

## Spinal dermal sinus coincides with an infected giant epidermoid cyst in an infant complaining of constipation: a case report

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Early diagnosis of spinal tumors is a prerequisite for achieving satisfactory neurological recovery. However, rare diseases tend to have a long diagnostic course.<sup>1</sup> In addition, the clinical presentation of spinal tumors in most children is not apparent, moreover, spinal tumors may not show clinical symptoms if they are located in the lumbar spine and sized no more than one vertebra.<sup>2,3</sup> Clinical presentations caused by tumors in the lumbar spine may include weakness of both limbs, impaired urination, and constipation. [*Paediatr Indones.* 2022;62:357-63 DOI: <https://doi.org/10.14238/pi62.4.2022.357-63> ].

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**E**pidermoid cells can present themselves aberrantly in inappropriate places, such as in the intraspinal space, and grow into benign neoplasms/tumors, either congenital or acquired. Although epidermoid tumors can be solid in consistency, “epidermoid cyst” is the most commonly used term. Most spinal epidermoid cysts (SECs) are acquired due to trauma that transfers epidermoid cells from the skin into the intraspinal space, such as surgery or lumbar puncture.<sup>4</sup> In contrast, congenital SECs originate from ectopic ectoderm remnants due to failure of complete separation between the neuroectoderm and skin ectoderm when the neural tube closes. Therefore, congenital SEC is often accompanied by other forms of spinal dysraphism, such as spinal dermal sinus, which provides a pathway for the transmission of microorganisms from the skin surface into the intraspinal space, leading to

infection.<sup>5,6</sup> Very large, infected congenital SECs very rarely affects infants.<sup>7,8</sup> This case report describes the clinical signs and symptoms, surgical intervention, and outcome of an infected giant spinal epidermoid cyst in an infant presenting initially with constipation followed by paraparesis.

### The case

A 10-month-old infant was brought to our hospital with complaints of difficulty defecating for the past three months, followed by weakness in both legs. Physical examination showed inferior paraparesis with a motor strength of approximately 3 out of 5 (with 0 corresponding to total paralysis 5 to normal strength). Sensory function was difficult to assess. Physical examination of autonomic function, i.e., a digital rectal examination was missed. Magnetic

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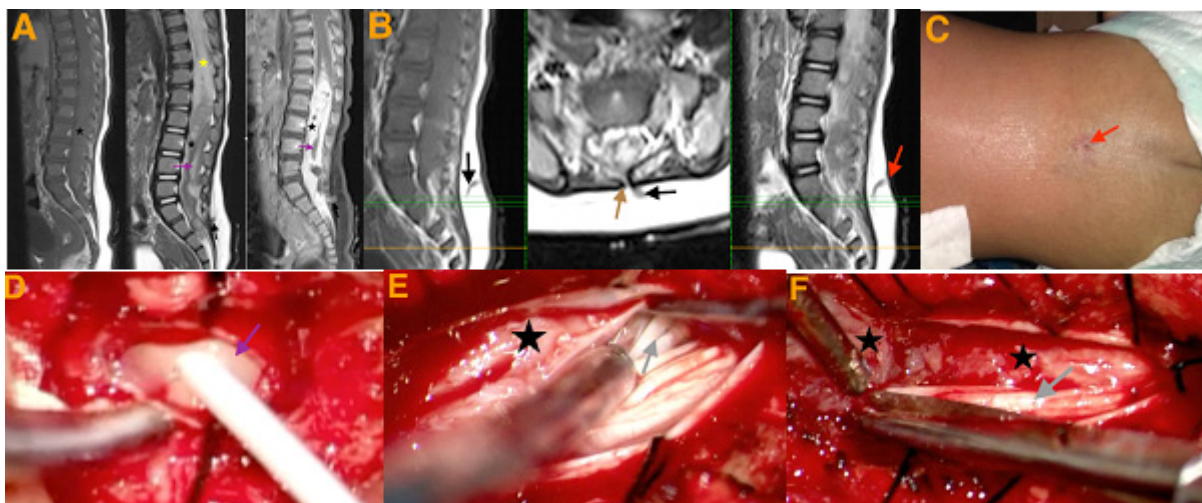
resonance imaging (MRI) of the spine showed an iso-intense tumor extending from the level of the 12th thoracic vertebra (T12) to the 2nd sacral vertebra (S2) with strong contrast enhancement throughout most of tumor (**Figure 1A**). MRI also showed spina bifida and dermal sinuses (**Figure 1B**). When the infant was on the operating table, we noticed a sinus ostium of a spinal dermal sinus visible as a dimple in the lumbosacral area, which initially was thought to be a needle puncture mark (**Figure 1C**). However, the infant had no history of lumbar puncture or spinal injection.

The infant was the first planned and expected child from the mother's second pregnancy. The mother suffered a miscarriage of her first pregnancy at three months' gestation, after 4 years of marriage. The parents were only able to conceive again three years later when the mother was 37 years old. The second pregnancy was discovered at two weeks' gestation and was routinely monitored by an obstetrician. Both mother and fetus were considered healthy and normal during pregnancy. History of illness during pregnancy and/or toxoplasmosis was denied. The infant was delivered by cesarean section on indication of breech presentation at 38 weeks' gestation and started crying immediately upon birth. Weight and length at birth was 2.6 kg and 40 cm, respectively; head circumference at birth was unknown but said to be normal.

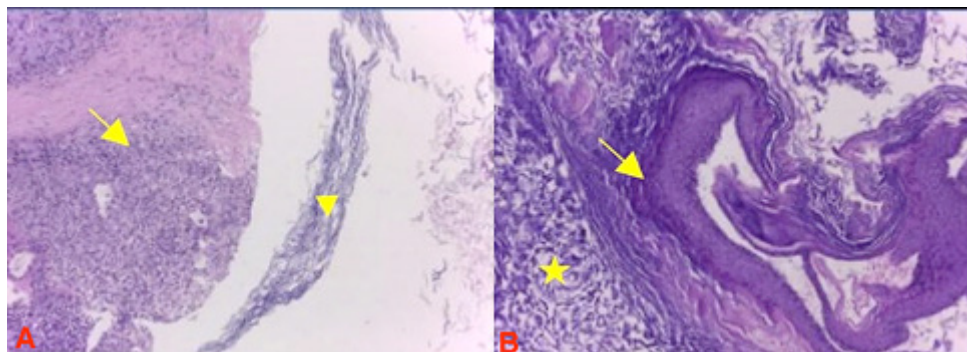
Surgery was planned in 2 stages. The first stage aimed to obtain tumor tissue for histopathological examination with L4 laminotomy. Intraoperatively, most of the tumor was found to have a solid consistency and adhere to the dura mater. A piece of tumor mass of about 0.5 cubic centimeters, which macroscopically resembled a tubercle, was taken and sent for histopathological examination. Upon incision of the tumor, pus exuded from its interior (**Figure 1D**). Subsequent tuberculosis tracing was negative. The second surgery was performed three months after the first operation with right-sided hemilaminectomy of the L2, L3, L4, L5, and S1 vertebrae to allow a gross total tumor resection. The tumor was firmly attached to the cauda equina and dura mater (**Figure 1E-F**). Most of the tumor was able to be removed, except for a few parts that were firmly attached to the nerve roots (**Figure 1 E-F**).

Histopathological findings of the specimens obtained from the first surgery showed acute and chronic inflammatory cell infiltrates and lamellar keratin (**Figure 2A**). The specimens from the second surgery showed stratified squamous epithelium with granular layer and lamellar keratin indicating an epidermoid cyst (**Figure 2B**). Based on these findings, we concluded that the tumor was an infected SEC.

The length of hospital stay for the first and second



**Figure 1.** A: a solid tumors (black stars: hypointensity on T1W and T2W with contrast enhancement) with spinal cord edema (yellow star) and pus (purple arrow: hyperintense on T2W and hypointense on T1 with contrast). B: the spinal dermal sinus/fistula (black arrows), the sinus ostium (red arrow), and spina bifida and fistula (bronze arrow). C: a small sinus ostium that does not form a dimple, the presence of a hair and a slight redness/erythema of the surrounding skin that we are not aware of at first. D: pus at the first surgery which corresponds to the MRI image on figure A (purple arrows). E: upper pole of the solid tumor (black stars) and the nerve roots / cauda equina (silver arrow). F: tumor (black stars) firmly attached to the nerve roots (silver arrow) on the previously infected area.



**Figure 2.** Histopathology of the first surgery (A) shows fibrotic tissue with acute and chronic inflammatory cells infiltrate (arrow) and lamellar keratin (arrowhead). H&E, 100X (A). Histopathology of the second surgery (B) reveals an epidermoid cyst that shows stratified squamous epithelium with a granular layer (arrow) and lamellar keratin (star). H&E, 100X (B).

surgeries was 14 and 5 days, respectively. Extended hospitalization following the first surgery was due to the need for empiric antibiotics for spinal abscesses. Prophylactic antibiotics for postoperative infection, namely intravenous cefotaxime, was replaced with intravenous meropenem for up to two weeks, followed by oral cephalexin for four weeks on an outpatient basis. The preoperative neurological deficits were restored six months after the second surgery. An MRI one month after the second surgery showed no residual tumor (**Figure 3**). Follow-up MRI one year after surgery has not been performed due to the COVID-19 pandemic. On telephone follow-up at two years after surgery, the parents reported that the child's growth and development were normal according to the child's age and there was no neurologic sequelae.

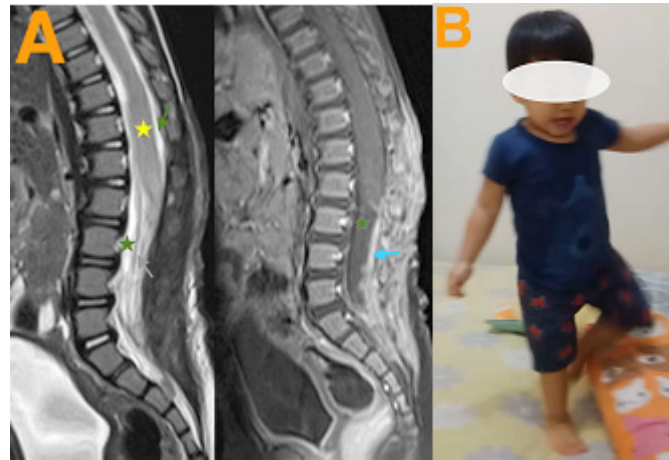
## Discussion

Spinal dermal sinuses, a form of occult spinal dysraphism, are formed due to the failure of complete separation of the skin ectoderm from the neuroectoderm, resulting in the formation of epithelial channels that connect the skin surface with nerve structures in the intraspinal space. Some skin ectodermal cells may also be trapped in the intraspinal space, these may later develop into tumors, including congenital spinal epidermoid cysts.<sup>5</sup> Spinal epidermoid cysts (SEC) can also develop from epidermal tissue inadvertently delivered into the intraspinal space during a lumbar puncture procedure, known as acquired SEC.<sup>7</sup> The shortest period from a lumbar puncture to tumor discovery is two years. Acquired SEC is considered more common

than congenital SEC.<sup>4</sup> However, the latest incidence of congenital SDS is currently unknown, as most publications are case reports.<sup>8</sup> In 1954, McIntosh et al (1954) reported an incidence of 1 in 2,500 live births.<sup>9</sup> Therefore, SDS is considered a rare disease, even more so when accompanied by epidermoid tumors.<sup>10-12</sup>

The recommended treatment for SDS is early surgical resection of the entire epithelial tract to avoid the development of devastating complications. Unfortunately, diagnosis is often delayed. Although it is a congenital disease and every newborn should undergo a thorough physical examination of the spine, the median age at which patients first visit a surgeon is 2.8 years, with a range of 1 month to 11 years.<sup>12</sup> A previous report showed that all 21 patients who underwent surgery had preoperative neurological deficits and not all patients experienced postoperative neurological improvement.<sup>13</sup> Therefore, recognizing the specific features of SDS is essential for early diagnosis. Spinal dysraphism, including SDS, is generally characterized by the presence of skin lesions in the midline along the spinal axis. The skin lesion that characterizes SDS is the sinus ostium, which resembles a dimple. Therefore, whenever a dimple or other skin lesion is found in the midline on the spinal axis on physical examination of the newborn and infant, radiological evaluation is recommended.<sup>14</sup> The best radiological evaluation or imaging modality for detecting spinal dysraphism is MRI.<sup>15</sup>

The incidence of spinal dysraphism in asymptomatic or otherwise healthy infants in the absence of a subcutaneous mass is low.<sup>16,17</sup> Radiological evaluations that are invasive or contain radiation hazards should be done selectively. The MRI is



**Figure 3.** A. MRI image one month after the second surgery shows that the subarachnoid space had opened (olive arrow) and the cerebrospinal fluid had filled the area previously occupied by the tumor (olive stars), the spinal cord edema had also reduced considerably (yellow arrow). Cyan arrows do not show tumor residue but inflamed scar tissue. B. The affected child at two years after surgery (aged 3 years) who shows normal growth and development, and the neurological deficit has recovered well.

considered an invasive examination in infants because it requires sedation. A non-invasive, widely available imaging modality is ultrasound. Previous studies have reported that spinal ultrasound has good diagnostic value in detecting spinal dysraphism and spinal cord abnormalities. Therefore, it can be used as a first-line imaging modality in detecting spinal dysraphism.<sup>18,19</sup> The MRI is done when ultrasound examination reveals abnormalities, or when atypical dimples are found. Dimples are considered atypical if they are multiple, have a cleft size of  $>5$  mm, are located  $>25$  mm from the anus, or are accompanied by other midline skin lesions.<sup>20</sup>

Neither the skin lesions on the spinal axis nor the initial clinical presentation of the complications accompanying SDS in our case was readily apparent. Indeed, sinus ostium may often go unrecognized until material leaks out of the defect.<sup>21</sup> On the other hand, the clinical presentation of spinal tumors located in the lumbar segment may also be indistinct or even asymptomatic. One of the clinical symptoms of tumors in the lumbar spine is constipation.<sup>13,22</sup> However, constipation in infants often occurs when they change their diet from liquid to solid, which does not require radiological examination.<sup>23</sup> Constipation needs to be followed up with radiological evaluation if on physical examination weak anal sphincter tone, abnormal bulbocavernosus reflex, or changes in perianal sensibility is found.<sup>24</sup>

SDS can be a pathway for germs to enter from the skin surface to the intraspinal space and cause CNS infections in the form of abscesses in the spinal cord and brain.<sup>25,26</sup> In infected cases, it is recommended to administer long-term broad-spectrum antibiotics as a complement to surgical resection of the SDS. The antibiotics of choice should cover commensal bacteria commonly found on the skin, including *Staphylococcus aureus*, *Escherichia coli*, and *Proteus mirabilis*.<sup>27</sup> Once the antibiogram has been established, antibiotics must be adjusted. The most common bacteria found is *Staphylococcus sp.*<sup>28</sup> Intravenous antibiotics are given for a period of two to six weeks, and oral antibiotics may be added for another four weeks.<sup>25,29</sup> One of the antibiotics that have good coverage against the three most common bacteria associated with infected SDS is meropenem.<sup>30</sup> In a previous report, meropenem was given for eight weeks in a case with recurrent abscesses. With incomplete resection of an infected or complicated dermal sinus tract, even with appropriate long-term antibiotics, infection or abscess may recur.<sup>31</sup>

Giant spinal tumors generally require a wide corridor to achieve satisfactory tumor resection, i.e., gross total resection. Therefore, multi-level laminectomy is the procedure of choice. However, there is a threat of postoperative deformity and spinal instability as complications that may require further surgery.<sup>32</sup> On the other hand, narrower corridors may complicate surgical maneuvers when there is massive bleeding



from the tumor, or when the tumor is firmly attached to or invades neural structures, and is gigantic in size. Recent evidence suggests that unilateral laminectomy is effective in achieving complete resection of spinal tumors along with reducing the threat of postoperative deformity and spinal instability.<sup>33,34</sup> Re-surgery may be required in the presence of a residual tumor or tumor regrowth, and in the case of postoperative spinal instability. The present case has met the definition of a giant tumor.<sup>35</sup>

After completing the treatment, monitoring for recurrence of infection and/or regrowth of epidermoid cysts should be of concern. Recurrence of infection may occur within months of discontinuing long-term antibiotics, and regrowth of epidermoid cysts can take months to years to produce clinical symptoms.<sup>31,36</sup> When referring to the acquired spinal epidermoid cysts, the period time from delivery of epidermal cells by lumbar puncture or spinal anesthesia to symptoms is between two and 20 years.<sup>7</sup> Therefore, long-term postoperative monitoring of spinal epidermoid cyst patients is recommended, considering that these types of tumors are generally attached to the nerve roots or spinal cord and there may be a small number of tumor tissue, both visible and invisible, that are not removed during surgery. Recurrence after a gross total resection of the spinal epidermoid cyst is believed to be rare. However, repeated recurrences can cause a malignant transformation of the tumor's nature, which increases the difficulty of efforts to achieve a satisfactory treatment outcome.

In conclusion, congenital SDS may show ill-defined skin markers, leading to delayed diagnosis. Considering that congenital SDS can be accompanied by spinal tumors and can be a gateway for pathogens into the intraspinal space, becoming a port of entry for CNS infections, we need to be wary of subtle clinical signs, such as constipation in infants, as possible early neurological symptom of complicated congenital SDS.

### Declaration of patient consent

The authors certify that the parents have obtained all appropriate patient consent forms. In the form, the parents have given their consent for their images and other clinical information to be reported in the journal. The parents understand that the patient names and initials will not be published, and due efforts will be

made to conceal their identity, but anonymity cannot be guaranteed.

## Conflict of interest

None declared.

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