#### **CASE REPORT**



# Radiologically occult cervical intradural dermal sinus tract: a case report and review of literature

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### **Abstract**

We report the unusual case of a 7-month-old girl presenting with congenital cervical dermal sinus tract in which the intradural tract was not detected on preoperative imaging and was identified intraoperatively. Considering possible devastating sequelae of infection, excision of dermal sinus tract might be justified even in the case with radiologically undetected intradural tract.

**Keywords** Dermal sinus tract · Intradural tract · Cervical

## Introduction

Congenital spinal dermal sinus tract occurs with an incidence of 1 in 2500 live birth. Of those, cervical dermal sinus tract is rare and accounts for only 1% of all dermal sinus tracts [1–3]. The depth of the tracts varies. When the tract extends into the dura, the risk of infection such as meningitis, subdural abscess, or intramedullary abscess increases [4]. In such cases, surgical excision of dermal sinus tract is recommended to prevent infection even in asymptomatic patients. In cases without an intradural tract, surgical excision may not be recommended due to reduced risk of infections. However, imaging studies may not be relied on detection of an intradural tract, and surgical indication still remains to be established.

Herein, we report the unusual case of a 7-month-old girl presenting with congenital cervical dermal sinus tract in which the intradural tract was not detected on preoperative imaging and was identified intraoperatively. Surgical exploration of the intradural tract may better be performed even in radiologically undetected cases.

# **Case report**

A 7-month-old girl presented with a cutaneous epithelial defect in the midline of her upper posterior neck. Discharge from the cutaneous defect had not been observed. She had no medical history including meningitis. Physical examination showed no other abnormalities nor neurologic deficit. Magnetic resonance imaging (MRI) demonstrated the subcutaneous tract originating from skin surface to the dura at the level of C1/2, but the intradural tract was unclear (Fig. 1).

Since there was possibility that subcutaneous tract penetrated into the dura and would cause subdural infection in the future, we proceeded to the surgical excision of the tract.

Combined linear and elliptical skin incision around the cutaneous defect was made. The dermal sinus tract was followed to the level of the fascia. There was a fascial defect around the tract, and the tract entered into the fascia. The fascia was incised, and the tract was further traced deeply. The tract penetrated the dura between the lamina of C1 and C2. C1 laminectomy was performed and elliptical dural incision was made around the tract. Intradurally, the tract was found attached to the spinal cord (Fig. 2). The tract was transected slightly above the cord level.

The postoperative course was uneventful. Pathological examination demonstrated the tract comprising a lumen lined with stratified squamous epithelium. At the last follow-up, 1 year after surgery, the patient had been stable with normal development (Figure 3).



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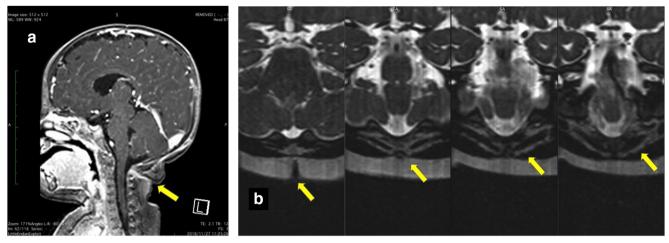


Fig. 1 Cervical magnetic resonance imaging (MRI) 1; Sagittal T1-weighted image with gadolinium [A] and T2-weighted image showed dermal sinus tract at the level of C1/2. The intradural tract was not detected

# Discussion

Congenital spinal dermal sinus tract occurs with an incidence of 1 in 2500 live birth. Of those, cervical dermal sinus tract is rare and accounts for only 1% of all dermal sinus tracts [1, 2]. The depth of the dermal sinus tracts can be various. When the tract extends into the dura, the risk of infection such as meningitis, subdural abscess, or intramedullary abscess increases, which also applies to the cervical dermal sinus tract [1, 4–8]. In such cases, surgical excision of dermal sinus tract is recommended to prevent infection even in asymptomatic patients. In cases without an intradural tract, surgical excision may not be recommended due to reduced risk of infections. However, imaging studies may not be reliable for detection of an intradural tract [9, 10]. Tisdall et al. reported that MRI underreported the presence of both an intradural tract (MRI

46%, operative finding 86%) and an intraspinal inclusion cyst (MRI 15%, operative finding 24%). In our present case, MRI could not detect the intradural tract which was identified intraoperatively, neither. Thus, surgical exploration of the intradural tract may better be performed even in radiologically undetected cases. The previous series comprised lumbar and lower thoracic dermal sinus tract [9, 10]. Our case is the first cervical dermal sinus tract in which the intradural tract was not detected by MRI and found intraoperatively.

Limited dorsal myeloschisis (LDM) has the resemblance with congenital dermal sinus tract, but there are different clinical importances between them [11, 12]. LDM is a closed skin defect and a solid tract without a lumen; thus, the possibility of infectious complications is considered low compared with congenital dermal sinus tract. Lee et al. reported several MRI features of LDM as higher visibility of the intrathecal

**Fig. 2** Intraoperative image showing a tract (arrow) penetrated the dura between the lamina of C1 and C2

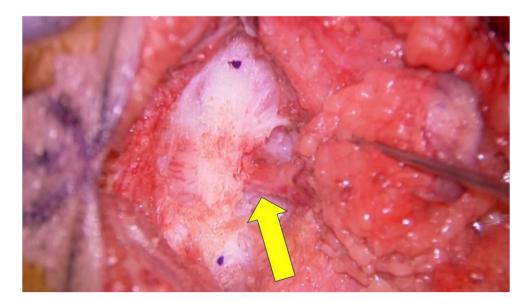
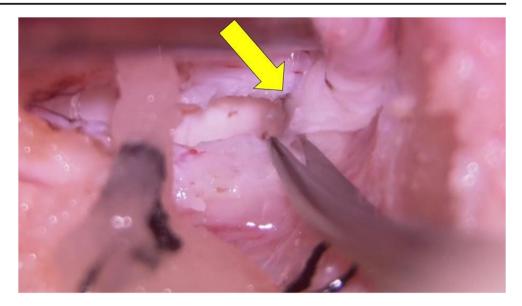




Fig. 3 Intraoperative image showing a thin tract (arrow) attached to the dorsal aspect of the spinal cord



tract, the tract attached to the cord, and dorsal tenting of the cord. In the present case, these features were not observed in MRI and congenital dermal sinus tract was more probable than LDM according to the findings of MRI, which also justified the surgical exploration.

## **Conclusion**

We report the first case of congenital cervical dermal sinus tract in which the intradural tract was not detected on preoperative imaging and was identified intraoperatively. Considering possible devastating sequelae of infection, excision of dermal sinus tract might be justified even in the case with radiologically undetected intradural tract.

## References

- Shen WC, Chiou TL, Lin TY (2000) Dermal sinus with dermoid cyst in the upper cervical spine: case note. Neuroradiology 42:51– 53. https://doi.org/10.1007/s002340050013
- Wang KC, Yang HJ, Oh CW et al (1993) Spinal congenital dermal sinus–experience of 5 cases over a period of 10 years. J Korean Med Sci 8:341–347
- Ceddia A, Di Rocco C, Pastorelli G (1990) The congenital cervical dermal sinus. A clinical case report and review of the literature. Minerva Pediatr 42:553–558
- Simon JK, Lazareff JA, Diament MJ, Kennedy WA (2003) Intramedullary abscess of the spinal cord in children: a case report and review of the literature. Pediatr Infect Dis J 22:186–192. https:// doi.org/10.1097/01.inf.0000048910.19136.49

- Dagcinar A, Konya D, Akakin A et al (2008) Congenital dermal sinus of the cervical spine in an adult. J Clin Neurosci Off J Neurosurg Soc Australas 15:73–76. https://doi.org/10.1016/j.jocn. 2006.05.022
- Mrowczynski OD, Lane JR, Shoja MM et al (2018) Double dermal sinus tracts of the cervical and thoracic regions: a case in a 3-yearold child and review of the literature. Childs Nerv Syst ChNS Off J Int Soc Pediatr Neurosurg 34:987–990. https://doi.org/10.1007/ s00381-017-3707-4
- Nicola Z, Antonio C, De Tommasi A (2014) Cervical dermal sinus complicated with intramedullary abscess in a child: case report and review of literature. Eur Spine J Off Publ Eur Spine Soc Eur Spinal Deform Soc Eur Sect Cerv Spine Res Soc 23(Suppl 2):192–196. https://doi.org/10.1007/s00586-013-2930-2
- Shah RK, Chaljub G, Swischuk LE (2003) Lower cervical dermal sinus tract and associated intraspinal abscess causing meningitis in a child. Emerg Radiol 10:160–162. https://doi.org/10.1007/s10140-003-0304-5
- Barkovich AJ, Edwards MS, Cogen PH (1991) MR evaluation of spinal dermal sinus tracts in children. AJNR Am J Neuroradiol 12: 123–129
- Tisdall MM, Hayward RD, Thompson DNP (2015) Congenital spinal dermal tract: how accurate is clinical and radiological evaluation? J Neurosurg Pediatr 15:651–656. https://doi.org/10.3171/ 2014.11.PEDS14341
- Lee SM, Cheon J-E, Choi YH et al (2017) Limited dorsal myeloschisis and congenital dermal sinus: Comparison of clinical and MR imaging features. Am J Neuroradiol 38:176–182. https:// doi.org/10.3174/ajnr.A4958
- Martínez-Lage JF, Almagro MJ, Ferri-Ñiguez B et al (2011) Spinal dermal sinus and pseudo-dermal sinus tracts: two different entities. Childs Nerv Syst ChNS Off J Int Soc Pediatr Neurosurg 27:609– 616. https://doi.org/10.1007/s00381-010-1308-6

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