Unusual Presentation of Congenital Dermoid Fistula of the Anterior Chest Region

Dear Editor

A congenital dermoid fistula of the anterior chest region (CDFACR) is a rare anomaly. It is mentioned in less than 10 papers. It consists of a skin orifice at the anterior border of the sternocleidomastoid muscle with a fistula extending caudally in the subcutaneous tissue near the sternoclavicular joint. A review of the literature contains various names and versions of this pathology (congenital fistula of the sternoclavicular joint area, congenital cutaneous fistula at the sternoclavicular joint, and congenital sternoclavicular dermoid sinus).

An 8-year-old girl presented to our hospital because she had a skin change in the projection of the right sternoclavicular joint. According to the mother's statement, skin change has been present since birth. Occasionally, leaks have occurred. Ultrasound showed a subcutaneous tubular structure that extended to the sternoclavicular joint. Operative treatment was initiated. The probe went through the small orifice to verify a 4.3 cm long fistula. It was marked [Figure 1A and B]. Skin incision was made to verify the fistula channel. The channel was resected and sent to the pathohistological analysis [Figure 1C and D]. Pathohistological analysis showed that it was keratincontaining tubular structure lined with keratinizing stratified squamous epithelium with the presence of adnexal structures. Following the pathohistological finding and review of the literature, it was concluded that

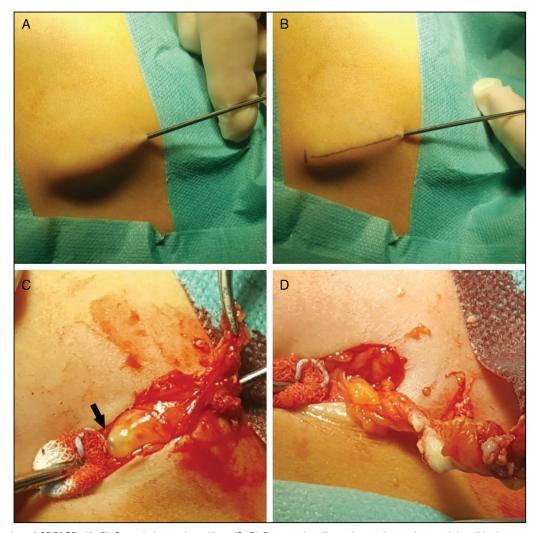


Figure 1: Resection of CDFACR. (A, B) Cannulation and marking. (C, D) Progressive dissection and complete excision (black arrow: probe tip)

it was a CDFACR. Long-term follow-up (2 years) did not show recurrence or late complications.

This anomaly has to be distinguished from other congenital anomalies and requires complete excision. The first description of this anomaly dates back to 1994 in the article of Matsunaga et al.[2] They argued that dermoid cysts were the result of the incorporation of epithelial elements in the deeper tissues along embryonic lines of fusion (inclusion type of dermoid). The embryonic sternoclavicular joint development has not been well described, and the pathogenesis of sternoclavicular dermoid sinuses and the reason for their left-sided predominance remain elusive.[3] In a series of patients in a study by Willaert et al.,[4] the leading symptoms were recurrent infections and abscesses that did not appear in our case. In the aforementioned study, fistulas were shorter (1-2cm), which was correlated with the age of patients (range 11–71 months). Instead of the catheter for cannulation, we used the probe as it is firmer. Although the articles mentioned computed tomography (CT) and magnetic resonance imaging (MRI) in the diagnosis of this anomaly, we considered the ultrasound to be optimal (given the age of the child), as confirmed by Hosokawa et al. [5] For visualization of cutaneous regions, sonography provides higher resolution images compared with CT or MRI. Unlike CT or MRI, sonographic examination of congenital anomalies does not involve radiation or require sedation, which carry small risks to infants and children. Although ultrasound is helpful in verifying the distal end of the fistula, we considered the tip of the probe must be the main guiding node in total resection. It is also interesting to note that in more than 90% of cases, this type of fistula appeared on the left,[1-5] whereas in our patient, the fistula was on the right side. Except the dominant side of the appearance of the fistula, we also witnessed that the leading reason does not have to be an infection or abscess, but aesthetics and leakage.

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Conflicts of interest

There are no conflicts of interest.

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