

Congenital Dermoid Fistula of the Anterior Chest Region: A Case Report

SANJAY M KHALADKAR¹, SUJITH KUMAR SAMUDRALA²

ABSTRACT

Congenital Dermoid Fistula of the Anterior Chest Region (CDFACR) is a subcutaneous fistula that ends caudally in the subcutaneous tissue close to the sternoclavicular joint, originating from the skin at the anterior border of the sternocleidomastoid. The fistulous track is lined by keratinised stratified squamous epithelium with hair follicles, sebaceous glands and sweat glands. It presents clinically as a cutaneous pit, sinus, or infected mass. It is rare and can be complicated by recurrent infection and abscess formation. Before surgery, imaging the complete extent of the sinus is essential to prevent recurrence. Authors hereby, report a case of a 20-year-old male presenting with discharge from the anterior chest wall. On radiological and histopathological examination, it was diagnosed as a congenital dermoid fistula. In the present report, authors present a case of CDFACR confirmed through comprehensive evaluation, including sonography, Computed Tomography (CT) sinography, Magnetic Resonance Imaging (MRI) and histopathology. The present case also represents the first documented instance of CDFACR from India, highlighting the importance of multimodal imaging in the diagnosis and management of this rare congenital anomaly.

Keywords: Abscess, Congenital anomaly, Sinus tract, Sternocleidomastoid

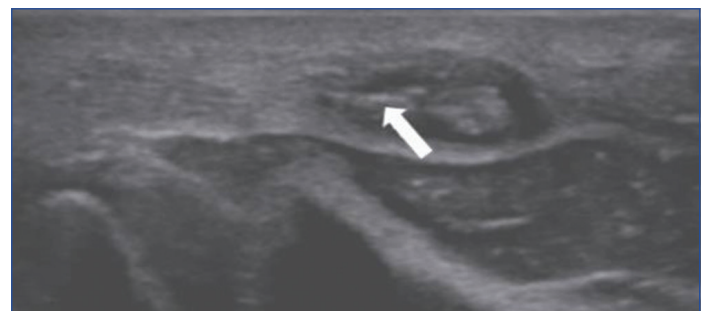
CASE REPORT

A 20-year-old male who was asymptomatic until 2.5 years ago noticed pain over the left infraclavicular region with minimal swelling and intermittent discharging pus from it [Table/Fig-1]. There was increased discharge, intermittent fever, weakness and giddiness over the last two months. Local examination revealed a pinpoint discharging sinus in the suprasternal notch on the left side, associated with erythema and tenderness. Ultrasound with a linear high-resolution probe revealed a linear hypoechoic track extending from the suprasternal notch on the left side, coursing inferiorly and slightly anteriorly, with its lower end at the medial edge of the left pectoralis major just superior to the manubrium sternum on the left side [Table/Fig-2].

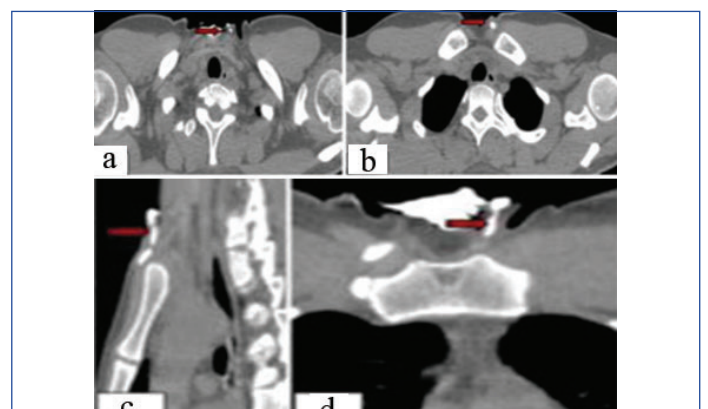


[Table/Fig-1]: Pinpoint discharging sinus in the suprasternal notch (left-side).

High-resolution Computed Tomography (HRCT) of the thorax and a plain CT scan of the thorax were done, followed by CT sinography. CT sinography was performed by injecting water-soluble contrast into the external opening in the suprasternal notch on the left side. Dilute contrast was injected through an external opening in the suprasternal notch on the left side of the midline. A linear, blind-ending sinus tract was noted, measuring approximately 23 mm in length and 3 mm in width. It was coursing inferiorly and slightly anteriorly, with its inner end at the medial edge of the left pectoralis major, just superior to the manubrium sternum on the left side [Table/Fig-3a-d]. No osteomyelitis or infective arthritis of the left sternoclavicular joint was noted. A diagnosis of CDFACR was given.



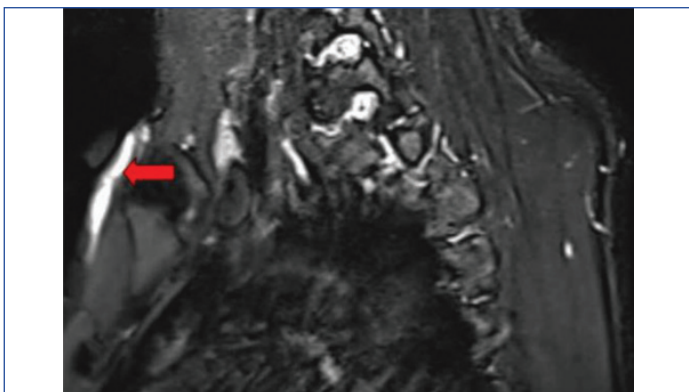
[Table/Fig-2]: High-resolution ultrasound showing linear hypoechoic sinus tract (white arrow) with central echogenic areas.



[Table/Fig-3]: CT sinography (a,b- axial; c- sagittal; d- coronal sections) showing linear sinus tract filled with contrast (arrow) extending from external skin opening, traversing through subcutaneous plane towards manubrium.

The MRI was done by taking the T1 and T2 Short Tau Inversion Recovery (STIR) sequences. The tract appeared hyperintense on T2/STIR [Table/Fig-4] and hypointense on T1WI.

A complete excision of the fistula tract was performed. Intraoperative Methylene blue was injected, a probe was inserted into the tract and dissection was done along the tract. The tract was excised in total and sent for histopathological examination. Histopathology of the excised fistulous tract: Haematoxylin and Eosin (H&E)-stained sections showed tissue lined by stratified squamous epithelium in places that formed cysts containing lamellated keratinous



[Table/Fig-4]: MRI sagittal thin T2-weighted sequence showing linear hyperintense sinus tract (arrow) extending from external skin opening, traversing through subcutaneous plane towards manubrium. Adjoining subcutaneous oedema was noted as appearing hyperintense on STIR. Underlying bones appeared normal. No osteomyelitis was noted.

flakes. Adjoining areas of mixed inflammation with prominent and congested small blood vessels and multiple hair adnexal structures and hair follicles were also seen. Findings were consistent with an infected dermoid sinus tract.

In the postoperative period, Ceftriaxone 1 gram intravenous was given twice a day for three days along with Injection Pantoprazole and Injection Tramadol. He was discharged three days after the surgery when the surgical site was healthy with no discharge. There was no soakage or discharge at the surgical site during the follow-up visit after two weeks.

DISCUSSION

The CDFACR is characterised by a skin orifice at the anterior border of the sternocleidomastoid. The fistulous track extends caudally into the subcutaneous tissue near the sternoclavicular joint [1]. Ohno M et al., suggested its origin from the remnants of the fourth branchial cleft [2]. Other terminologies used include congenital sternoclavicular dermoid sinus and congenital cutaneous

fistula at the sternoclavicular joint [1-3]. It is also called congenital sternoclavicular sinus [3]. The congenital dermoid sinus of the anterior chest region presents clinically as the cutaneous pit, sinus, or infected mass [4]. Although present at birth, it presents only if it becomes infected. If left untreated, it can become infected with abscess formation. Osteomyelitis of the underlying bone rarely occurs. Postoperative complications include hypertrophic scars and keloids. It can be misdiagnosed as a cyst with an abscess [5]. Recurrent infection and abscess formation can complicate it [6]. Imaging the complete extent of the sinus before surgery is essential to prevent a recurrence. As it is a blind-ending structure, Shin HB et al., proposed the term "sinus" rather than "fistula" [4].

The differential diagnosis includes a sinus tract, congenital skin fistula and dermoid cyst [6]. A congenital skin fistula with a sternal cleft occurs in the anterior chest wall in the midline [7]. Congenital cutaneous fistulas like the median nasal dermoid fistula, sinus of the upper lip and sacral dermal sinus occur in the median region [8]. These occur along the epithelial embryonic fusion line in the developmental stage at sites like the mid-ventral, mid-dorsal line and branchial cleft [9]. The congenital dermoid fistula predominantly occurs on the left side of the midline [10]. The congenital dermoid fistula extends caudally while the branchial fistula or sinus extends cranially [10]. A dermoid cyst is seen in the midline and lacks excretory glands [5].

To prevent recurrence, complete resection is essential. Preoperative diagnosis of this entity can be done via ultrasound, CT scan and MRI. Ultrasound provides high-resolution images. The fistulous track is seen as a hypoechoic track coursing caudally through the subcutaneous plane. Its distal end is usually blind, ending at the level of the sternoclavicular joint. No visible connection is noted with the joint or pectoralis major muscle. Ultrasound lacks radiation, does not require sedation and can visualise the caudal end of the fistula [10-12]. Both CT and MRI can detect its extent and rule out underlying bony involvement [5].

A comprehensive review of the literature on CDFACR is shown in [Table/Fig-5] [1,3-6,8,10,11,13-16].

Author	Descriptive term	N	Gender	Age	Side	Symptoms	Imaging and findings	Management	Recurrence	Complications
Matsunaga W et al., 1994 [10]	Sternoclavicular dermoid	18	12 female, 6 male	N/A	17 left, 1 right				None	None
Numajiri T et al., 2008 [8]	Congenital dermoid fistula	1	Female	1 year	Left	1 mm pit, recurrent infection and abscess	CT scan showed a nodule of 25 mm with attenuation similar to that of muscle	Surgical excision	None	Hypertrophic scar, mild
Muto J et al., 2004 [13]	Congenital dermoid fistula of the anterior chest region	1	Male	1 year	Midline	Dermal pit with recurrent swelling	Ultrasound showed a 1 cm, cord-like low-density area	Surgical excision	None	None
Adachi K et al., 2009 [6]	Congenital dermoid fistula of the chest region	1	Female	11 months	Bilateral	5 and 20 mm subcutaneous nodule with a brown fossa.	MRI revealed two well-circumscribed masses located symmetrically in the subcutaneous tissue of the sternoclavicular joint. Both lesions showed low signal intensities on T1-weighted images and high signal intensities on T2-weighted images.	Surgical excision, no relation to mediastinum	None	None
Hosokawa T et al., 2017 [14]	Congenital dermoid fistula of the anterior chest region	2	2 Female	11 month, 4years	2 left	Dermal pit with discharge	Case 1- Sonography revealed a fistula extending caudally from the pit toward the left sternoclavicular joint Case 2 Sonography showed a fistula extending caudally from the dermal pit toward the left sternoclavicular joint and MRI revealed a well-circumscribed mass in the subcutaneous tissue in the sternoclavicular joint area (T1 -low signal, T2- high signal)	Surgical excision	None	None

Willaert A et al., 2015 [1]	Sternoclavicular dermoid sinus	8	4 female, 4 male	11-71 months	All left	Small orifice at level of sternoclavicular joint, 1 accompanying skin tag, Recurrent infections, 2 abscess	In 2 patients an ultrasonography had showed tubular structure reaching sternoclavicular joint	Surgical excision	None	2 hypertrophic scars. No infection.
Kubo T et al., 2008 [15]	Congenital peristernal dermal sinuses	7	6 female, 1 male	1 month - 14 year	5 left, 2 right		Ultrasonography revealed a subcutaneous tract extending toward the pectoralis major muscle	Surgical excision	None	None
Nakamura Y et al., 2018 [16]	Peristernal dermal sinus	1	Female	17 years	Left	Punctum with scar-like lesion at caudal side	Ultrasound revealed a sinus tract of low echogenicity extending to the pectoralis major muscle	Surgical excision	None	None
Bašković M and Kljenak A 2020 [11]	Congenital dermoid fistula of the anterior chest region	1	Female	8 years	Right	Skin change with leaking since birth.	Ultrasound showed a subcutaneous tubular structure that extended to the sternoclavicular joint	Surgical excision	None	None
Shin HB et al., 2021 [4]	Congenital dermoid sinus of the anterior chest region.	11	8 female, 3 male	16-47 months	10 left, 1 right	Small congenital pit on the skin located between the lower neck and upper chest wall, with a small mass inferior to the pit.	Ultrasound was performed in 9 patients (72.7%) and demonstrated a localised hypoechoic mass-like lesion with a small tract extending from the pit to the sternoclavicular joint or pectoralis major muscle downward -Rest of the patients were diagnosed by physical examination	Surgical excision	None	None
Yang G and He T 2021 [3]	Congenital sternoclavicular sinus	88	48 female, 40 male.	1-4.5 years	73 left, 15 right	Pit of the skin over the sternoclavicular joint along the line of the anterior margin of sternomastoid muscle since birth	Ultrasound retrospectively reviewed in patients.	Surgical excision	1 of 88	None
Alenezi M 2023 [5]	Congenital dermoid sinus of the anterior chest.	1	Female	4	Right	Sinus, present since birth, intermittently discharging white matter	Ultrasound showed small superficial hypoechoic tract-like area measuring 0.1cm in diameter without deep extension	Surgical excision	None	None
Present case 2024	Congenital dermoid fistula of the anterior chest region	1	Male	20	Left	Pain over the left infraclavicular region with minimal swelling and intermittent discharging pus from it	Ultrasound (linear hypoechoic sinus tract with central echogenic areas.), CT sinography (linear sinus track filled with contrast extending from external skin opening, traversing through subcutaneous plane towards manubrium.), MRI (thin T2-weighted sequence showing linear hyperintense sinus tract)	Surgical excision	None	None

[Table/Fig-5]: Summary of findings of CDFACR from the literature [1,3-6,8,10,11,13-16].

CONCLUSION(S)

Congenital dermoid sinus of the anterior chest region is rare. It should be suspected if there is a cutaneous pit, sinus, or infected mass near the anterior border of the sternocleidomastoid. It can be misdiagnosed as an infected cyst. Due to the high incidence of recurrence and infection, the complete extent of the sinus tract should be evaluated by radiological imaging prior to surgery.

REFERENCES

- Willaert A, Bruninx L, Hens G, Hauben E, Devriendt K, Vander Poorten V. Congenital sternoclavicular dermoid sinus. *International Journal of Pediatric Otorhinolaryngology*. 2016;81:65-67.
- Ohno M, Kanamori Y, Tomonaga K, Yamashita T, Migita M, Takezoe T, et al. Congenital cutaneous fistula at the sternoclavicular joint-not a dermoid fistula but the remnant of the fourth branchial (pharyngeal) cleft?. *International Journal of Pediatric Otorhinolaryngology*. 2015;79(12):2120-23.
- Yang G, He T. The congenital sternoclavicular sinus: A single-institution retrospective study of 88 patients. *Orphanet Journal of Rare Diseases*. 2021;16:01-05.
- Shin HB, Park HS, Park EH, Jeong YJ. Congenital dermoid sinus of the anterior chest region. *Pediatric Dermatology*. 2021;38(1):132-36.
- Alenezi M. Congenital dermoid sinus of anterior chest: Case report and review of literature. *International Journal of Surgery Case Reports*. 2023;109:108591.
- Adachi K, Yoshida Y, Yamamoto O. Symmetrical congenital dermoid fistulae of the anterior chest region. *European Journal of Dermatology*. 2009;19(2):181-82.
- Miyamoto T, Hosoda Y, Fujimoto Y, Fujishima M, Sasaoka R, Mihara M. Congenital skin fistula with sternal cleft. *British Journal of Dermatology*. 1995;132(3):492-94.
- Numajiri T, Nishino K, Uenaka M, Sowa Y. Congenital dermoid fistula of the anterior chest region. *Acta Dermato-Venereologica*. 2008;88(5):538-40.
- Vittore CP, Goldberg KN, McClatchey KD, Hotaling AJ. Cystic mass at the suprasternal notch of a newborn: Congenital suprasternal dermoid cyst. *Pediatric Radiology*. 1998;28:984-86.
- Matsunaga W, Ishihara T, Yasuno K. Congenital dermoid fistula of the anterior chest region. *Nishinohon J Dermatol*. 1994;56(1):34-39.
- Bašković M, Kljenak A. Unusual presentation of congenital dermoid fistula of the anterior chest region. *Journal of Cutaneous and Aesthetic Surgery*. 2020;13(2):183-84.
- Hosokawa T, Takahashi H, Miyasaka Y, Ohira K, Tanami Y, Sato Y, et al. Ultrasound evaluation of dermal sinuses/fistulas in pediatric patients. *Journal of Ultrasound in Medicine*. 2019;38(12):3107-22.
- Muto J, Mori N, Konohana I, Sato H. Congenital dermoid fistula of the anterior chest region. *Clinical and Experimental Dermatology: Viewpoints in Dermatology*. 2004;29(1):96-97.

- [14] Hosokawa T, Yamada Y, Takahashi H, Sato Y, Tanami Y, Suzuki K, et al. Congenital dermoid fistulas of the anterior chest region (CDFACR): Usefulness of sonography for complete resection. *Radiology Case Reports*. 2017;12(3):628-31.
- [15] Kubo T, Takagi T, Yamaguchi Y, Yano K, Hosokawa K. Congenital peristernal dermal sinuses: A new entity? *British Journal of Dermatology*. 2008;159(3):763-65.
- [16] Nakamura Y, Takamuki R, Fujisawa Y, Okiyama N, Watanabe R, Ishitsuka Y, et al. Congenital peristernal dermal sinus: A case report and published work review. *The Journal of Dermatology*. 2018;45(9):e242-43.

PARTICULARS OF CONTRIBUTORS:

1. Professor and Head, Department of Radiodiagnosis, Dr. D.Y. Patil Medical College, Hospital and Research Centre, Dr. D.Y. Patil Vidyapeeth, Pune, Maharashtra, India.
2. Resident, Department of Radiodiagnosis, Dr. D.Y. Patil Medical College, Hospital and Research Centre, Dr. D.Y. Patil Vidyapeeth, Pune, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Sujith Kumar Samudrala,
Resident, Department of Radiodiagnosis, Dr. D.Y. Patil Medical College,
Hospital and Research Centre, Dr. D.Y. Patil Vidyapeeth, Sant Tukaram Nagar,
Pimpri-Chinchwad, Pune-411018, Maharashtra, India.
E-mail: docsujithsam@gmail.com

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