



Decreased MEPs during subcutaneous dissection for untethering surgery of a “true” lipomyelomeningocele: aggravated traction of the spinal cord by release of the sac from the original nest

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Abstract

Purpose It is well known that the preoperative enlarged sac in occult spinal dysraphism, in which the spinal cord is attached to its dome, can be associated with neurological deficits by aggravation of spinal cord traction. We experienced a similar phenomenon during the early stage of untethering surgery for “lipomyelomeningocele in a strict sense (LMMC).”

Clinical presentation We report a case of a 3-month-old girl with LMMC, which showed decreased motor evoked potentials (MEPs) in the lower extremities during the early stage of untethering surgery. The sac was released from the original nest by incision with separation of the overlying skin and dissection from the muscle fascia. The sac became rounder, and the spinal cord attached to the dome of the sac was further stretched. After aspiration of cerebrospinal fluid from the sac, the MEPs recovered. Postoperatively, the patient had no neurological deficits.

Conclusions Although this phenomenon rarely occurs, release of the herniated sac of LMMC from the original nest during the early stage of untethering surgery may result in neurological changes. Application of intraoperative neurophysiological monitoring helps to detect electrophysiological deterioration and can prevent neurological deficits.

Keywords Intraoperative neurophysiological monitoring · Lipomyelomeningocele (LMMC) · Untethering · Spinal cord traction

Introduction

Lipomyelomeningocele (LMMC) refers to lumbosacral lipomatous malformation in a broad sense [7, 9, 10]. However, the dural sac which contains the spinal cord and cerebrospinal fluid (CSF) should be herniated through fascial defect and attached to the subcutaneous fat tissue in the “LMMC in a

strict sense” or “true LMMC” [1, 4]. In the present article, we used the term “LMMC” in a strict sense.

It is well known that the preoperative enlarged sac of the LMMC or terminal myelocystocele (TMC), in which the distal end of the spinal cord is attached to its dome, can be associated with neurological deficits by aggravation of spinal cord traction.

Intraoperative neurophysiological monitoring (IONM) allows electrophysiological monitoring and mapping during neurosurgical procedures. In particular, IONM is especially essential in untethering surgery to identify functioning roots or cords. In addition, overtraction of the spinal cord may cause IONM abnormalities, and this feedback warns the surgeon not to overstretch the spinal cord.

We report a rare case of LMMC in which IONM abnormalities occurred during the early stage of the untethering operation by release of the sac from the original nest and the procedures associated with aggravated traction of the spinal cord. This study was approved by the institutional review board of the Seoul National University Hospital (IRB No. H-1809-094-973).

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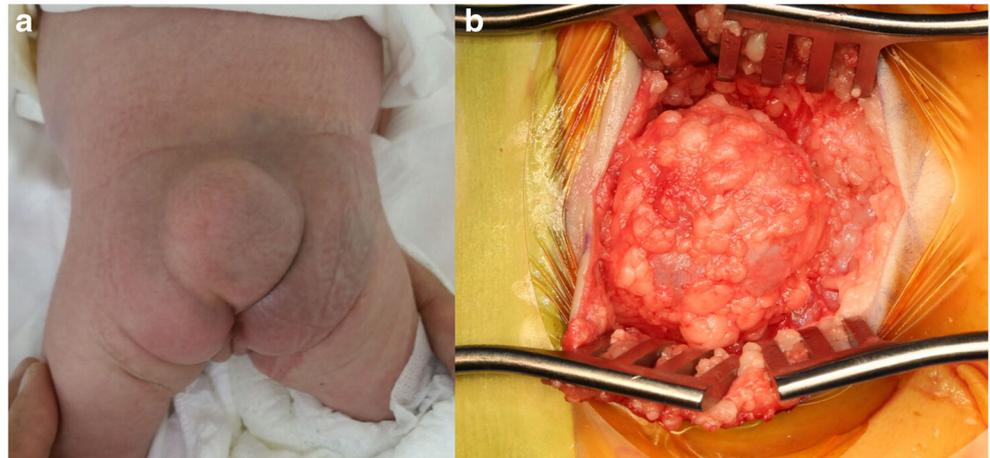
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Fig. 1 **a** A gross photo of the patient shows a cystic mass in the back. **b** An operative photo shows the cystic mass herniated through the fascial defect (right—cephalad side of the patient)

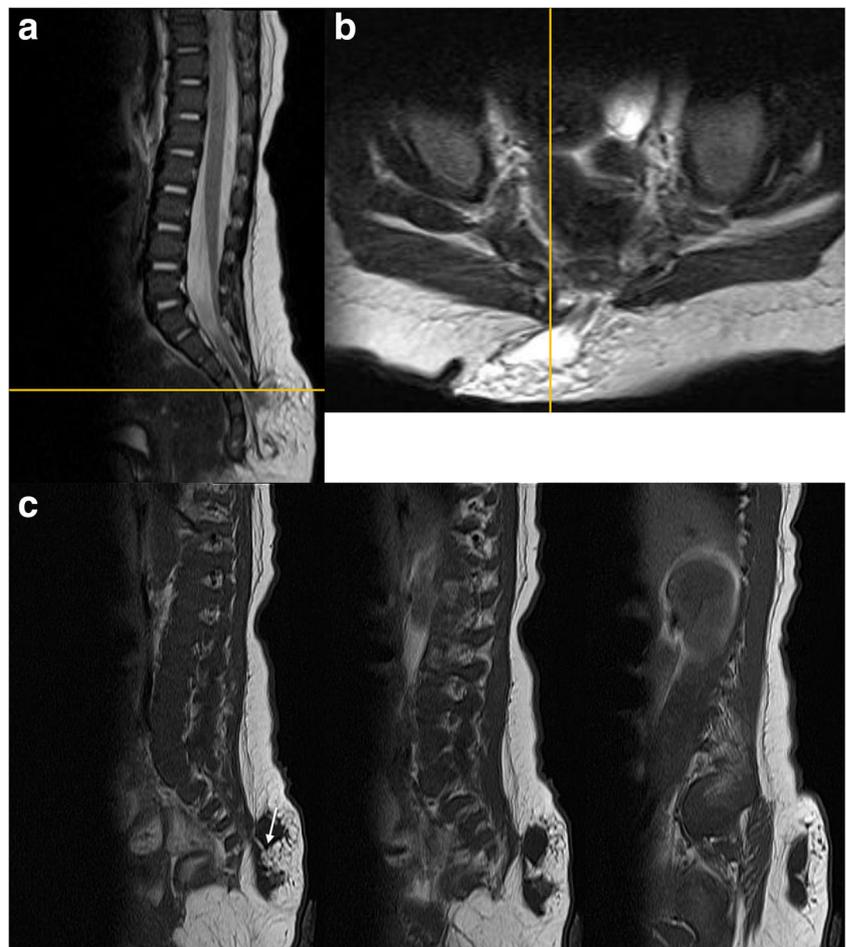


Case presentation

The female patient was one of triplets, born at 34 weeks and 2 days of gestation with a birth weight of 1800 g. A transilluminating sacral cystic mass was found at the time of birth. Postnatal ultrasonography (US) of the lumbosacral area showed posterior herniation of the meninges and spinal

cord through the sacral lamina defect. During the follow-up, the cystic lesion gradually increased in size, and we decided to perform untethering surgery (Fig. 1a). There was no motor weakness or sensory deficits upon neurological examination. Preoperative magnetic resonance imaging (MRI) of the spine revealed a herniated spinal cord attached to the cystic dome and a lipoma at the end of the cord (Fig. 2).

Fig. 2 Preoperative magnetic resonance imaging (MRI) demonstrated that the spinal cord and the dural sac are herniated through a fascial defect (**a** sagittal T2-weighted images, **b** axial T2-weighted images). Fat tissue attached to the spinal cord is also seen in sagittal T1-weighted images (**c**)



No trumpet-shaped myelocystocele was observed. Diagnosis was made with LMMC. Preoperative electromyography (EMG) and urodynamic study (UDS) showed no abnormal findings.

Surgery was performed at the age of 3 months. The patient underwent surgery in a prone neutral position and motor evoked potentials (MEPs), somatosensory evoked potentials (SSEPs), and bulbocavernosus reflex (BCR) were monitored during surgery. The baseline MEPs were recorded at the start of surgery (Fig. 3a). When the subcutaneous layer was dissected to expose the sac and to identify any fascial defects (Fig. 1b), the MEPs of both lower extremities disappeared (Fig. 3b). At that time, the MEPs of the upper extremities (abductor pollicis brevis), which were recorded as a control, were well maintained. The dural sac was not opened yet, and any structures within the dural sac and the spinal canal were untouched and undisrupted. After ceasing all manipulation for a period, MEPs were repeatedly measured. However, there were no improvements. When the surgical field was observed carefully, we found that the dural sac became rounder and looked slightly swollen compared with its size at the start of the operation.

We speculated that the decrease in MEPs was caused by a deterioration of the tethering effect, as the sac was released

from its original nest during dissection. Therefore, we tried to aspirate CSF from the sac, and after draining approximately 6 mL of CSF, the sac shrank. MEPs began to recover immediately (Fig. 3c) and were fully recovered after 20 min (Fig. 3d).

The subsequent procedure was performed. Proximal to the terminal part of the spinal cord, which is distal to where the last root arises, electrical stimulation was performed at an intensity of 10 mA to ascertain the site where the response became negative (Fig. 4a). The untethering was completed successfully by amputation at the response-negative site (Fig. 4b). MEPs and BCR remained stable until the end of surgery, and the patient recovered without neurological deficits.

Discussion

One of the questions in this case is what caused deterioration of the patient's lower extremity MEPs. The event occurred during the early stage of surgery, within 30 min from the start. At that time, only the dorsal aspect of dural sac exiting through the fascial defect was exposed. The dural sac was not opened,

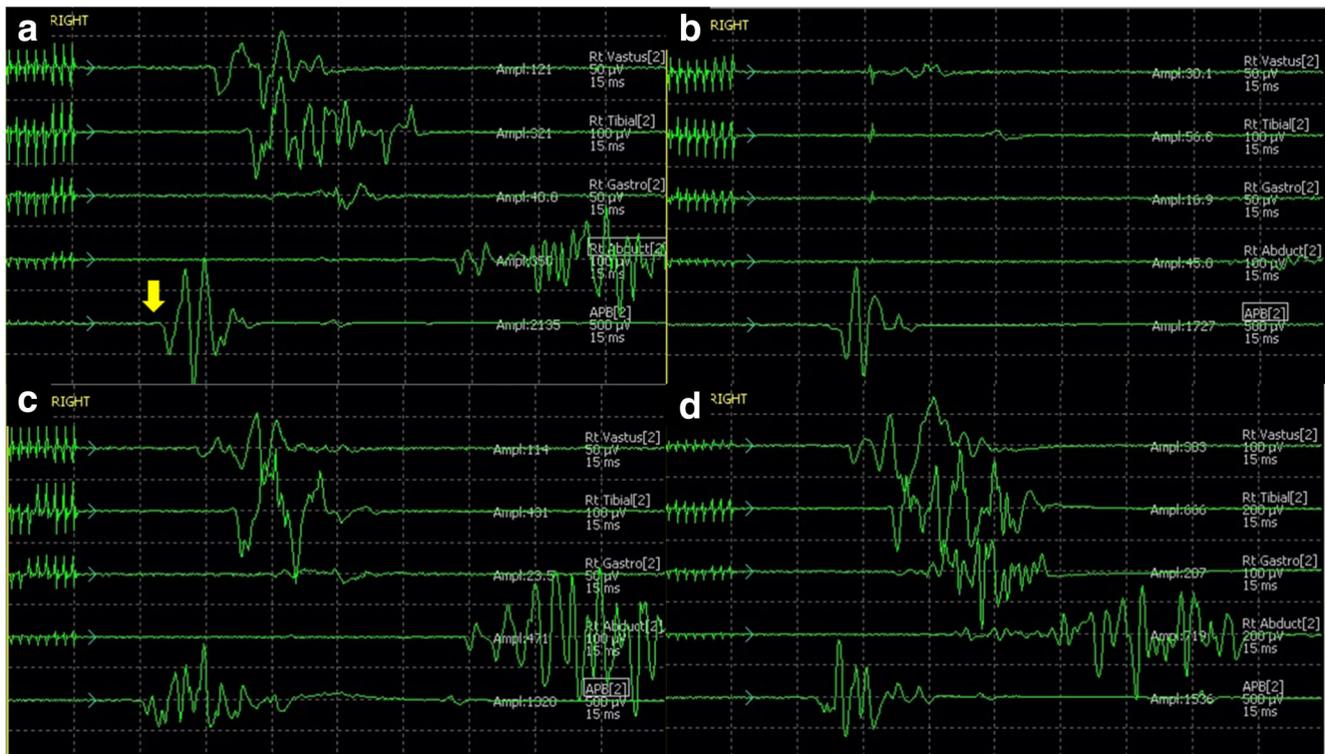
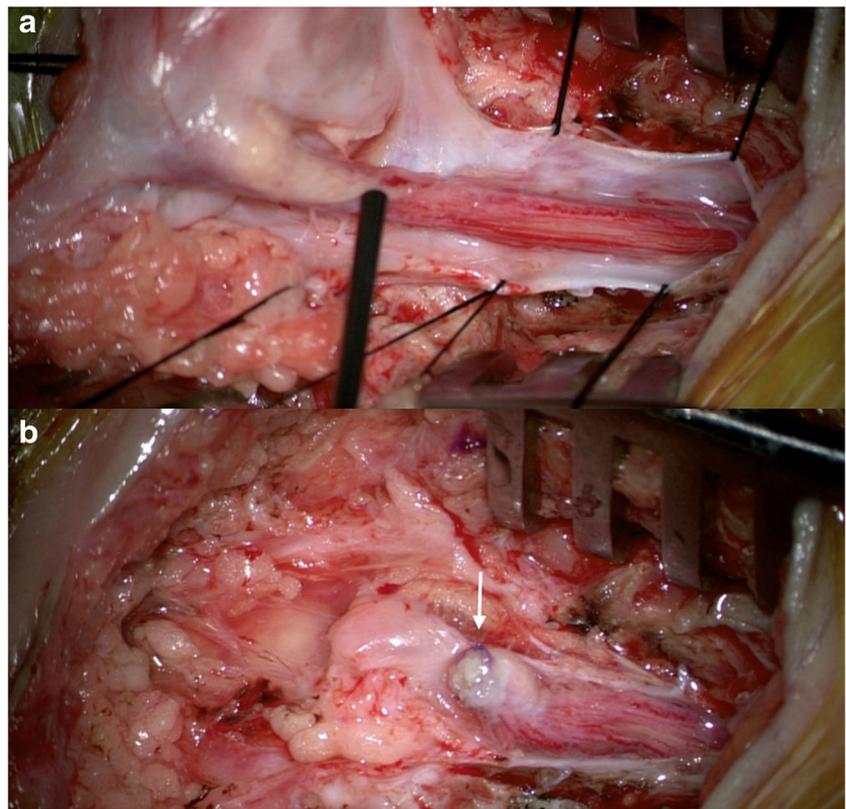


Fig. 3 Tracings of the motor evoked potentials (MEPs) demonstrate changes in MEPs during the operation. The baseline MEPs were recorded at the start of surgery (a). Decrease in MEPs in both lower extremities was noticed. Only the right-side tracings are shown in this figure (from top to bottom, vastus medialis, anterior tibialis, gastrocnemius, abductor

hallucis, abductor pollicis brevis). Please note that there is no change in the control MEPs in the upper extremity from abductor pollicis brevis (arrow) (b). After aspiration of cerebrospinal fluid, the signal appeared again (c) and was fully recovered (d)

Fig. 4 After detecting the nonfunctioning part of the spinal cord (a), untethering was performed (b)



and no manipulation of any neural structure was performed. Therefore, it is difficult to suspect direct neural damage as the cause of the deterioration. A prolonged flexed-hip prone posture, which can sometimes lead to deterioration even during untethering surgery, is also less possible considering the time of occurrence [2, 3]. We observed that the dome of the sac became elevated as it was released from the original nest, and the MEPs were recovered immediately after the aspiration of CSF. Therefore, we speculate that the spinal cord was stretched as the sac was dissected and that the stretching of the spinal cord was worsened.

Spinal dysraphism, such as TMC, is known to cause neurological deterioration when the terminal cyst enlarges [6]. Moreover, dramatic recovery has been reported to be possible if surgical intervention is performed with early detection. This case is also similar to those TMC series. The dural sac was exposed with subcutaneous dissection, and thereby, the structure supporting the sac disappeared dorsally. This process seemed to cause the sac to enlarge dorsally, leading to further stretching of the spinal cord.

IONM has been increasingly highlighted as important in the field of neurosurgery, including during untethering operations. IONM is helpful for predicting postoperative motor deficits [5, 8, 11]. In this case, with the help of meticulous IONM, electrophysiological deterioration was noted at an early point

in the event, and the cause could be defined quickly and reversibly. Furthermore, it is unique in that this rare event was fully recorded with changes in IONM.

Conclusion

Although rare, the release of the herniated sac of LMMC from the original nest during the early stage of the untethering surgery may result in neurological changes. Application of IONM facilitates the detection of electrophysiological deterioration and can prevent neurological deficits by aspiration of CSF.

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Compliance with ethical standards

Conflict of interest The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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