Original Article

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Mapping and Monitoring During Surgery for Congenital Spinal Malformation

Konjenital Spinal Malformasyon Cerrahisinde Haritalama ve Monitörizasyon

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ABSTRACT

Objective: Surgery of complex congenital spinal malformations has a risk to damage functional neural structures. Intraoperative neurophysiological monitoring for congenital spinal pathologies is suggested to reduce this risk for neural tissues and thus improve the surgical results. Our aim is to evaluate which patients are liable to reveal critical neurophysiological changes during surgery by presenting our intraoperative neurophysiological approach and the early clinical outcome of patients with congenital spinal malformations.

Methods: Nineteen patients (8 males and 11 females) were included in the study. Demographic data, symptoms and signs, radiological investigations, and other diagnostic tests for accompanying pathologies were evaluated together with neurophysiological findings.

Results: The mean age of patients was 13.6 years (range: 16 months-62 years). Neurophysiological changes were seen in 7 patients (36.8%) during surgery. Most of them had mass lesion including dermoid and epidermoid tumor or intradural abscess. Motor evoked potential (MEP) changes were seen in 3 patients without any new postoperative motor deficit. Bulbocavernosus reflex (BCR) changed in 4 patients; stimulation threshold (15-25 mA) increased in 3 of them while the other one had additional morphological changes in BCR response and worsening of preexistent urological dysfunction.

Conclusion: Mapping is the most critical part of surgery in terms of neurophysiology before cutting any structure in the operating

ÖZ

Amaç: Kompleks spinal malformasyonların cerrahisi fonksiyonel nöral yapılara hasar verme riskine sahiptir. Kompleks spinal patolojiler için kullanılan intraoperatif nörofizyolojik monitörizasyon, nöral yapılar için olan bu riski azaltmak ve dolayısıyla cerrahi sonuçları iyileştirmek için önerilmektedir. Amacımız konjenital spinal malformasyonlu hastalarda intraoperatif nörofizyolojik yaklaşımlarımızı ve erken klinik bulguları sunarak, cerrahi boyunca hangi olguların kritik nörofizyolojik değişikliklere yatkın olduğunu değerlendirmektir.

Yöntemler: On dokuz hasta (8 erkek and 11 kadın) çalışmaya dahil edildi. Demografik bilgiler, semptom ve bulgular, radyolojik değerlendirmeler ve eşlik eden patolojiler için diğer testler nörofizyolojik bulgularla birlikte değerlendirildi.

Bulgular: Hastaların ortalama yaşı 13,6 (dağılım: 16 ay-62 yıl) idi. Cerrahi süresince nörofizyolojik değişiklikler hastaların 7'sinde (%36,8) görüldü. Çoğunda dermoid, epidermoid veya intradural abseyi içeren kitle lezyonu vardı. Motor uyarılmış potansiyel (MEP) değişikliği postoperatif yeni motor defisit olmaksızın üç hastada görüldü. Bulbokavernöz refleks (BKR) 4 hastada değişti: Üçünde uyarı eşiği artarken diğerinde BKR yanıtında ek olarak morfolojik değişiklik ve postoperatif mevcut ürolojik disfonksiyonda kötüleşme oldu.

Sonuç: Postoperatif nörolojik defisiti önlemek için cerrahinin en kritik kısmı, cerrahi alandaki herhangi bir yapıyı kesmeden önce nörofizyolojik olarak haritalama yapmaktır. MEP ve BKR değişiklikleri, spinal konjenital malformasyonlar içerisinde izole

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area for preventing neurological deficit postoperatively. Among the spinal congenital malformations, MEP and BCR changes seem to occur during surgery for concomitant space occupying lesions or diastometamyelia and dermal sinus tract rather than isolated filum lipoma or tethered cord.

Keywords: Spinal congenital anomaly, dermal sinus tract, diastomatomyelia, mapping, neuromonitoring

filum lipomu veya gergin omurilikten ziyade eş zamanlı yer işgal eden lezyon, diastematomyeli veya dermal sinus traktının cerrahisinde görünmektedir.

Anahtar Sözcükler: Spinal konjenital anomali, dermal sinus traktı, diastematomyeli, haritalama, nöromonitörizasyon

Introduction

Congenital spinal malformations may present in a wide spectrum; ranging from mere junctional defects in the posterior vertebrae elements in its simplest benign form to severe neural malformations accompanied by neurological, orthopedic and urological disorders. Functional neural structures may be damaged during surgery of these serious malformations. Neurophysiological methods are routinely used for the diagnosis of subclinical findings identifying the severity of pathology or surgical decision making in the state of inconsistent clinical and magnetic resonance findings. Intraoperative neurophysiological monitoring (IONM) for congenital spinal pathologies is usually recommended to improve surgical results and reduce the risk of potential damage to the functional neural tissues (1-4). It helps the surgeon to distinguish between the roots and filum terminale, and conus medullaris in tumors even if the pathological condition has disrupted the normal anatomy of surgical region. In general, mapping and monitoring techniques including electromyography (EMG), motor evoked potential (MEP), somatosensory evoked potential (SEP) and bulbocavernosus reflex (BCR) are used during spinal surgery for congenital pathologies. In this paper, we present our neurophysiological approach and the early clinical outcome of patients who are treated for congenital spinal malformations to predict which patients are liable to reveal critical neurophysiological changes during surgery and neurological deficit postoperatively.

Methods

Patients

Nineteen patients (11 females, 8 male) having congenital spinal malformations were analyzed retrospectively. The symptoms and the physical examinations were assessed before and after surgery. All patients were evaluated with spinal magnetic resonance imaging (MRI), computerized tomography (CT) scans and other diagnostic tests including urodynamic and neurophysiological techniques for coexisting pathologies before surgery. Demographic data of the patients are presented in Table 1. Standard surgical techniques for diastometamyelia, tethered cord, dermal sinus tract (DST) and intradural tumors were applied by a single neurosurgeon (A.K). IONM data and early outcome of the patients were documented. All procedures performed in this study were in accordance with the 1964 Helsinki Declaration and its later amendments or comparable ethical standard. Informed consent was taken from all study participants.

Intraoperative Neurophysiology

IONM was performed using the Cadwell elite IONM system for monitoring by the same neurophysiologist (E.T). Mapping and monitoring techniques were used during the surgery. Mapping was used to identify nerve roots and to distinguish filum terminale while monitoring was performed to preserve the functionality of sensory and motor pathways and reflex circuits.

Neurophysiological setup included free run EMG, triggered EMG, MEP, SEP and the BCR techniques.

The MEPs were elicited by transcranial electrical stimulation of the motor cortex from C1-C2 or C3-C4 according to the 10-20 International EEG system and recording from extremities and sphincter muscles for evaluating motor system structures. In a standardized setup, constant voltage stimulation with short train technique including 5 to 8 stimuli, pulse duration of 0.5 ms, ISI of 3-4 ms, and stimulus intensity of maximum 400V were used to elicit MEP responses. At least 30 microvolt was accepted for adequate muscle MEP amplitude. Double stimulation technique was tried if the response could not be elicited by standard short train technique.

Tibial SEPs were constitutively elicited by stimulation of the posterior tibial nerve at the ankle (rectangular pulses with duration 0.3-0.5 ms, repetition rate of 4.3 Hz, intensity up to 45 mA) and recorded from scalp electrodes at Cz′-Fz or Cz'-C3'/Cz'-C4', whereas median SEP eliciting median nerve stimulation and recording from C3'-Fz/C4'-Fz was used as control except in one patient with cervical encephalocele.

The BCR was recorded from external anal sphincter (AS) muscles by electrical stimulation of dorsal nerve of the penis/clitoris with train of 4-5 stimuli, ISI of 2.5 ms, stimulus intensity of 25-80mA for stimulation by surface electrode; 5-50mA for needle electrode, duration of 0.3-0.5 ms.

Muscles to be monitored were selected based on the level of vertebra. Iliopsoas, Quadriceps femoris ve vastus lateralis arasına ,adductor longus vastus lateralis, tibialis anterior, gastrocnemius, abductor hallucis (AH), and AS muscles were monitored for thoracic and lumbar levels using bilateral upper extremity muscles as control.

Mapping was performed by classical monopolar/bipolar or concentric bipolar stimulation probe with constant current stimulation (0.05-8 mA) on the nerve roots or intradural neurological structures and recording compound muscle action potentials from related muscles. A bite block was placed to prevent tongue injuries during MEP monitoring.

Table 1. Demographic and clinical findings of the patients

Patient	Age (yrs)/ gender	Symptom	Diagnosis	Coexisting anomalies	Preoperative sensorimotor deficit	IONM	Postoperative exam
1	5/M	Skin sign	FTL + TC + dermoid tumor	Absent	Absent	Loss of MEP amplitude 70-85% in AH bilaterally	No new deficit
2	11/M	Pain	DST + intradural abscess	Bladder dysfunction	Weakness in the left leg	Loss of MEP amplitude 80-95% in AH and AS bilaterally	No new deficit
3	15/F	Pain	DST + epidermoid tumor	Absent	Absent	Loss of MEP in R AH-MEP	No new deficit
4	62/F	No	dermoid tumor + intradural abscess	Voiding and bowel dysfunction	Absent	ST↑ 20mA in R BCR, 30 mA in L and morphological change	Worsened urological dysfunction
5	5/F	Pain	FTL + TC + epidermoid tumor	Aabsent	Absent	ST↑ 15mA in BCR	No new deficit
6	8/F	Skin sign	diastometamyelia + TC	Pes cavus	Absent	ST↑ 20mA in BCR	No new deficit
7	6/F	Skin sign	diastometamyelia + TC + DST	Absent	Absent	ST↑ 25mA in BCR	No new deficit
8	6/M	Skin sign	FTL + TC+ DST	Absent	Absent	No change according to baseline	No new deficit
9	13/F	Pain	FTL +TC	Ureteral duplication, cloacal extrophy, esophageal atresia	Weakness in the left leg	No change according to baseline	No new deficit
10	4/M	Skin sign	FTL +TC	Absent	Absent	No change according to baseline	No new deficit
11	4/F	Pain	FTL +TC	cloacal extrophy	Absent	No change according to baseline	No new deficit
12	35/F	Pain	FTL +TC	Absent	Absent	No change according to baseline	No new deficit
13	21/F	Pain	FTL +TC	Absent	Absent	No change according to baseline	No new deficit
14	2/F	No	FTL +TC	Anal atresia	Absent	No change according to baseline	No new deficit
15	10/M	Pain	FTL + TC + epidermoid tumor	Absent	Weakness in the legs	No change according to baseline	No new deficit
16	3/M	Skin sign	FTL + DST	Absent	Weakness in the right leg	No change according to baseline	No new deficit
17	13/F	Pain	FTL + TC	Absent	Limited neck movement, no walking ability	No change according to baseline	No new deficit
18	21/M	Pain	Diastometamyelia + TC	Pes cavus	Weakness and hypoesthesia in the left leg	No change according to baseline	No new deficit
19	27/M	No	Cervical encephalocele + DST	Absent	Absent	No change according to baseline	No new deficit

FTL: Filum terminale lipoma, TC: Tethered cord, DST: Dermal sinus tract, MEP: Motor evoked potential, ST: Stimulus threshold, BCR: Bulbocavernous reflex, R: Right, L: Left, F: Female, M: Male

Amplitude decrease more than 50% and threshold increase more than 100V for MEP, amplitude decrease more than 50% for SEP were accepted as warning criteria. Regarding BCR, all changes (threshold increase, amplitude decrease, wave form abnormalities) were considered as a warning criterion because of our limited knowledge on its effect on surgical results.

Anesthesia

Total intravenous anesthesia consisting of propofol (1.5-2 mg/kg for anesthesia induction and 6-10 mg/kg/h for maintenance) plus remifentanil (0.15 µg/kg/min) was used in all patients except those below 5 years of age to avoid propofol infusion syndrome. Inhalation anesthesia (sevoflurane, MAC 0.5, BIS 40-60) was used for younger patients. A short-acting muscle relaxant (rocuronium, 0.5 mg/kg) was used only before endotracheal intubation.

Statistical Analysis

The statistical package for the social sciences (SPSS) 22.0 was used for data analysis. Data were presented as percentage distributions for categorical variables and median with ranges for continuous variables. Comparisons between groups were made using Mann-Whitney U test for numerical data. Statistical significance was defined as $p<0.05$.

Results

The mean age of 19 patients (8 males and 11 females) was 13.6 years (median: 11 years, range: 16 months-62 years).

Eleven patients had filum terminale lipoma (FTL) and tethered cord (TC); four of whom had additional pathologies (epidermoid tumor in 2, dermoid tumor in one and DST in one). Other patients had FTL + DST, diastometamyelia + TC, diastometamyelia + TC + DST, dermoid tumor/DST + intradural abscess, DST + epidermoid tumor/cervical encephalocele (Table 1).

The most common complaint was pain in 12 patients, followed by voiding or bowel difficulties in 2. Dermal findings were detected in 6 patients, motor deficit in 5, sensory deficit in 1, and orthopedic deformity in 2 (Table 1).

The MRI revealed TC in 14 of all patients, diastometamyelia in 3, mass lesion in 15 (3 epidermoid, 2 dermoid, 12 lipomas, 2 abscess), DST in 6, and encephalocele in 1 patient. On spinal CT scans, 11 patients showed fusion defect on the posterior elements, 2 had scoliosis and 2 had fusion of adjacent vertebral bodies.

The MEP was recordable in all patients including those with preoperative motor deficit. Threshold values were not different significantly between age the group below 17 years and adult age group. When the patients <5 years old were evaluated as a separate group, there was a difference for threshold values between groups (Mann-Whitney U test $p<0.05$). SEPs were recordable and stabil for all patients including children except one adult. This patient had SEP recordings with severe artefacts during surgery. But it was the same according to baseline at the end of the surgery.

Regarding IONM, MEP and/or BCR changes were noted while SEP did not change during the surgery. Neurophysiological changes were seen in 7 out of 19 patients (36.8%) during surgery. Five of them had mass lesion including dermoid tumor, epidermoid tumor or intradural abscess, 2 had diastometamyelia with DST/TC.

The surgeon and anesthesiologist were immediately informed when significant changes were seen in monitoring during surgery. Changes related to anesthesia were checked. If there were any changes such as end tidal carbondioxide, minimum alveolar concentration of the inhalation anesthetic or mean arterial blood pressure, they were managed by the anesthesiologist. But if the changes were still the same, the surgeon checked his maneuvers and the placement of surgical instruments. Steroid was administered if there was no improvement in potentials with these corrections.

The MEP changes which met the warning criteria were seen in 3 patients: amplitude decreasing in 2 and loss of MEP in 1. All had free run EMG abnormalities in advance of MEP changes. No new motor weakness or worsened clinical findings were observed in the postoperative period of these patients (Table 1).

Patient 1 with FTL + TC + dermoid tumor had no motor deficit preoperatively. TC was released by cutting filum terminale concurrent with mapping and MEP check after the procedure. The left side of the cord was invaded by dermoid tumor. Gross total resection was performed. The last recording of MEP showed a decrease of 70-85% in MEP amplitude of AH muscles bilaterally.

Patient 2 had DST with intradural abscess and proximal 4/5, distal 2/5 motor deficit in the left leg and also bladder dysfunction preoperatively. At the baseline recordings of this patient, monophasic response was elicited on the right side for BCR while the left side had more turn counts. Loss of BCR response was seen immediately after a 95% decrease of MEP amplitude in left AH and loss of MEP in AS muscles bilaterally while abscess formation was evacuated during the surgery. Afterwards, left AH response was lost with a decrease of MEP amplitude in right AH muscle. MEP response of left AS improved partially during dural closure.

Patient 3 had DST with sacral epidermoid tumor without motor deficit preoperatively. During surgery, DST was followed and resected until the dura. Some of the sacral nerve roots and rootlets were seen inside the tumor. Gross total resection was performed and the tumor capsule attached to nerve fibers was left due to loss of MEP in the right AH muscle during resection.

The BCR was recordable in all patients except 2 (pt 11 and 16). Pt 11 who was 4 years old female had FTL and TC with coexisting cloacal extrophy. This patient was one of our first patients. We could not put recording electrodes into the bulbocavernosus muscle instead of external AS due to cloacal extrophy. Pt 16 was 3 years old male having FTL and DST without preexisting voiding and bowel dysfunction. BCR could not be recorded. It was probably because of the inhalation anesthetics. We could even stimulate his motor cortex with electrical stimulation in high voltage (450 Voltage, 8 pulses with a pulse width 75ms). BCR changes were seen in 4 patients (Table 1). Stimulation threshold (ST) increased 15-25 mA in 3 of them while the rest had additional morphological changes in BCR response. Postoperatively, urinary function worsened in only one patient having ST increase and morphological change as turn count decreased. The patient was 62 -year-old female having constipation and also urinary incontinence before surgery. She had dermoid tumor with intradural abscess formation. Bladder and voiding complaints worsened slightly immediately after surgery. She needed intermittent catheterization for 10 days postoperatively. However, she was discharged without catheterization as her antibiotherapy was completed.

Discussion

Congenital spinal malformation surgery demands nerve root mapping and spinal cord monitoring for the protection of motor pathway and segmental reflexes like bulbocavenosus. There is still no established warning criterion for nerve root monitoring in terms of MEP monitoring according to guideline (5). Some researchers used 50-80% amplitude reduction criteria for nerve root monitoring in the spinal surgery (6,7) and they noticed false positive responses in their studies (6,7). Because of limited number, we took all MEP changes into consideration for this study. MEP changes were seen in only 3 patients; amplitude decrease of more than 50% in 2 and loss of amplitude in 1 who had severe preoperative motor deficit in the related muscle. No new motor weakness was observed in the postoperative period of these patients. Regarding the risk of postoperative motor weakness, MacDonald et al. (8) stated that it was difficult to predict motor deficit after surgery in patients with amplitude reduction due to overlapping of radicular innervation, limited sampling and variabilities in MEP responses. MEP loss may be reliable criterion for patients without preoperative motor deficit. However, we cannot clearly express this opinion about warning criteria when considering our results.

The MEP was recordable in all patients including those with preoperative motor deficit in this study. The difficulty of MEP recording for especially lower extremities was shown for children under 7 years of age and those with preoperative severe motor deficits (9). In fact, we have difficulty to elicit MEP for patients with severe paralysis according to our experiences. Therefore, we examine the patients preoperatively and add the best recordable muscle to the IONM set up according to level of surgery. However, we had no patients with muscle strength lower than 3/5 according to the Medical Research Council system in this study.

It is known that threshold value varies with age because of the myelinization and maturation of corticospinal tract, and interneuronal synaptic connections either at the cortex or spinal cord (10,11). Motor thresholds are reduced during the early development toward mid to late adolescence, and MEP amplitude is increased with age (10). Our threshold values were not different significantly between age groups. When the patients <5 years old were evaluated as a separate group, there was a difference for threshold values between groups. The reduction in thresholds with age could be related to the phasic and synchronous activation of spinal motor circuits because of completed myelination (12). One more reason of this difference might be probably related to usage of inhalation anesthetics as we showed in our previous work (13). In addition, we had also recordable MEP in children under 2 age in this study. But, we could record MEP with more pulses and higher voltage stimulation and if necessary double stimulation technique in this group, as another study carried out in infants (14). On the other way, SEPs were recordable and stabil for all patients including children except one adult. It may be due to the fact that we record from different montages such as Cz'-C3' and Cz'-C4' for the best result.

There are different practical methods reported for recording and stimulation parameters of BCR in the literature. Rodi and Vodusec stimulated pudendal nerve by cup electrodes with rectangular electrical stimuli of 0.5 ms duration and 40 mA intensity for eliciting BCR in adults (15). Skinner et al. (16) suggested double train technique for improving intraoperative BCR acquisition using needle electrodes to stimulate and recommended it as an effective practical method (17). A new study which used surface electrodes for stimulation recommended a biphasic 8-pulse stimulation with 2-msec intervals as the optimal stimulation paradigm (18). Same authors reported that this method had technical failure for eliciting BCR in some patients, because 8 pulses stimulation made it impossible to discriminate between BCR response and stimulation artifact in another study (19). We used surface electrodes in men and needle electrodes in women for stimulating dorsal penile/clitoral nerves using stimulus criteria recommended by Skinner and Vodusec (17). Our response elicited by needle electrodes with bilateral stimulation was more sensitive for recording and correlated with outcome as well.

With respect to interpretation of BCR, no widely accepted BCR warning criteria are ascertained for clinical outcome. Skinner and Vodusec (17) advocate the warning criterion as disappearance of the response under unchanged anesthetic conditions and preserved perfusion. Recently, Morota published an article about BCR warning criteria according to results of 164 operations (20). He notified using the cutoff value of BCR amplitude reduction at 75% for conus spinal lipoma. There is no information yet about threshold increase criterion for eliciting BCR. We observed threshold increase (15-30 mA) for BCR in 4 patients in our center while only one patient showed worsened voiding dysfunction and new fecal incontinence in the postoperative period. The patient showed stimulus threshold increase of 20 mA on the right side, 30 mA on the left with decreasing turn accounts. Congruently, Skinner and Vodusec (17) presented a patient having an obvious reduction of waveform complexity and amplitude (more than 50%) during surgery and who required intermittent catheterization after surgery. The take home message of this patient was that threshold increase with waveform abnormality in BCR response might be a potential risk for new deficits with this sensitive method.

Mapping technique and tracking free run EMG are used technically to prevent an injury of functional roots and rootlets. We performed mapping using different stimulator probes and stimulus intensities depending on the aim of surgery. Basically, stimulus parameters used in different centers show a discrepancy even if the modalities are the same (21,22). These differences are generally relevant to stimulation type (constant current or constant voltage), stimulus duration and stimulation probes (classical or concentric, monopolar or bipolar). In fact, the most important thing is the experience of each center. Regarding other stimulation probes, Sala et al. used bipolar concentric probe, 0.05-0.2 mA stimulus intensity, a single stimulus of approximately 0.2 to 0.5 ms duration for mapping the functionality of root and rootlets and 2.5 mA for hunting up functional rootlets in the filum or pathological tissue (21). Hoving et al. (22) used classic bipolar and monopolar probe with voltage current, duration of 0.2 ms for TC surgery. They resected the structure if the voltage threshold was three times over the voltage threshold of a previously stimulated nerve root in the operating field. Although we currently started to use bipolar concentric probe in our center, we used to benefit either classical bipolar or monopolar stimulating probe with single stimulus 0.2 ms duration, 0.05-8 mA stimulation in order to distinguish filum terminale and evaluate the functionality of the nerve roots. In our experience, we considered to increase the stimulus duration and intensity up to 8 mA with 0.2 ms to excite degenerated fibers. In this setting, the surgeon was able to cut the structure after mapping as planned without a new motor deficit in any of the patients.

Study Limitations

This study had some limitations. Major limitation of this study was small number of patients. Advanced statistical analysis could not be measured because of limited patients. The last limitation was that this study was a retrospective study.

Conclusion

This study showed that free run EMG, MEP and BCR changes seemed to occur during surgery for concomitant space occupying lesions or diastometamyelia and DST rather than isolated filum lipoma or TC among the spinal congenital malformations. Mapping before cutting any structure in the operating field is the most critical part of surgery to protect neural structures and thus improve postoperative neurological functions.

Ethics

Ethics Committee Approval: All procedures performed in this study were in accordance with the 1964 Helsinki Declaration and its later amendments or comparable ethical standard.

Informed Consent: Informed consent was taken from all study participants.

Peer-review: Externally peer reviewed.

Authorship Contributions

Surgical and Medical Practices: E.T., R.K., B.K., B.T., A.M.K., Concept: E.T., R.K., Design: E.T., A.M.K., Data Collection or Processing: B.K., B.T., Analysis or Interpretation: E.T., R.K., B.K., A.M.K., Literature Search: E.T., B.K., B.T., Writing: E.T., R.K., B.T.

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