International Journal of Medical Sciences And Clinical Research

VOLUME Vol.06 Issue03 2026
PAGE NO. 18-23
DOI 10.37547/ijmscr/Volume06Issue03-04

Clinical, Neurophysiological and Imaging Correlation in Surgery for Tethered Cord Syndrome with Closed Spinal Dysraphism In Children

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Received: 30 January 2026; **Accepted:** 25 February 2026; **Published:** 19 March 2026

Abstract: In this study, clinical changes, neurological changes before and after surgery were compared in children born with closed spinal cord dysraphism, and their correlation was determined using neurophysiological examination methods.

Keywords: Spina bifida, spinal cord, dysraphism, MRI, neurophysiology.

Introduction: Spina bifida occulta (closed spinal dysraphism) is a congenital absence of the spinous process and various portions of the vertebral arches, without an overt pathology of neural tube formation itself, in which there is no contact between the meninges or neural tissue and the external surface.

Closed spinal dysraphism may be represented by a thickened terminal fi-lum, lipomyeloschisis, a dermal sinus, diastematomyelia, and syringomyelia of the terminal segment of the spinal cord (Figure 1).

The tethered spinal cord syndrome is identified in various forms of spi-nal dysraphism, in patients with post-traumatic and post-inflammatory cicatri-cial and proliferative changes, as well as in other pathological conditions. Clin-ically, it manifests as a mosaic combination of sensory disturbances, weakness in the lower extremities, pelvic dysfunction, trophic disorders, and osteoarticu-lar deformities [1–7, 13, 14].

The true incidence of tethered spinal cord syndrome in closed forms of dysraphism is unknown; however, the incidence of neural tube defects in which tethered spinal cord syndrome develops remains relatively high even in devel-oped countries. Thus, in Japan it is approximately 0.2 cases, and in the United States from 0.8 to 1.4 cases per 1,000 newborns [8, 15, 17].

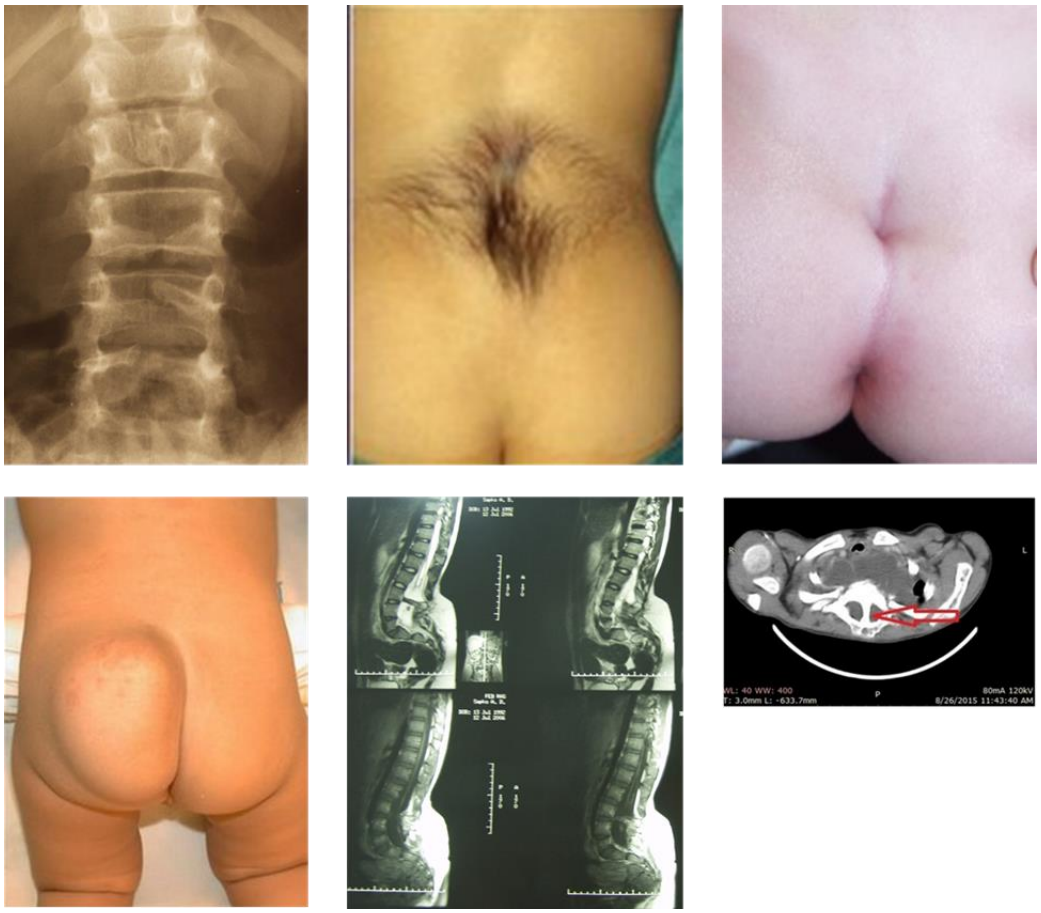

Figure 1. Various forms of spina bifida occulta, or the so-called closed spinal dysraphism.

The pathophysiological mechanisms of the development of tethered spi-nal cord syndrome are associated with traction of the caudal segments of the spinal cord between the distal pair of denticulate ligaments and any non-elastic structure that fixes it caudally [3, 4].

Surgical treatment of tethered spinal cord syndrome consists of eliminat-ing fixation factors, in particular: excision of the pathologically altered filum terminale (fatty degeneration, thickening, shortening, decreased elasticity), re-moval of pathological tissue (lipomas, diastemas, dermoids, dermal sinus tracts), and elimination of cicatricial and arachnoid adhesions [1–7, 9, 14]. Af-ter surgery aimed at eliminating fixation, in some patients (from 5 to 50 per-cent) the clinical and neuroimaging manifestations of tethered spinal cord syn-drome may recur [10, 16]. It has been established that with incomplete elimina-tion of fixation, the recurrence rate may reach 80 percent [12]. Surgical inter-ventions for lipomyelomeningocele are accompanied by recurrence of tethered spinal cord syndrome in nearly 60 percent of cases, requiring repeat surgery [10].

Thus, success in the treatment of tethered spinal cord syndrome largely determines the subsequent quality of life of patients [12, 13]. Significant diffi-culties in the diagnosis and treatment of tethered spinal cord syndrome are caused by the manifestation of concomitant pathological conditions (hydro-cephalus, Arnold–Chiari malformation, syringohydromyelia) [12, 13], as well as by latent and asymptomatic forms of the disease [16]. The development of standards and treatment recommendations for children with various forms of tethered spinal cord syndrome, including those combined with other develop-mental anomalies and their consequences (hydrocephalus, Arnold–Chiari mal-formation, syringohydromyelia), appears to be highly relevant.

Objective of the study. Within the framework of research on closed spi-nal dysraphism, it was necessary to establish a correlation between clinical manifestations, in particular the level of the lesion, and intraoperative neuro-physiological parameters, as well as imaging changes of pathological spinal tracts in tethered spinal cord syndrome associated with this anomaly in chil-dren of different age groups.

METHODS

The study included 28 patients aged from 5 months to 12 years with tethered spinal cord syndrome caused by closed spi-nal dysraphism. These patients underwent baseline and intraoperative neu-romonitoring with recording of spontaneous and evoked bioelectrical activity of the striated muscles of the lower extremities and the perineum, as well as diffusion-weighted

magnetic resonance tractography.

The neurological status was assessed in the preoperative and postopera-tive periods.

Considering that one of the main factors determining the quality of life and the degree of social adaptation of children with tethered spinal cord syn-drome is the level of motor deficit [8], the functional motor level of all patients was assessed to objectify the clinical examination data before and after surgery [9]. For this purpose, during the neurological examination, the most proximal level of the spinal cord was identified at which muscle strength in the lower ex-tremities was at least 3 out of 5 points according to the Medical Research Council scale. In patients with asymmetric motor deficit, the more proximal level of involvement was taken into account. Groups of patients were distin-guished in whom voluntary movements in the lower extremities were complete-ly absent (motor level Th), in whom flexion and adduction in the hip joints were preserved (motor level L1–L2), knee extension was preserved (motor lev-el L3–L4), ankle extension was preserved (motor level L4–L5), as well as pa-tients in whom all voluntary movements in the lower extremities were pre-served, but weakness of the knee flexors or foot muscles was identified (motor level S).

Before and after surgery, motor potentials were recorded from the mus-cles of the lower extremities (m. rectus femoris, m. tibialis anterior, m. gas-trocnemius). The amplitude and latency of the motor responses were assessed.

During the study, a statistically significant correlation was determined between neuromonitoring parameters and the data from diffusion-weighted magnetic resonance tractography (DW-MRI), as well as neurological symp-toms.

After intubation of the child, motor M-responses were recorded from target muscles: m. quadriceps femoris, m. biceps femoris, m. tibialis anterior, m. adductor hallucis, and m. sphincter ani. Additionally, the overall response of the pyramidal tract was recorded—transcranial electrical stimulation with registration of motor-evoked potentials from the leg muscles and external sphincter. Anesthetic sedation was performed using intravenous propofol, since the use of inhalation anesthetics significantly increased the threshold of the motor response (Figure 2).

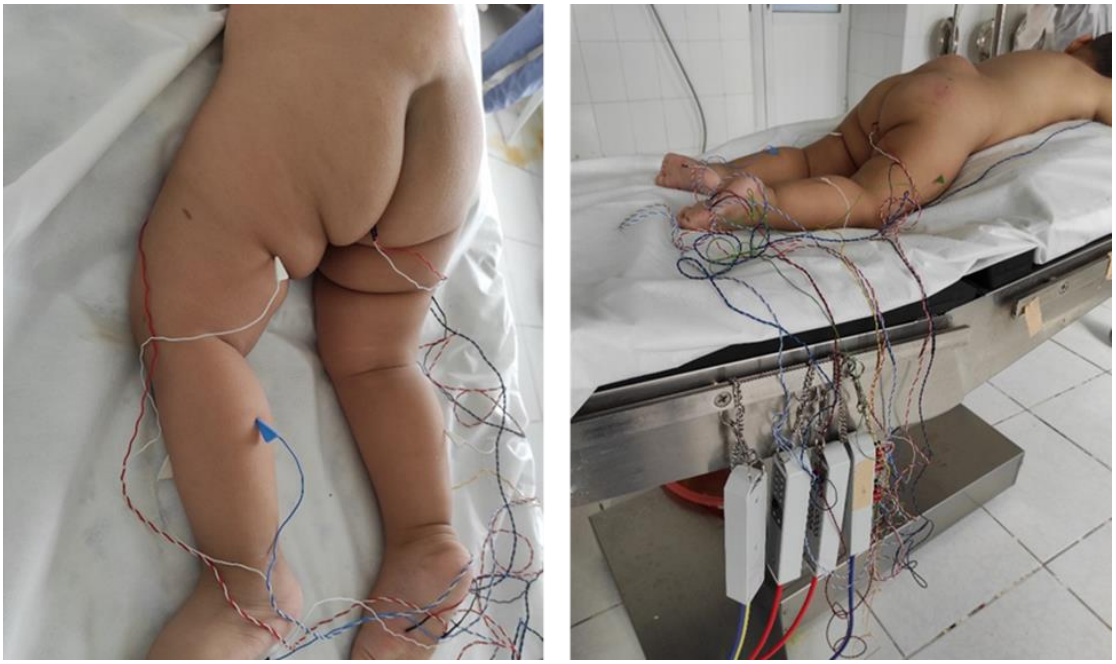

Figure 2. Electrode placement for intraoperative neuromonitoring.

Spinal cord tractography is a diagnostic method based on diffusion-weighted magnetic resonance imaging (DW-MRI), which allows visualization of the orientation and integrity of conducting pathways in vivo. Using tractog-raphy data, we also assessed the level of tract interruption (LTI) (Figure 3).

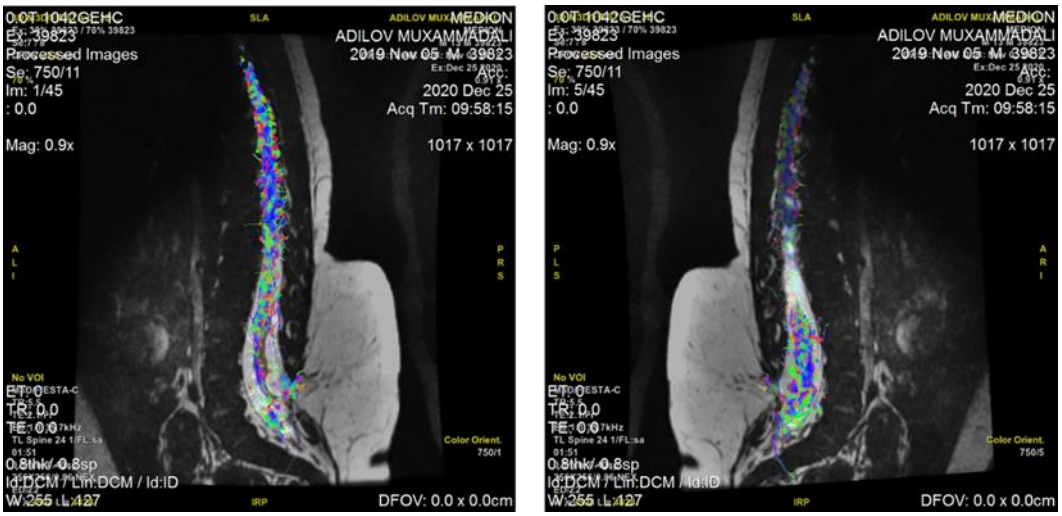

Figure 3. Spinal cord tractography—a diagnostic method based on diffusion-weighted magnetic resonance imaging.

Since tethered spinal cord syndrome is a dynamic process, we also con-sidered the age aspect of the child.

RESULTS AND DISCUSSION

The study of the clinical picture revealed that the characteristic clinical manifestations in these patients were moderate sensory and motor disturb-ances, which did not correspond to specific myotomes or dermatomes, as well as uniform suppression of tendon reflexes, disruption of pelvic function regu-lation (paroxysmal incontinence, retention), progressive scoliotic deformation of the spine, and shortening of one of the lower extremities accompanied by foot deformity. The development of clinical symptoms was associated with pe-riods of accelerated growth in children. Thus, Table 1 reflects the correlation between the child's total motor deficit and the tractography data regarding tract interruption (Table 1).

Dependence of clinical manifestations on the level of tract interruption accord-ing to DW-MRI tractography (Table 1).

Functional motor level	The level of tract interruption L5—S1	The level of tract interruption L4—L5	The level of tract interruption L3—L4
S (n=11)	9 (81,1%)	2 (18,1%)	0
L4—L5 (n=10)	0	8 (80%)	2 (20%)
L3—L4 (n=7)	0	0	7 (100%)

From the given table, it can be seen that there is a direct and strong cor-relation between the clinical manifestations of spinal cord tension syndrome and the tractography data. As shown in the table, 4 out of 28 patients did not have a correspondence between the clinical manifestation and the level of tract interruption. These patients belonged to the age group over 5 years, which in-dicates and demonstrates the dynamic nature and a wider extent of clinical manifestations in the proximal direction.

After the surgery aimed at eliminating spinal cord fixation, regression of the clinical manifestations of SFMS was observed in all patients of this group. This was also confirmed by ENMG data, including the nerve conduction veloc-ity (NCV) and the nerve excitability threshold (NET).

To determine the correspondence between intraoperative direct stimula-tion activity of the nerves and the visualization of spinal tracts, all 28 patients underwent stimulation EMG at the site of fixation of the neural structures: 1) to the meningeal coverings; 2) to the deep tissues; 3) neurophysiological identi-fication of the terminal filament assumed based on tractography (Table 2).

Table 2.

The relationship between intraoperative stimulation EMG and tractography DV.

Evoked EMG	The number of matches between stimulation and tractography data	Reliability
Stimulation of the superficial nerve roots	22 (78,5%)	P<0,05
Stimulation of the deep nerve roots	12 (42,8%)	P<0,05
Stimulation of the terminal filament	25 (89,2%)	P<0,05

This table illustrates the correspondence between direct evoked activity of the nerve roots and DV MRI tractography data.

In 19 out of 28 patients, a direct and strong correlation was observed be-tween the data from stimulation electromyography and MRI features during the registration of nerve structures associated with superficial tissues. Stimula-tion of the deep nerve roots did not reveal a statistically significant correlation. However, the terminal filum was clearly identified on tractography, which cor-responded to the findings of stimulation, and, moreover, it was visually diffi-cult to confuse it with nerve structures.

CONCLUSIONS

1. The clinical manifestations of a tethered spinal cord reliably correspond to the data obtained from diffusion-weighted MRI tractography.

2. In the age group older than five years, the clinical signs of tethering shift in a proximal direction.

3. Active stimulation electromyography and tractography serve as com-plementary intraoperative methods for specifying the location of nerve structures.

4. Nerve structures that are fixed to the overlying tissues coincide with their locations on diffusion-weighted MRI tractography in 78.5% of cas-es. A statistically significant strong direct correlation between electro-myography data and tractography was observed in the patients of the study group.

5. Deeply located nerve structures do not demonstrate a statistically signif-icant correlation, and diffusion-weighted tractography cannot be used in planning the release of tethered nerve structures.

6. Diffusion-weighted tractography reliably identifies features of the termi-nal filum and structures to be released, which are not at risk for causing neurological deficits.

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