



# Double dermal sinus tracts of the cervical and thoracic regions: a case in a 3-year-old child and review of the literature

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## Abstract

**Introduction** Dermal sinus tracts are rare congenital abnormalities characterized by an epithelium-lined tract that extends from the subcutaneous tissue to the underlying thecal sac or neural tube. These developmental anomalies can present asymptotically with a cutaneous dimple or with devastating complications including recurrent episodes of meningitis, or neurological complications including paralysis. Dermal sinus tracts generally occur as single lesions, and the presentation of midline double dermal sinus tracts of the cervical and thoracic regions has not been previously described.

**Methods** Here, we present the case of a 3-year-old girl suffering from recurrent episodes of myelitis, paraparesis, and intramedullary intradural masses, who was diagnosed with double dermal sinus tracts of the cervical and thoracic regions. We also present a summary of all previous reported cases of multiple dermal sinus tracts.

**Results** Our patient was successfully treated surgically and is now 2 years status post her last procedure with a significant improvement in her neurologic function and normal muscle strength and tone for her age, and there was no recurrence of her symptoms.

**Conclusions** Early treatment with prophylactic surgery should be performed when possible, but removal of these lesions once symptoms have arisen can also lead to success, as in the case presented here. Complete excision and intradural exploration is required to excise the complete tract.

**Keywords** Dermal sinus tract · Dermoid · Spinal dysraphism · Cyst

## Introduction

Dermal sinus tracts are rare defects in development characterized by an epithelium-lined tract which can extend from the subcutaneous tissue to the thecal sac and neural tube [1, 2]. These developmental anomalies occur in 1 to 2500 live births due to the dermal and neural elements of the neuroectoderm failing to separate [3, 4]. Dermal sinus tracts most commonly present as a single lesion at one of the extremes of the spinal cord, either

cervical or lumbar [5]. A case of midline double dermal sinus tracts has only been reported once previously and describes asymptomatic double dermal sinus tracts of the cervical region [6]. Herein, we describe the case of a 3-year-old girl who presented with severe meningitis and paralysis, who was found to have double dermal sinus tracts of the cervical and thoracic regions with accompanied dermoid tumors. This is the first described case of midline double dermal sinus tracts in the cervical and thoracic regions that presented with severe complications and was successfully treated. We also present a summary of all previous reported cases of multiple dermal sinus tracts.

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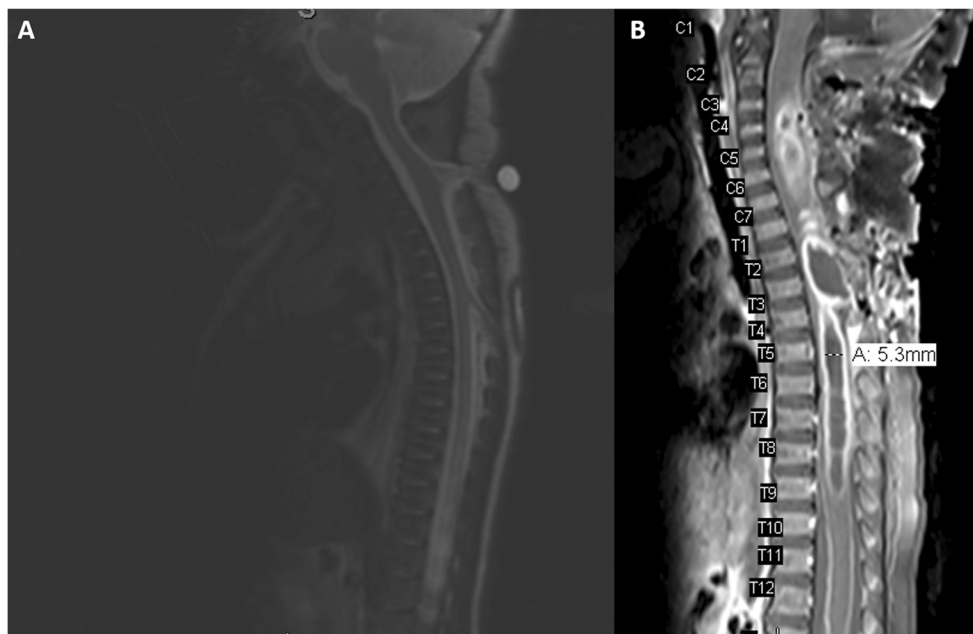
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## Case

A 3-year-old, full-term normal vaginal delivered baby girl was found to have two skin defects in the cervical and thoracic regions, shown by MRI (Fig. 1a). Surgical excision was performed at an outside institution where the cervical

**Fig. 1** **a** MRI at initial presentation demonstrating double dermal sinus tracts of the cervical and thoracic regions with accompanying skin defects. **b** MRI of the spine at the time of patient presentation at our institution when the patient had severe respiratory distress and paraplegia. MRI showed an enlarging intradural intramedullary enhancing mass with severe myelitis



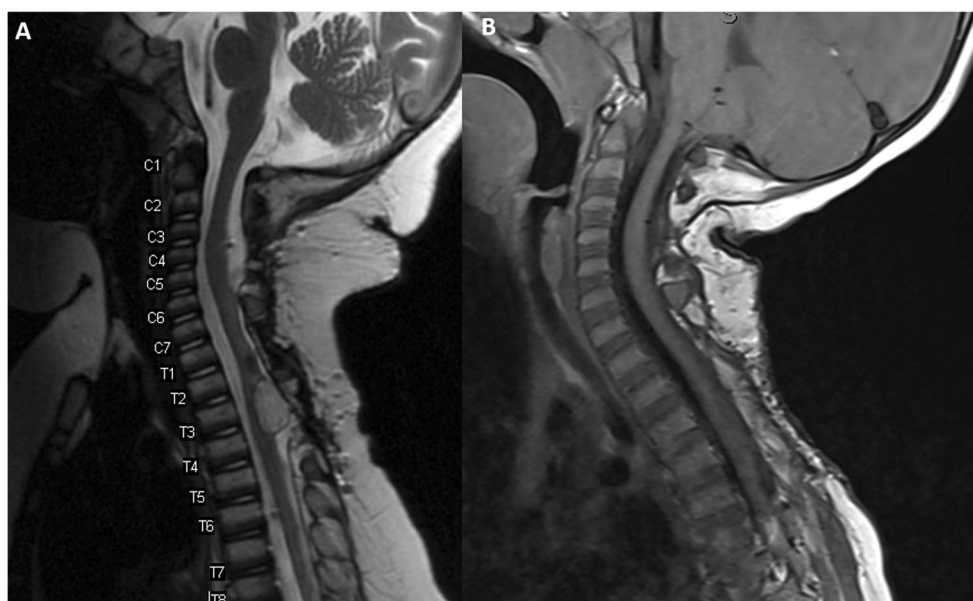
tract was excised intradurally and the thoracic tract was amputated at the level of the spine at the dura. Keratinous material with squamous epithelium was noted in the mass. She did well until she re-presented to our medical center with paraplegia and respiratory distress. MRI of the spine showed an enlarging intradural intramedullary enhancing mass with severe myelitis (Fig. 1b). The child was taken to the operating room to obtain specimen for cultures and was started on IV antibiotic and steroids. Repeat MRI showed resolution of myelitis and a residual intradural intramedullary mass at the T4-T5 level (Fig. 2a). The mass was excised and was determined to be a dermoid (Fig. 2b). There was no presence of an intramedullary spinal abscess or infected cyst. Pathology demonstrated the mass contained a portion of

connective tissue with squamous epithelium and keratinous material, which was consistent with epidermoid. At her 2-year follow-up, the patient continues to be with full strength in her upper and lower extremities appropriate for her age and normal reflexes. The patient had no episodes of hydrocephalus throughout her care. The patient has had no recurrence of her symptoms or new episodes of myelitis or paralysis.

## Discussion

Dermal sinus tracts are developmental defects which begin at the subcutaneous tissue and can extend to the underlying

**Fig. 2** **a** Repeat MRI after the patient was started on antibiotics and steroids showed resolution of myelitis and a residual intradural intramedullary mass at the T4-T5 level. **b** MRI after complete excision and intradural exploration to excise the complete tract. The mass excised was determined to be a dermoid



**Table 1** The present and previous reported cases of multiple dermal sinus tracts

Study	Age	Symptoms	Sinus tracts	Treatment	Outcome
Khashab et al. (2008) [6]	5 years old	Occasional headache	Double cervical dermal sinus tracts	No infection or intradural extension was found so no procedure was performed	Patient in same state of health
Nejat et al. (2003) [10]	16 days old	Poor feeding and low-grade fever	Double lateral retro-auricular sinus tracts	Complete excision and intradural exploration	Child was asymptomatic and was developing normally 1 year later
Ansari et al. (2009) [11]	1 month old	1 episode of bacterial meningitis with no neurological deficit	Triple dermal sinus tracts of the lumbar region	Surgical excision	Postoperative period was uneventful
Current report, 2017	3 years old	Paraplegia and respiratory distress	Double dermal sinus tracts of the cervical and thoracic regions	Complete excision and intradural exploration	Significant improvement and normal muscle strength and tone for her age

structures, including the thecal sac and neural tube [7]. These developmental anomalies frequently present with other developmental abnormalities including myelocoele, tethered cord, and dermoid tumors and come to clinical attention in diverse ways [2]. A physician may visualize a cutaneous dimple or patch of hair on the skin along the spine [8]. The sinus tract may also act as a passage of organisms and a child may thus present with a life-threatening infection such as meningitis, myelitis, or neurologic deficits including paralysis [9]. A review of all reported cases of multiple sinus tracts was performed. Multiple sinus tracts are exceedingly rare and only one case of midline double, one case of lateral retro-auricular double, and one case of triple dermal sinus tracts have been reported previously [6, 10, 11]. A table of these previous cases and the current case is shown (Table 1). Our search included MEDLINE/PubMed and Ovid with all combinations of “double,” “triple,” “multiple,” and “dermal sinus tracts” from inception to present, and the reference lists were manually searched for further relevant articles. The previous case of midline double dermal sinus tracts was both cervical and the case of triple dermal sinus tracts was all lumbar. The present case is the first with severe symptoms to present with midline double dermal sinus tracts which include the thoracic region that were successfully excised and treated with positive outcomes.

Our patient presented with episodes of myelitis, paraparesis, and a history double dermal sinus tracts with residual dermoid. The surgical removal of the associated dermoids has led to a significant recovery at this time of 2 years after her initial surgery. This case demonstrates the possibility of double dermal sinus tracts in the cervical and thoracic regions, and complete excision and intradural exploration is necessary to excise the complete sinus tract. The patient currently has normal muscle strength and tone for her

age, with normal reflexes. MRI shows complete resolution of inflammation. The patient has not had recurrence of any of her previous symptoms or episodes of hydrocephalous.

## Conclusions

We present the case of a 3-year-old child who presented with midline double dermal sinus tracts of the cervical and thoracic regions and a review of the previously reported cases of multiple dermal sinus tracts. This is to our knowledge the first known case of midline double dermal sinus tracts which included the thoracic region and presented with severe symptoms and were successfully treated. The patient had two surgeries to remove both dermal sinus tracts and associated dermoids and is now 2 years post her last operation with significant improvement and normal muscle strength and tone for her age. Although significantly less common, double dermal sinus tracts including the thoracic region are possible. Early treatment of these lesions via physician detection of cutaneous abnormalities early in child development is crucial to prevent devastating complications such as recurrent myelitis or paraparesis. Prophylactic surgery should be performed when possible, but removal of these lesions once symptoms have arisen can also lead to success, as in the case presented here. Complete excision and intradural exploration is required to excise the complete tract.

## Compliance with ethical standards

**Conflict of interest** On behalf of all authors, the corresponding author states that there is no conflict of interest.

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