

Congenital Dermoid Fistula of the Anterior Chest Region

Toshiaki Numajiri, Kenichi Nishino, Maki Uenaka and Yoshihiro Sowa

Department of Plastic and Reconstructive Surgery, Kyoto Prefectural University of Medicine, Kyoto 602-8566, Japan. E-mail: prs-bin@koto.kpu-m.ac.jp
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Sir,

Congenital dermoid fistula (CDF) of the anterior chest region is a congenital cutaneous fistula that has its skin orifice at the sternoclavicular joint and a tract extending caudally in the subcutaneous tissue (1). Since CDF is rare it is sometimes incised and drained after misdiagnosis as an inflamed atheroma, but the affected region and the pathological and clinical findings are, in fact, characteristic. Only limited cases without radiological findings have been documented previously. We report here a case of CDF with computed tomography (CT) images.

CASE REPORT

A 1-year-old girl with a cutaneous pit at the left sternoclavicular joint present from birth was referred to our hospital. Physical examination revealed a 1 mm in diameter pit and subcutaneous cord-like tract that extended caudally, measuring about 4 cm in length. She had no previous history of illness. The CT images (Figs 1b, 1c) showed a narrow funicular lesion from the

neck to the anterior chest, with an area of high density at the subcutis. The lesion ended as a nodule of 25 mm in diameter with an attenuation similar to that of muscle (Fig. 1c), of which the capsule showed high density. The internal contents were almost homogeneous. The nodular lesion ruptured due to infection before the operation, with formation of an abscess in the anterior chest (Fig. 1a). Excision was performed under general anaesthesia. The abscess, funicular lesion, and skin orifice were totally excised. The fistula terminated on the fascia of the pectoralis major muscle. Histopathological analysis showed that the fistula was lined by keratinizing stratified squamous epithelium, with numerous hair follicles and sebaceous glands. The lumen was dilated in several areas by its keratin contents. Eccrine glands were also seen (Fig. 1d). Infiltration of neutrophils and foreign body giant cells was observed in the surrounding area. Thus, the diagnosis of CDF of the anterior chest region was made. The postoperative course was uneventful, although the scar was slightly hypertrophic. No clinical recurrence was evident at an 11-month follow-up.

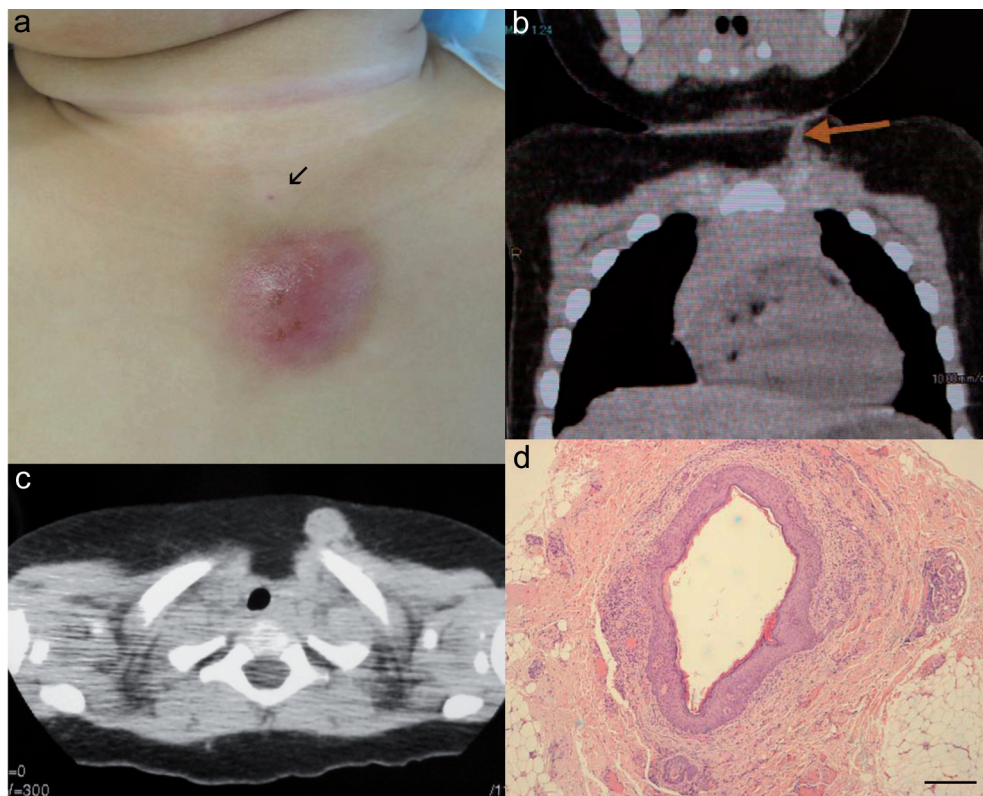


Fig. 1. (a) A 1 mm pit is seen at the left sternoclavicular joint (arrow). A reddish nodule exists caudally above the sternal bone. (b) A narrow funicular lesion is observed at subcutis from neck to chest wall (arrow). (c) Computed tomography (CT) image showing a nodule of 25 mm with attenuation similar to that of muscle. The internal contents were almost homogeneous. (d) The lumen is lined by keratinizing stratified squamous epithelium, with hair follicles and eccrine glands (haematoxylin and eosin stain; scale bar = 250 μ m).

DISCUSSION

CDF of the anterior chest region was first described by Matsunaga et al. in 1994 (1) as a subcutaneous fistula that started from a skin pit at the sternoclavicular joint and ended its tract caudally in the subcutaneous tissue near the sternal bone. Histology showed a fistula that was lined by keratinizing stratified squamous epithelium, with hair follicles, sebaceous and sweat glands. The lumen contained keratin. Muto et al. (2) also reported a fistula lined by a keratinizing squamous epithelium with numerous hair follicles and sebaceous glands, but saw no eccrine or apocrine glands. Matsunaga et al. (1) labelled the lesion as CDF of the anterior chest region, because the characteristics of the lesion histologically and clinically resembled those of a median nasal dermoid fistula (3), which occurs on the roof of the nose.

Although clinical differential diagnoses include epidermoid cyst and a branchial cleft derived fistula, such as a lateral cervical cyst, these conditions have different histological features. In our patient, in addition to finding many hair follicles, sebaceous glands, and a wall that consisted of squamous epithelium, eccrine glands were clearly observed. Miyamoto et al. (4) reported a congenital skin fistula that was similar to CDF with a sternal cleft. However, this was considered to be different from CDF because CDF are short with a blind end in the subcutaneous fatty tissue.

Vittore et al. (5) reported the CT appearance of CDF as a low-attenuation mass on the manubrium, which was well defined without septations or calcifications. The internal contents were homogeneous. In our patient, the CT images showed a narrow funicular lesion from neck to anterior chest with high density at the subcutis. This tract ended as a nodule with an attenuation similar to that of muscle, of which the capsule showed high density. Finding that the nodule shows a cystic mass with homogeneous contents in the CT images is typical.

Congenital cutaneous fistulae, including median nasal dermoid fistula, sinus of the upper lip, and sacral dermal sinus, predominantly occur near the median region. This is understandable because these fistulas occur along the

epithelial embryonic fusion line in the developmental stage at sites such as mid-ventral, mid-dorsal line, or the branchial clefts (5–7). By contrast, CDF is preferentially prevalent on the left side (1), and occurrence in the median is rare (5). There are too few reported CDF cases to determine whether this prevalence is caused by differences in the developmental stage.

As many CDFs show abscess formation with infection, surgical therapy is usually chosen. Total excision of the fistulous tract is necessary for the optimal management of this condition because recurrence is frequently reported (1, 2). Because adequate excision in the anterior chest in the Asian patient frequently causes hypertrophic scars or keloid formation (2), suturing with regard to these concerns and prevention of scarring with aftercare is required. Although CDF of the anterior chest region is uncommon, we should be familiar with this entity. CDF should be considered in the differential diagnosis of cervical and pectoral congenital fistulae of infants.

The authors declare no conflicts of interest.

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