CASE REPORT - SPINE



Intramedullary holocord abscess secondary to infected dermoid cyst

Dorota Tassigny¹ · Edward Fomekong¹ · Guus Koerts¹ · Christian Raftopoulos¹

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Abstract

In the literature, less than ten cases of holocord intramedullary abscess in children have been described. A 15-month-old girl presented with flaccid paraplegia and dermal sinus in the sacral region. MRI highlighted an infected lumbar dermoid cyst. The child underwent surgery to remove the cyst and purulent collection. Five days after surgery, she developed upper limbs paresis. An MRI showed a holocord abscess. A catheter was inserted through a cervical myelotomy into the abscess for drainage with a good postoperative recovery. A rapid management, even for extended or recurrent intramedullary abscess, can prevent potential severe neurological dysfunctions.

Keywords Abscess · Holocord · Intramedullary · Dermal sinus · Dermoid cyst · Myelotomy · Laminoplasty

Abbreviations

MRI Magnetic resonance imaging

Introduction

Intramedullary dermoid cysts account for about 0.8-1.1% of all intraspinal tumors. This congenital tumor results from inclusion during the embryonic period of cutaneous ectodermal cells during neural tube closure. In half of the cases, they are associated with a dermal sinus and could be a source of intramedullary abscess [4]. First reported in 1830, spinal intramedullary abscesses remain rare, dermal sinuses being the main cause [6]. In the literature, only about 100 cases of intramedullary abscesses have been reported, with only 40 occurring in children. Less than ten cases of intramedullary abscesses with holocord extension were described in the pediatric group [2, 5, 9–11]. We report such a case.

Case report

A 15-month-old girl presented with a 3-month history of gait disturbance and progressive bilateral lower limb weakness. Clinical examination showed the dimple of a dermal sinus in sacral region (Fig. 1) and flaccid paraplegia. The child presented no facial response or leg movement when stimuli were applied to her lower limbs. The plantar reflexes were absent. The spinal magnetic resonance imaging (MRI) confirmed a dermal sinus, as a T1-weighted hypointense tract extending from the skin to the epidural space at S1-S2 junction. T2-



Fig. 1 Presence of a small dermal sinus (arrow) in the sacral region

Christian Raftopoulos christian.raftopoulos@uclouvain.be

¹ Department of Neurosurgery, Cliniques Universitaires Saint Luc, Avenue Hippocrate, 10, 1200 Brussels, Belgium

Fig. 2 a Spinal sagittal MRI (T2weighted) highlighted the dermal sinus (*arrow*) connected to an infected lumbar dermoid cyst with an abscessed cavity. **b** A syringomyelic cavity from C5 to conus medullaris was also present (*arrows*)



weighted images of the lumbar spine showed from L1 to S2 an intradural mass with heterogeneous intensity, mass effect, and perilesional cord edema (Fig. 2a). After contrast injection, an enhancing peripheral rim appeared. A syringomyelic cavity from C5 to conus medullaris was also present (Fig. 2b). The bladder was distended. A diagnosis of infected dermoid cyst with an abscess was considered.

Based on the clinical and radiological findings and after swab of pus coming out of the tract, an empirical antibiotic therapy was started before emergency surgery. A linear incision was made in the lumbosacral region with an ellipsoidal incision surrounding the sinus tract. The dermal sinus was followed into the intradural compartment and ligated. A laminectomy from L1 to S3 was performed. The lower end of conus medullaris was scarcely located. A midline myelotomy was performed to access the lumbosacral dermal cyst containing pus-like material, hair, and squamous epithelium. The dermal sinus tract and the dermoid cyst were totally removed and the upper purulent collection was drained by a puncture myelotomy at the level of L2. The dura was closed watertight. Postoperative cultures revealed *Escherichia coli*.

Five days after surgery, the child developed progressive left upper limb paresis (grade 3/5). MRI showed a holocord abscess within the syrinx cavity in association with an important cervical cord edema (Fig. 3a). A second surgery was immediately performed. A three-level (C6 to D1) cervical laminotomy was realized. The dura mater was open over less than 2 cm. Through a short midline myelotomy, the abscess was drained. Then, the smallest pediatric feeding tube (no. 6 French) was inserted through the myelotomy (Fig. 3b). It was gradually advanced into the enlarged central canal down to the lumbar region. Then the tube was slowly withdrawn with negative pressure applied to the connected syringe. This procedure was repeated cranially; 12.0 ml of purulent fluid was obtained. The spinal cord became lax after drainage. The dura was closed and the laminotomy bone flap was replaced and fixed. A few days after surgery, the child showed complete recovery of the left upper limbs motion. One month after surgery, her neurological examination showed no longer bladder sampling, sensitivity recovery below umbilical region (not in the lower limbs), and proximal movement of the lower limbs (grade 2/5). Before leaving the hospital for the rehabilitation center, a MRI with gadolinium showed complete removal of the dermoid cyst and disappearance of the holocord high-intensity lesion (Fig. 4).

Discussion

Infected dermoid cyst is a rare cause of intramedullary abscess. In half of the patients, the infection is caused by contiguous spread from an associated dermal sinus [1, 7, 8, 12]. In children with a preexisting anatomical defect, the median age of presentation is 15 months [5]. In most of the cases, the infection starts in the gray matter, extends peripherally into the white matter, and then along the fiber tracts. The average extension is usually confined to 3–6 levels. An involvement of the entire cord is exceptional [1, 2]. In our patient, such a wide extension was probably favored by a preexisting syringomyelic cavity and the first surgery.

In the literature, an early surgical management of intramedullary abscess is recommended [1, 5, 7, 9, 12]. The



Fig. 3 a Spinal sagittal MRI with gadolinium 1 week postoperatively showing a holocord abscess (*arrows*). b During a second surgery, a catheter was inserted through a small cervical myelotomy into the abscess for drainage

aim of the surgical procedure is to excise the dermal sinus tract to remove the dermoid cyst and to drain the abscess.



Fig. 4 One month after the second surgery, a spinal sagittal MRI with gadolinium showed complete removal of the dermoid cyst and disappearance of the holocord high-intensity lesion

First reported in 1969, the technique of passing a thin catheter into the medullar abscess cavity was proposed to avoid an extensive laminectomy even when the abscess involves the entire cord [3]. This procedure is recommended when the abscess is located in one cavity or with thin-walled septae [10].

For the surgical spine approach in pediatric patients, in order to avoid postoperative progressive spinal deformity, a limited laminoplasty rather than long-segment laminectomy is recommended [1, 5, 9].

The prognosis of children with extended intramedullary abscess depends mainly on the preoperative status [2]. A complete neurological recovery is achieved in only 20% of patients [12]. Improvement failure could be due to spinal cord microinfarctions. About 25% of intramedullary abscesses are known to recur [9].

Conclusions

Early recognition of dermal sinus allows their rapid management and could prevent intramedullary infection frequently responsible of potential permanent severe neurological dysfunctions. A rapid management, even of an extended or recurrent intramedullary abscess, can help the patient to recover.

Compliance with ethical standards

Consent The parents have consented to submission of this case report to the journal.

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