu jest tylko jedyna możliwość. Leczenie tych wrodzonych wad polega na zabiegu chirurgicznym, ale zakres operacji zależy od lokalizacji, obecności stanu zapalnego i wielkości zmiany. Problem z diagnostyką, doborem właściwego badania radiologicznego, a następnie zaplanowaniem właściwego leczenia chcielibyśmy przedstawić w formie raportu z opieki nad siedemnastoletnią dziewczynką przyjętą do Kliniki Otorynolaryngologii Dziecięcej Dziecięcego Szpitala Klinicznego Uniwersyteckiego Centrum Medycznego Warszawskiego Uniwersytetu Medycznego.

Introduction

This study was conducted to analyze the literature focused on congenital abnormalities of head and neck as branchial cleft cysts and fistulas. As well as we want to present the case of a patient whose fistula has an abnormal localization. The patient has sixteen years old in the day of admission to Clinical Department of Pediatric Otorhinolaryngology in Pediatric Teaching Hospital of University Clinical Center Medical University in Warsaw.

Case report

Seventeen-year-old girl was admitted to Clinical Department of Pediatric Otorhinolaryngology in University Clinical Center Medical University in Warsaw with diagnosis of

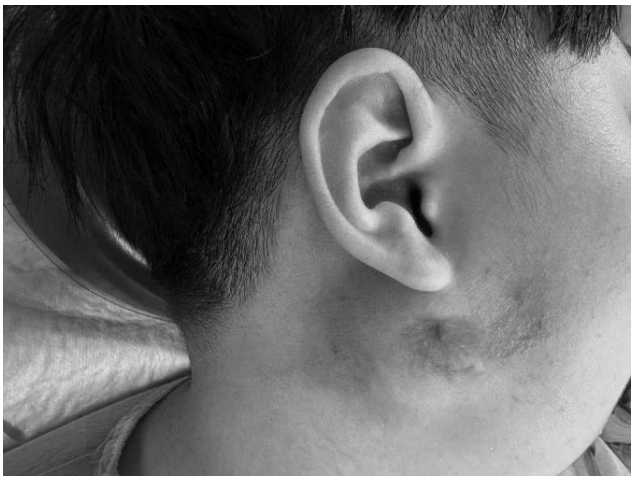

Fig. 1.

buccal cyst (fig. 1). During the interview upon admission, the patient reported a small hole on the skin in region of right mandibular angle, recurrent inflammation of the right side of neck treated with multiple antibiotic therapy usually with trimethoprim and sulfamethoxazole with temporary improvement, secretion of purulent content which was leaking outside through epidermal ending of fistula and as well through mucosal ending in pharynx. Patient reported also minimum one episode of drainage of abscess of the neck on the right. No facial nerve palsy was reported. Due to secretion from fistula, we take microbiological samples and start empirical antibiotic therapy starting with amoxicillin with clavulanic acid. Patient was referred to radiological diagnostic started with ultrasonography of neck and after the USG the contrast enhanced magnetic resonance imaging was planned. In MRI the result was: "The examination revealed the presence of a fistula tract in the soft tissues of the neck on the right side. The tract runs through the parapharyngeal space and the parotid space, and from the medial side, it has a narrow band of connection with the right palatine tonsil. Then it is quite wide (7-9 mm in diameter), runs upwards, laterally and backwards. The tract runs forwards from the external carotid artery in its initial section, running directly to it. Then, in the section between the superficial and deep lobe of the parotid gland, the tract lies forwards from the facial nerve and is directly adjacent to it. Then the tract turns sharply downwards and narrows. The last approx. 2.5 cm has a narrow lumen of approx. 1-2 mm and an external opening forwards from the tragus of the ear. A very narrow fistula to the bottom of the external auditory canal is also possible. The image raises suspicion of a first branchial pouch cyst with a fistula to the skin surface and penetration into the right palatine tonsil". Patient spend in our clinic 2 weeks due to antibiotic therapy and all consultation needed to qualify to surgery. As a result of athypical morphology of fistula to plan the surgery we decided to make fistulograhpy which is Computed Tomography with contrast injected directly to epidermal ending of fistula to assess the exact course of the whole fistula. In CT examination result was: "Neck CT scan performed in a single phase after administration of a contrast agent (Visipaque in dilution) through a drain to the external opening of the fistula, without intravenous administration of the agent. The examination revealed two parallel fistula channels penetrating from the openings on the skin deep into the parotid gland P and soft tissues of the neck. The fistulas with a very tortuous course, directed obliquely upwards and medially at a height of 2 cm from the skin openings, tangentially to the right styloid process (medially from the parotid gland), then a present tortuous blindly ended dilatation with total dimensions of approx. 50 x 9 x 7 mm extending downwards to the area of the lower pole of the right parotid gland. Medially, the described lesion extends at the level of the skin openings towards the right palatine tonsil, at the top of which it opens into the nasopharynx (dimensions of the area approx. 35 x 12 x 10 mm)" (fig. 2-4). After visualization

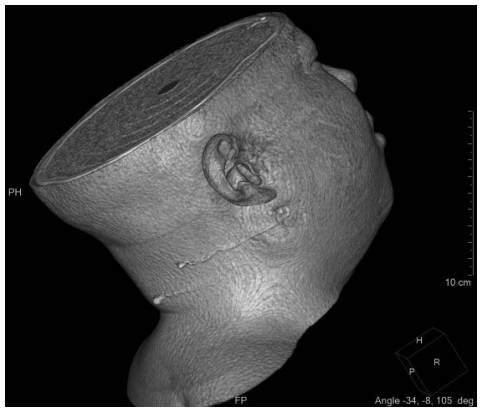

Fig. 2.

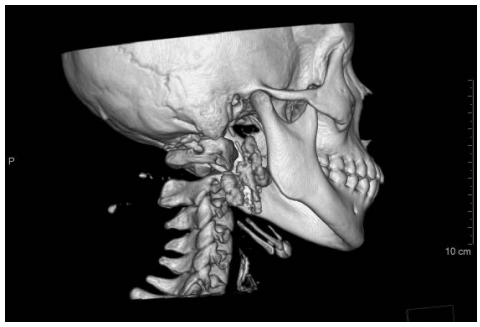

Fig. 3.

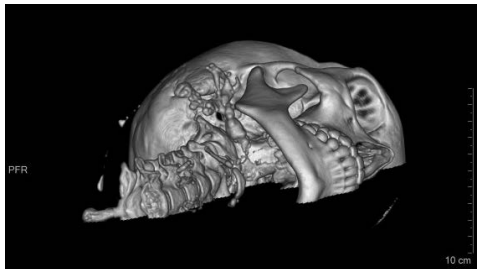

Fig. 4.

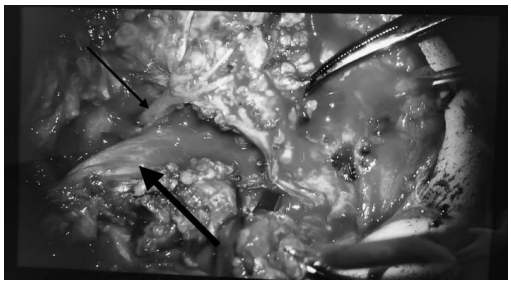

Fig. 5.

of exact fistula, the surgery was planned and as a atypical course patient was informed that surgery will include partial parotidectomy with high risk of intraoperative bleeding due to connection with external carotid artery and need to operate through parapharyngeal space. The surgery on the begin was very challenging with localization and identification of the facial nerve, after dissection of main trunk of facial nerve the superficial parotidectomy was performed and fistula was successfully dissected from facial nerve (fig. 5). In the parapharyngeal space after identification of external carotid artery the fistula was dissected safely to internal ending near pharyngeal tonsil. After surgery the patient feels well and only with partial facial nerve insufficiency of marginal branch III stage of Houde-Brackmann scale was discharged home in third day after surgery.

Discussion

Etiology

Branchial cleft fistulas and cysts are in generally benign anomalities which can occure in any age. Usually presents as a different size lump or small hole on the skin of the head or neck. Typically with worsening during the respiratory tract infections and after treatment it gets better. Most common is the second branchial cleft cyst with presention of along the anterior margin of sternocleidomastoid muscle (SCM). The next one is the is first branchial cleft fistula or cyst located typically in triangle between auricula, mentum and hyoid bone. Most common localization is mandibular arch. First branchial cleft aberrations occur in 5-25% of all branchial cleft anomalies (1-3).

Classification of first branchial anomalies

The first branchial cleft anomalies are classified by Work's Classification on two types:

- Type I:
  - Ectodermal
  - Created as a result of multiplication of membranous external auditory canal
  - Histology – cyst with squamous epithelium
  - Laterally to parotid gland and facial nerve
- Type II:
  - Ectodermal and mesodermal
  - Created as a result of multiplication of membranous and cartilaginous external auditory canal
  - Histology – squamous epithelium, cartilage cells, adnexal structures
  - Medially to parotid gland and facial nerve, inferiorly to mandibular angle.

That classification was made in 1972 and looks that Work's classifications is still up to date to differentiate the types of first branchial cleft cysts and fistulas (2-4).

Diagnostic

Diagnosis of first branchial cleft fistula is usually not very problematic that bigger challenge is to diagnose first branchial cleft cyst. The fistula presents small hole on the skin and during patients examination we can see it and then refer patient to radiological examination as Ultrasound examination of neck and to be sure we can do fistulography – CT scan with contrast injected directly to fistula. To diagnose branchial cyst which is typically presented for the first time during upper respiratory tract infection as an lump on the neck that suddenly increases in size, transformed in some cases in abscess. After drainage with local improvement after the treatment it can reappear in the same localization and that is situation that we need to think about branchial cleft cyst and refer patient to CT or MRI. In younger patients sedation may be need to get that examination proceed (1, 2, 5, 6).

Surgical management

Definitive diagnosis of first branchial cleft cyst or fistula is after surgical excision and following histopathological examination. Surgery is highly recommended as aa effective treatment. Incision or drainage of cyst is not recommended and should be avoided due to high recurrence rate. In type II branchial cleft fistula the identification of facial nerve and superficial parotidectomy may be needed. High risk of surgical treatment complications may occur and should be discussed with patient and patients parents before surgery (1-7).

Prognosis

Facial nerve palsy can be presented after surgery as a temporary or permanent complication. Some authors report the effectiveness of Tumescent solution (0,9% NaCl, Lidocaine, Epinephrine, sodium bicarbonate) before parotidectomy to prevent bleeding and lowering risk of facial nerve damage. Others uses Methylthioninium Chloride to inject into fistula and track it during operation. Carefully performed operation can lower the risk of facial nerve palsy, recurrence rate and need for reoperation (5, 8, 9).

Conclusions

Proper diagnostic process followed by surgery is the one and only way of treatment patients with first branchial cleft cyst or fistula.

Conflict of interest
Konflikt interesów

None
Brak konfliktu interesów

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