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Skleroterapia z zastosowaniem doksycykliny w leczeniu zatoki skrzelopochodnej u dziecka

Doxycycline sclerotherapy in paediatric branchial cleft sinus

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Streszczenie

Anomalie kieszonek skrzelowych są drugą pod względem częstości występowania u dzieci zmianą w obrębie głowy i szyi (po torbieli przewodu tarczowo-językowego). Są to zmiany wrodzone wynikające z nieprawidłowego przebiegu procesu inwolucji aparatu skrzelowego. Najczęściej stosowaną metodą leczenia jest resekcja chirurgiczna, która pozwala uzyskać dobry efekt terapeutyczny. Jednak w ostatnim czasie pojawiły się także publikacje, w których badano skleroterapię jako praktyczną opcję leczenia, eliminującą konieczność interwencji chirurgicznej. W pracy przedstawiono przypadek siedmioletniej dziewczynki z nawracającym wyciekiem treści surowiczej w obrębie szyi. Objaw utrzymywał się od czterech lat. U pacjentki rozpoznano torbiel pierwszej kieszonki skrzelowej oraz wdrożono skuteczne leczenie metodą skleroterapii z zastosowaniem doksycykliny.

Słowa kluczowe: torbiel skrzelopochodna, skleroterapia, doksycyklina

Abstract

Branchial cleft anomalies are the second most common head and neck lesion in children after thyroglossal cyst. They are congenital lesions resulting from improper involution of the branchial apparatus. The most common treatment of choice is surgical excision, which gives a good outcome. Nevertheless, there has been recent literature exploring sclerosant therapy as a viable alternative to nonsurgical treatment. We present the case of a seven-year-old girl with first branchial cleft cyst who presented with recurrent serous discharge persisting for four years and was successfully treated with sclerotherapy injection with doxycycline.

Keywords: branchial cleft cyst, sclerotherapy, doxycycline

INTRODUCTION

The branchial apparatus consists of embryonic structures that give rise to multiple structures in the head and neck region. It is also known as the pharyngeal apparatus and comprises arches, grooves, and pouches. It is covered by mesenchyme at the core, externally by ectoderm, and internally by endoderm. Invagination of ectoderm into the cleft will form branchial cleft in which its anomalies account for approximately 30% of congenital neck diseases[1]. Incomplete obliteration of branchial cleft during embryogenesis can present as sinuses, a fistula, or cysts[2]. A sinus is a blind tract which opens into the skin, while a fistula leaves a communication between the skin and mucosa. A cyst occurs when those structures fail to resolve and become fused without communication with the skin or mucosa[2]. Second cleft abnormalities are the most common among the four types of branchial cleft anomalies. They account for 90% of all anomalies, followed by first cleft anomalies which account for 8% of cases. The remaining third and fourth cleft anomalies are rare[3].

Anomalies of the first branchial cleft occur due to incomplete closure of the ectodermal part of the first branchial cleft. The anomaly may start at the floor of the external auditory canal (EAC), extend downward along the seam between the mandible and hyoid arches, and terminate at the submandibular region[4]. First branchial cleft anomalies are often closely related to the superficial part of the parotid gland and are most often diagnosed as an intraparotid cyst. Clinical diagnosis of the first branchial cleft cyst located near or in the parotid gland can be challenging without surgical exploration[5]. Work classification is mainly used to assist in the clinical diagnosis of the first branchial cleft cyst. It divides the first branchial cleft anomalies into two groups based on their clinical and histological features. Type I anomalies present as a cystic mass of ectodermal origin, while type II anomalies may present as a cyst, sinus, fistula (or in any combination of those), and are of ectodermal and mesodermal origin[5]. Anatomically, type I anomalies occur medial to the concha and most typically extend to the postauricular crease. They pass anterior and deep to the ear lobule, superior to the facial nerve, parallel to the EAC, and finally end on the bony plate at the level of the mesotympanum. Type I is rare compared to type II[5].

Type II anomalies are usually described as duplication anomalies of the membranous part of the EAC and pinna. They are usually diagnosed after an episode of infection that requires incision and drainage at the level below the angle of the mandible. Superiorly, the lesion extends up to the level of the ramus of the mandible, passing either lateral or medial to the facial nerve and ending up at the inferior to the membranous part of the EAC. Neither type I nor II anomalies are associated with a pretragal cyst or sinus[5].

CASE REPORT

A healthy seven-year-old girl presented with recurrent serous discharge at her left neck since the age of three years old.

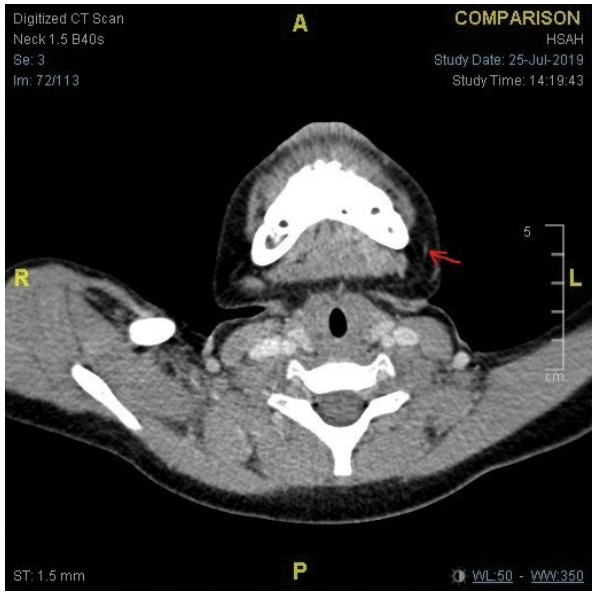

Fig. 1. Axial cut of the CT of the neck showing sinus tract marked by red arrow

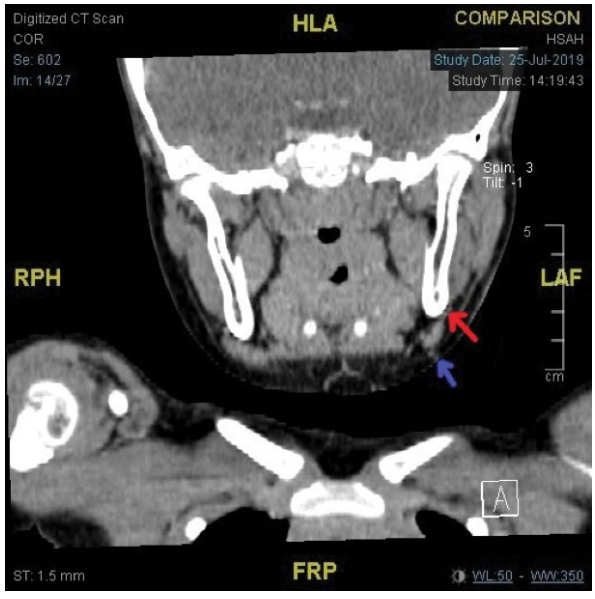

Fig. 2. Coronal cut of the CT of the neck showing the sinus tract ending blindly in the parotid gland, marked by red arrow. The external opening of the sinus at skin is marked by blue arrow

She also had multiple episodes of swelling at the similar site, which required antibiotics. However, no surgical drainage was needed. There were no episodes of recurrent parotitis or any other significant ear, throat, or nasal symptoms. Examination of the neck revealed a sinus opening at the left angle of the mandible, anterior to the sternocleidomastoid muscle, with surrounding erythematous skin. There was no tenderness on palpation, and no pus discharge upon the application of pressure. Facial nerve examination was normal. The rest of the ear, nose and throat assessment was unremarkable.

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Computed tomography (CT) of the neck was done and showed enhancement with irregularity of skin at the level of the left mandible, possibly due to abscess. It was associated with a subtle small tract going up into the subcutaneous tissue until the level of the upper mandible into the superficial parotid gland area which ended blindly into the gland (Figs. 1, 2).

The patient underwent intraoral examination under general anaesthesia, followed by left branchial cleft sinus irrigation with a sclerosant agent which, in this case, was doxycycline foam. The doxycycline foam base sclerosant was prepared for injection. Firstly, a 5 mL syringe was used for mixing 100 mg doxycycline constituted in 2.5 mL normal saline mixed with an equal volume of 25% human albumin, producing a total of 5 mL of solution (Fig. 3). Another 5 mL syringe was used to create an agitated foam base solution by mixing 1 mL gel foam with 4 mL saline water (Fig. 4). Both syringes were forcibly agitated using three-way stopcocks and mixed until particulate doxycycline foam was generated with the final content of 10 mg doxycycline per 1 mL foam. Before the injection of the sclerosant, we injected methylene blue into the Stenson's duct of the left parotid gland to ensure that there was no connection between the parotid gland and the fistula. Then, the external fistula opening was cannulated using an 18G intravenous cannula and the foam mixture was injected until it overflowed from the external opening (approximately 1.5 mL of solution) (Fig. 5). Intra-operative findings showed an external sinus opening at the left angle of the mandible, with minimal pus discharge. There was no connection between the parotid duct and the branchial cleft sinus. Rigid otoscopy of the left ear showed no sclerosant material in the external auditory canal.

The patient was discharged in a good general condition the next day after surgery. The were no immediate or long-term complications such as wound infection or recurrence. The patient was followed up until 12 months post procedure, and showed a well-healed and dry fibrotic scar at the previous external sinus opening site (Fig. 6).

DISCUSSION

Surgical excision is the most common choice of treatment for branchial cleft anomalies, ensuring good clinical resolution. It has been reported that the risk of recurrence is as low as 2.12%, while the rates of other complications, such as wound infection, range from 4.25% to 6.75%[2,6]. Recently, the emergence of treatment using sclerotherapy in branchial cleft anomalies gaining a lot of roles in managing this lesion due to its efficacy, easily performed, low morbidity and fewer complications. There are various sclerosant agents that can be used, such as trichloroacetic acid (TCA), OK-432 (picibanil), doxycycline, and ethanol[7–10]. In this case, we used doxycycline due to its nature as a gentler sclerosant, with lesser side effects, easily available, and inexpensive. TCA acts by inducing denaturalisation, precipitation, and destruction of the tissue. Thus, the fistula tract is obliterated by the destruction of the inner epithelium by TCA and then replaced by granulation tissue.

Fig. 3. Particulate doxycycline foam is created by mixing 100 mg doxycycline capsule with 2.5 mL 20% human albumin and 2.5 mL saline

Fig. 4. Preparation of agitated foam-based solution using 4 mL normal saline with 1 mL gel foam

OK-432 is a lyophilised mixture of a low-virulence strain of *Streptococcus pyogenes* incubated with benzylpenicillin potassium. It is mainly used as an immunotherapy agent for malignant tumours. It is gaining popularity as a sclerosant agent after it was found to successfully treat plunging ranula[11]. OK-432 acts by causing inflammatory cells to invade the cyst and stimulates the immunological cells to secrete a variety of cytokines such as interleukin-6, tumour-necrosis factors, and interferon-gamma. Through the release of cytokines into the circulation, it also increases the permeability of the endothelium; thus, it facilitates excretion from the lesion into the lymphatic vessels, resulting in the contraction of the cyst[8]. Doxycycline is a tetracycline antibiotic which is used widely to treat bacterial infections. It is the most preferred gentler type of sclerosant especially when dealing with lesions that communicate with the airway. It is hypothesised that it acts

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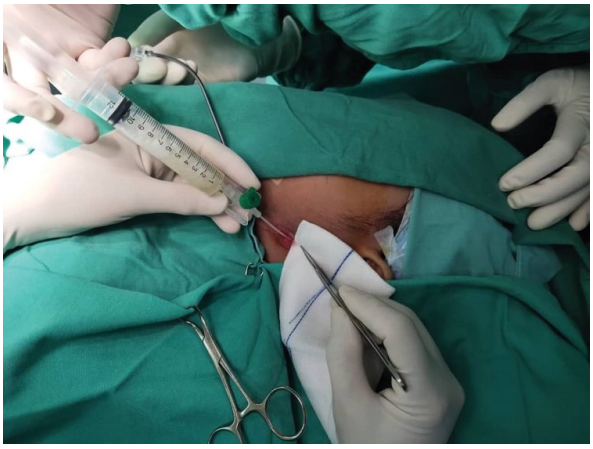

Fig. 5. External opening of the fistula was cannulated using an 18G branula and doxycycline foam mixture was injected

Fig. 6. Well-healed scar at the previous opening of the external sinus

by the destruction of mesothelial cells lining the cyst, inhibition of fibrinolysis, and induction of fibroblast growth factors. Combined with its anti-bacterial action, it causes adherence of tissue surfaces together without stimulating inflammation[12]. Complications of sclerotherapy include scarring, which was an expected outcome in this case, though from another perspective fibrosis and scarring helps in tract occlusion in this patient. We were using the existing fistulous opening to avoid another disfiguring scar in the neck region. Other complications include nerve injury, cellulitis, and skin excoriation due to leaking of the doxycycline foam solution from the catheter. No such complications were observed in this case.

Image-guided sclerotherapy has recently gained popularity because this minimally invasive procedure can be performed on an outpatient basis, eliminating the need for general anaesthesia. The procedure should be one of the options available in all centres having Intervention Radiology service. In this procedure, doxycycline foam is injected under X-ray guidance as the dilator is slowly retracted from the fistula. Patients can be discharged home after the procedure if no complications are observed[9].

CONCLUSIONS

In cases of benign lesions, such as branchial cleft fistulas, sclerotherapy treatment with doxycycline should be one of the treatments considered. It is minimally invasive, easily available, and cost effective, and it is associated with good outcomes with a lower risk of complications.

Conflict of interest

The authors report no financial or personal relationships with other individuals or organisations that could adversely affect the content of the publication and claim ownership of this publication.

Author contributions

Original concept of study: KNAR. Collection, recording and/or compilation of data: KNAR. Analysis and interpretation of data: KNAR. Writing of manuscript: KNAR. Critical review of manuscript: KBN, NG, SSMH, AZ. Final approval of manuscript: AZ.

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