İntraoperatif Nöromonitörizasyon ile Birlikte Serbestleştirme İşlemlerinin Bulguları: Okült Spinal Disrafizm ve Myelomeningosele Sekonder Gergin Omurilik Sendromu

The Results of Untethering Procedures with Intraoperative Neuromonitoring: Occult Spinal Dysraphism and Tethered Spinal Cord Secondary to Myelomeningocele

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ÖΖ

GİRİŞ ve AMAÇ: İntraoperatif nöromonitörizasyon, hem primer hem de sekonder gergin omurilik operasyonlarında, operasyonun daha güvenli gerçekleşmesi ve kompleks operasyonlarda cerraha yol göstermesi adına bir gereklilik olarak kabul edilir. Bu makalede, çocuklarda iki farklı operasyonun nöromonitörizasyon sonuçları ve postoperatif klinik değişimleri yer almaktadır: okült spinal disrafizm ve geçmiş myelomeningosel onarımına sekonder gergin omurilik.

YÖNTEM ve GEREÇLER: Yirmi dört operasyon üç modalite ile monitörize edildi: transkraniyal motor uyarılmış potansiyeller (TcMEP), sürekli elektromiyografi (EMG) ve direkt sinir stimülasyonu (DNS). Grup 1'de okült spinal disrafizme bağlı gergin omurilik operasyonu geçirmiş 14 hasta, grup 2'de geçmiş myelomeningosel onarımına sekonder gergin omurilik operasyonu geçirmiş 10 hasta bulunuyordu.

BULGULAR: Alt ekstremitelerden TcMEP yanıt elde etme oranı grup 1'de %92, grup 2'de %80 olarak bulundu. Anal sfinkterden TcMEP yanıt elde etme oranı grup 1'de %83, grup 2'de %60 olarak bulundu. Her iki grupta da operasyonlar esnasında TcMEP değişikliği görülmedi. Her iki grubun postoperatif ürodinami sonuçları 1 yıl periyodu içerisinde iyileşti (grup 1'de %78, grup 2'de %43). Hipoaktif mesaneli hastalarda TcMEP'te anal sfinkter yanıtları alınamadı. Yeni ya da kötüleşmiş postoperatif nörolojik defisit görülmedi.

TARTIŞMA ve SONUÇ: Hem asemptomatik okült spinal disrafizm hem de MMC'ye sekonder TCS'de spinal kordun serbestleştirilmesi bozulmuş ürodinami sonuçlarını iyileştirebilir. İntraoperatif nöromonitörizasyon ve direkt stimülasyon daha güvenli bir ameliyat için bilgi sağlayabilir ve özellikle sekonder serbestleştirmede cerrahi manevraları yönlendirebilir. İntraoperatif nörofizyolojik monitörizasyon, kalan motor fonksiyonları korumak adına nörolojik defisitli MMC hastalarının operasyonlarında yararlıdır. Anal sfinkter fonksiyonları ile mesane fonksiyonları arasında korelasyon olduğundan hasta hipoaktif mesaneye sahipse anal şfinkterden TcMEP yanıtı alınamayabilir.

Anahtar Kelimeler: İntraoperatif nöromonitörizasyon, gergin omurilik sendromu, myelomeningosel, motor uyarılmış potansiyeller

ABSTRACT

INTRODUCTION: Using intraoperative neuromonitoring in both primary and secondary tethered cord operations is accepted as a necessity for a safer operation and guiding surgeon in complex surgeries.

METHODS: Twenty four operations which were monitored with three modalities; transcranial motor evoked potentials (TcMEP), free-run electromyography and direct nerve stimulations. In group 1, there were 14 patients underwent tethered cord operations due to occult spinal dysraphism, in group 2 there were 10 patients underwent tethered cord operations secondary to previous myelomeningocele repair.

RESULTS: TcMEP responses of lower extremity were elicited in 92 % in group 1, 80 % in group 2. TcMEP responses of anal sphincter were elicited in 83 % in group 1, 60 % in group 2. No TcMEP change was observed during the surgeries in both group. Postoperative urodynamic results of both group were improved in 1 year period (78 % in group 1, 43 % in group 2). In patients with hypoactive bladder, we could not take anal sphincter responses in TcMEP.

DISCUSSION and CONCLUSION: Untethering of spinal cord both in asymptomatic occult spinal dysraphism and TCS secondary to MMC, can improve impaired urodynamic results. Intraoperative neuromonitoring and direct stimulation provides information for a safer surgery and guide surgical maneuvers especially in secondary untethering. Intraoperative neurophysiological monitoring is beneficial for operations of MMC patients with neurological deficits, to preserve their residual motor functions. Since anal sphincter functions are correlated with bladder functions, it is possible to get no anal sphincter TcMEP response if patient has hypoactive bladder.

Keywords: Intraoperative neuromonitoring, tethered spinal cord, myelomeningocele, motor evoked potentials

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INTRODUCTION

Intraoperative neurophysiological monitoring (IONM) is a continuously progressing area for today's neurosurgery. It is not proven that intraoperative neuromonitoring decreases postoperative neurological deficits in tethered cord surgery because it is difficult to conduct a prospective study to compare with a similar control group. On the other hand, the benefit of neuromonitoring on surgical strategy and low postoperative neurological complications in tethered cord operations was shown in many studies (1-6).

Therefore, using intraoperative neuromonitoring is accepted as a necessity for a safer surgery during tethered cord operations today. Myelomeningocele (MMC) is the most common (1per /1000 births) form of neural tube defects which requires a closure operation immediately after birth for preventing infection and trauma to open spinal cord (7). In postoperative years, many patients develop tethering of spinal cord due to previous MMC operation which is known as secondary tethered spinal cord (8). Untethering operations of secondary tethered spinal cords due to MMC are more challenging because of the scar tissue, attachments and abnormal neuroanatomy. Both in primary and secondary tethered cord operations, surgeons need to differentiate functional neural tissue from nonfunctional structures. Intraoperative transcranial motor evoked potentials (TcMEP) provide a continuous information about spinal cord integrity and direct nerve stimulation guides the manipulations of surgeon to find and protect functional nerves in surgical area. In our institution, we are performing intraoperative neuromonitoring together with mapping techniques in all type of tethered cord operations.

In this paper, we presented the results of IONM in two different type of operations in children: TCS due to occult spinal dysraphism and TCS secondary to previous myelomeningocele repair. Monitoring results and postoperative clinical changes compared between two groups of patients.

METHODS

Twenty four operations which were performed by same neurosurgeon, were monitored with three modalities; TcMEP, free-run EMG and direct nerve stimulations. We divided surgical procedures into 2 subgroup, group 1: tethered cord operations of occult spinal dysraphism, group 2: tethered cord operations of secondary tethered cord due to previous myelomeningocele repair. Group 1 includes 14 patients 9 female, 5 male with occult spinal dysraphism. The mean age was 44 months (between 1 months-6 years 5 months). Preoperative lower extremity motor functions and urodynamic studies are shown in **table** 1.

No	age	gender	Preoperative scoring of motor deficits	Preoperative urodynamic studies	Spinal Dysraphism	IONM Modalities	Postoperative 1th day motor functions	Postoperative urodynamic changes in 1 vear
1	4 m	F	Normal	OB	Lipoma+ dermal sinus	EMG	No change	Better
2	ly5m	М	Normal	OB + DSD	diastematomyelia	TcMEP+EMG	No change	Better
3	6y5m	F	Normal	OB	TCS	TcMEP+EMG	No change	Better
4	3 m	F	Normal	OB	diastematomyelia	TcMEP+EMG	No change	Better
5	3 у	F	Unilateral newly started distal weakness	OB	diastematomyelia	TcMEP+EMG	No change	Better
6	бm	F	Normal	OB	diastematomyelia	TcMEP+EMG	No change	No change
7	lm	F	Normal	OB + DSD	Dermal sinus	EMG	No change	Better
8	llm	F	Normal	OB	Diastematomyelia	TcMEP+EMG	No change	•
9	2 y	F	Normal	Normal	TCS	TcMEP+EMG	No change	•
10	бу	М	Unilateral newly started distal weakness	OB + DSD	TCS	TcMEP+EMG	No change	No change
11	4 y	М	Normal	Normal	Dermal sinus	TcMEP+EMG	No change	•
12	3 m	М	Normal	OB + DSD	TCS	TcMEP+EMG	No change	Better
13	13 m	М	Normal	OB	Diastematomyelia	TcMEP+EMG	No change	•
14	2y6m	F	Normal	•	Diastematomyelia	TcMEP+EMG	No change	•

Group 2 includes 10 patients 3 female, 7 male with tethered spinal cord secondary to myelomeningocele repair. The mean age was 19 month (between 2 months-4 years). Preoperative lower extremity motor functions and urodynamic studies are shown in **table** 2.

In 21 of 24 operations, intraoperative neurophysiological monitorings were done with Cadwell Cascade Elite System (Cascade Laboratories, WA, USA) while in last 3 operations Xltek Protektor 16 System were used. Anesthesia was maintained with total intravenous anesthesia with propofol and fentanyl infusion. Short acting muscle reluctant was used only for intubation.

needle recording electrodes were Subdermal inserted into M.quadriceps femoris (L3-L4), M.tibialis anterior (L4-S1), M.gastrocnemius (S1-S2) and external anal sphincter muscle (S2-S4). Patients were grounded from lateral hip and all impedances were kept below 2 kOhm. Free-run EMG and direct stimulations were recorded from same muscle groups. Free-run EMG were followed from first incision to the end of the operation. Surgeons were informed about any repeated and prolonged high frequency train activity in free-run EMG. Direct stimulations were done by a monopolar probe (reference electrode were placed on the paravertebral muscles) with a maximal 5 mA current intensity. Constant current was delivered with a 2.82 Hz stimulation frequency and 200 µs pulse width. Band-pass filter was set from 30 to 3000 Hz.

myelomeningocele								
No	Age	G	Preoperative scoring of motor deficits	Preoperative urodynamic studies	Lesion level	IONM Modalities	Postoperative 1th day motor functions	Postoperative urodynamic changes in 1 year
1	1 y 4 m	М	4	OB	L4-L5	TcMEP+EMG	No change	Better
2	4 m	М	3	OB + DSD	L4-L5	TcMEP+EMG	No change	Better
3	2 y 6 m	М	3	OB + DSD	L2-L3	TcMEP+EMG	Better (2)	Better
4	4 y	М	2	OB + DSD	\$1-\$2	TcMEP+EMG	No change	•
5	1 y 8 m	F	2	HB + DSD	L5-\$2	TcMEP+EMG	No change	No change
6	2 y 8 m	М	4	HB + DSD	L2 and below	TcMEP+EMG	No change	No change
7	1y7m	М	3	OB + DSD	L4 and below	TcMEP+EMG	No change	No change
8	2 y 1 m	F	2	OB + DSD	L4 and below	TcMEP+EMG	No change	•
9	2 m	М	1	OB + DSD	L4-S2	TcMEP+EMG	No change	No change
10	2 y 2 m	F	1	OB + DSD	L5-S1	TcMEP+EMG	No change	-

For TcMEP monitoring, corkscrew electrodes were placed over C3-C4 sites on the scalp according to International 10-20 System and stimulation parameters were adjusted to 5-7 pulses, 100-600 V, rectangular pulses with 50 μ s pulse width. We started with 100 V intensity and increased it until a compound muscle action potential was elicited at least 100 μ V in amplitude. Based on the reports about correlation between TcMEP loss and postoperative motor deficits, we prefer to use all or nothing criteria in our operations (9).

RESULTS

Intraoperative Changes in Neuromonitoring

TCMEP responses were followed throughout the operations. In group 1, we could not use TCMEP in 2 patients because of neuromuscular blocker usage. We took TcMEP responses from extremity muscles in 11 of 12 TcMEP monitoring (92 %), in 1 patient (3 month age) we could not elicit TcMEP at the start of the operation (8%). However, extremity response (tibial muscle CMAP) was obtained just after untethering of spinal cord. We took anal sphincter TcMEP responses from 10 of 12 TcMEP monitoring (83%). No change was observed during the surgeries in group 1 (**Table 3**).

Table 3. Intraoperative monitoring modalitiesand responses of patients in Group 1 (occultspinal dysraphism)							
Patient	Usage of	TcMEP	TcMEP	Intraoperative			
No	direct stimulation	Extremity responses	Anal sphincter	TcMEP change			
	Sumulation	responses	response	change			
1	+	Not used	Not used	Not used			
2	+	+	+	No change			
3	+	+	+	No change			
4	+	-	+	+ extremity			
				MEP response			
5	+	+	+	No change			
6	-	+	+	No change			
7	+	Not used	Not used	No change			
8	-	+	+	No change			
9	-	+	+	No change			
10	+	+	+	No change			
11	+	+	-	No change			
12	+	+	-	No change			
13	+	+	+	No change			
14	-	+	+	No change			

In group 2, TcMEP responses from lower extremity muscles were elicited in 8 of 10 patients (80%), in 2 patients (4 month and 2 month age) we could not take a response from lower extremity and also anal sphincter too. Anal sphincter TcMEP responses were elicited in 6 of 10 patients (60%). Two of those patients had lower extremity TcMEP responses while no response from anal sphincter (**Table** 4). No loss of intraoperative TcMEP response was seen in group 2 patients.

Table 4. Intraoperative monitoring modalities							
and responses of patients in Group 2 (TCS							
secondary to MMC)							

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Patient No	Usage of direct stimulation	TcMEP Extremity responses	TcMEP Anal sphincter response	Intraoperative TcMEP change
1	+	+	+	No change
2	-	-	-	No change
3	-	+	+	No change
4	+	+	+	No change
5	+	+	-	No change
6	-	+	-	No change
7	-	+	+	No change
8	+	+	+	No change
9	+	-	-	No change
10	+	+	+	No change

Surgeons were warned in one operation from group 1, according to repeated responses in freerun EMG. In 10 of 14 operations in group 1, and 6 of 10 patients in group 2, direct stimulation with monopolar probe was used. Filum was stimulated if it was planned to dissected, no response were taken with maximum 5 mA current intensity. In 2 cases, we directly stimulated unidentifiable tissue and took responses from lower extremity muscles and surgical plan was changed accordingly.

Postoperative clinical changes

The motor functional evaluations were done 24 hours after the operations by same neurosurgeon. Urodynamic studies were performed minimum 3 months - maximum 12 months after surgery. When we look at the results of postoperative motor functions, there is no worsened or new postoperative motor deficits in both group of patients. In only one case from group 2, a better motor function was observed in distal muscles in 24th hours after the operation (Table 2). When urodynamic results were evaluated in 1 year period, we found a difference between two groups. In group 1, we could obtain postoperative urodynamic results in 9 of 14 patients. Seven of 9 were better compared to preoperative results (78%). In group 2, only 7 of 10 patients got results of postoperative urodynamic studies yet. Three of 7 results were better compared to preoperative studies (43%).

DISCUSSION

previous studies, it was shown In that preoperative motor deficits correlates with lower TcMEP monitorability for spinal cord surgeries In our results, anal sphincter TcMEP (9.10).responses and extremity TcMEP responses in occult spinal dysraphism patients were obtained in 83 % and 92 % respectively. Our results of TcMEP monitorability rates are similar with Sala and friends (11). They found the monitorability rates 97 % for limb muscles and 74 % for anal sphincter in children who underwent complex spinal cord untethering. Furthermore, the youngest patient with TcMEP response was 4-month old. In our results, the youngest patient with monitorable TcMEP was 2 month-old.

In TCS patients secondary to MMC, TcMEP responses were obtained in lower rates (60 % anal sphincter, 80 % extremity muscles). The lower monitorability rate of TcMEP in TCS patients secondary to MMC could be related to the more severe preoperative motor deficits. However, one of the two patients who had no extremity TcMEP responses in group 2 (patient number 2 and 9) had normal motor functions and the other had severe weakness in lower extremity (number 2). On the other hand, we could obtain extremity TcMEP responses from patients with more severe motor deficits in group 2 (Table 2, Table 4). Accordingly, those results are not enough to make a conclusion about the correlation between extremity TcMEP and lower extremity motor deficits in TCS patients secondary to MMC. The other possible factor interfere to monitorability of TcMEP in those patients may be the young ages of children. The three patients with no TcMEP responses were at 2 (in group 1), 3 and 4 month of ages (in group 2). Immaturity of central nervous system could be another reason of unresponsiveness of motor cortex (12).

The patient in group 1 who had no extremity TcMEP response, had monitorable anal sphincter responses. There was irregular on/off responses on tibial muscle at the start of the operation. After untethering the spinal cord the response become more stable and monitorable in the end of the surgery. She had normal motor functions preoperatively, so there was no postoperative clinical confirmation of intraoperative change. The intraoperative improvement of TcMEP in tethered cord was anecdotally reported on case basis. It can be related to sudden changes in impaired oxidative metabolism in conus (11).

The correlations between anal sphincter TcMEP responses and preoperative clinical findings were also examined. In 2 patients from group 1, we could not elicit anal sphincter TcMEP response. Those patients had normal preoperative motor functions and extremity TcMEP responses were present. The corkscrew electrodes were at C3-C4 sites which induces more muscle twitch than other electrode montages (12). Since the transcranial stimulation induced a strong body twitch in those operations we could not increase the stimulation intensity to get an anal sphincter response. Therefore, the reason of absent anal sphincter TcMEP response may be due to inadequate intensity of transcranial stimulation. Since the electrode montage of C3-C4 limited us, (after the preparation of present paper) we continue TcMEP monitoring with C1-C2 electrode montages in our institution. In group 2, 4 patients had no TcMEP responses from anal sphincter. However, 2 of 4 patients had also no response from extremity muscles. The other 2 patients had hypoactive bladder in urodynamic studies preoperatively. Hypoactive bladder indicates an injury at lower motor neuron at sacral level- conus or sacral roots which also innervate external anal sphincter muscles. It can be deduced that hypoactive bladder is in correlation with absent anal sphincter TcMEP response in group 2.

Kumar and colleagues were used intraoperative mapping with direct stimulation at sacral level in patients with conus and cauda lesions including spinal dysraphism (14). He emphasized that mapping was helpful for dissection especially who have undergone surgery previously. Our results support same conclusions. In our operations, we did not dissect filum terminal without stimulation in a maximum 5 mA intensity. No muscle responses were obtained with filum stimulation and no postoperative worsened functions were found in patients. In 2 operations, we elicited muscle responses with direct stimulation of undefinable tissue in surgical area and manipulations were changed according to responses. Direct stimulation helped surgery in a critical direction in those patients.

There are few studies focused on neuromonitoring in TCS syndrome secondary to MMC. Pouratian and colleagues reported the results of guided electrophysiologically untethering of secondary tethered spinal cord 38 patients who had undergone perinatal myelomeningocele repair (4). They use free-run EMG and direct stimulation of tissues to identify functional neural elements during untethering procedures. We use free-run EMG and direct stimulation during the closure of MMC lesions routinely. The reason to not use TcMEP in MMC closure is the safety questions of transcranial electrical stimulations in newborns. In secondary TCS operations as in the present study, we also add TcMEP technique to follow the integrity of spinal cord. Multimodality may provide a wide possibility both roots and spinal of monitoring cord simultaneously.

There are studies about TCS operations with intraoperative neuromonitoring which evaluated postoperative urological functions. The percentages of postoperative improvement in bladder functions were between 14-46 %. In most of them, only urological symptoms were evaluated, patients were adults or the pathologies were mixed (lipoma, myelomeningocele, tight filum etc.) (1,6,15). In very young children it is not possible to recognize bladder dysfunction due to tethering cord. Urodynamic studies are helpful for an early recognition of TCS and prevents a delay in surgery and irreversible injury. Hoving and colleagues used video urodynamics for evaluations (1). They found stable urological functions postoperatively in high risk group of TCS patients (lipomyelomeningocele or/and split cord). However, the time of the postoperative urodynamics did not mentioned which is suggested to perform at least 3 months after operations generally. In follow-up periods they found an improvement in urologic functions in the 4 of 40 patients (10 %). Pouratian and colleagues were evaluated bladder with cystometrogram and found a 20 % urological improvement postoperatively in TCS patients secondary to MMC (4). Untethering of spinal cord with intraoperative monitoring both in

asymptomatic occult spinal dysraphism and TCS secondary to MMC, may contribute to improvement of impaired urodynamic results by helping to preserve root functions. Our postoperative urodynamic change rates are high in both group according to literature. However, this can be resulted from the small number of patients in the groups which make difficult to draw a conclusion about postoperative urodynamic changes. Further studies need with large number of patients to investigate the postoperative changes in bladder functions after TCS operations with IONM.

Conclusion

The use of intraoperative monitoring and mapping techniques together is feasible and efficient during TCS operations (2,3,4,5,6). In our results, there is no new or worsened neurological or urological functions. Untethering of spinal cord can improve urodynamic results in both occult spinal dysraphism and MMC patients. Besides, we may not elicit anal sphincter TcMEP response, if patient has hypoactive bladder. Continuous information about spinal cord integrity with TcMEP gives a reassurance to surgeon about their strategy and mapping with direct stimulation make easier to find and preserve functional tissue especially while doing maneuvers in distorted neuroanatomy. Even the most of the patients with MMC had neurological deficits, it is feasible to use TcMEP and direct stimulation in those operations. Moreover, to preserve the residual functions in lower extremities during the operations is essential for those children to maintain their life quality.

Disclosure

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Conflict of Interest

All authors stated no conflict of Interest.

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