Surgical treatment of tethered cord syndrome showed promising outcome in young children with short duration

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The inclusion criteria were 1) TCS patients who were clinically and radiologically (MRI) confirmed; 2) patients who underwent neurophysiological monitoring during surgery. Patients with previous lumbar surgery, tumor, trauma, and other neurological diseases were excluded. This study was performed in accordance with the Helsinki declaration and approved by the ethical committee of Wuhan Children’s Hospital.

Surgical Procedure
All children underwent general anesthesia in the prone position, in order to prevent excessive loss of cerebrospinal fluid due to the gravity. The sacrococcygeal region was raised by putting an air pillow at the hip joint and the lumbosacral mass was gently cut. Needle electrodes were then implanted in the anal sphincter, bilateral quadriceps femoris, anterior tibial and gastrocnemius muscle to assess neurophysiological condition of patients during the surgery.

The thickened filum terminale was explored and the spinal cord adhesion was peeled under electrophysiological monitoring. For the assessment of myelomeningocele type and lipoma type, the neck of the bulging capsule was fully exposed, and a longitudinal dural incision was made from top to bottom in order to expose the bulging spinal nerve. Electromyographic activity was measured using an electrophysiological probe after stimulation and resting potential. Among the lipomatous infiltrations, filar lipoma is relatively simple to remove. For the lipomyelocele type, a cavitron ultrasonic surgical aspirator (CUSA) was applied for sharp segmented resection of lipoma under electrophysiological monitoring. This device was safe enough to protect the spinal nerve, avoiding repeated pulling and sharp damage. The spinal cord wound was tightened and sutured appropriately to prevent re-adhesion.

Evaluation and Postsurgical Follow-up
The medical history and demographic data of patients were collected from the hospital. SBNS (Spina Bifida Neurological Scale) score was adopted for evaluation of clinical observations pre- and post-surgery. The recovery of neurological symptoms was evaluated according to the Hoffman classification. Normal functions of the bladder and bowel and muscle strength normal sensation were considered as cure criteria. To be considered as improved, enhancement of urination, defecation, and neuropathy were taken into account. The stationary term was assigned to unchanged factors, while the deteriorated term was assigned to newly developed or worsened symptoms.

In children without pre-surgery neurological symptoms, the occurrence of urination or defecation dysfunctions, without affecting daily life, was defined as a “mild progression. Severe progression in the postsurgical follow-up was defined as the occurrence of the above dysfunctions affecting daily life. For at least two years, all children with TCS were followed up on at the clinic or by phone.

Statistical Analysis
Chi-squared test or Fisher’s exact test was used for data analysis. Beneficial post-surgery factors were identified and test by univariate and multivariate analysis respectively. All statistical analysis was performed using SPSS Software Version 25.0 (IBM Corp., Armonk, NY, USA) and p-value <0.05 was considered as significant.

Results
Clinical Characteristics
Among the 80 children with TCS, 43 were male and 37 were female. Cutaneous abnormalities were the most common signs and ladder dysfunction was the most common symptom before surgery. The median duration of preoperative symptoms was 3 months (range: 1 to 156 months). The median SBNS score of all children was 13.2 (range: 8.0 to 15.0). Lipoma was frequently observed in children, and lipoma with thickened filum terminale was accounted for 52.5% of pathological types. The transitional lipoma location was more frequently observed than other locations (Table I).

Surgical Outcomes
In 79% of children with preoperative symptoms, the surgical treatment led to the cure or improvement (Figure 1A). Table II shows the detailed cases of cure/improvement and station/deterioration among each symptom. In 74% of children with asymptomatic TCS, neurological deficits can be prevented by surgery (Figure 1B). Table III depicts the detailed types of progressive symptoms in the follow-up. Figure 1C displays that SBNS score is significantly increased after surgery. Due to the neurophysiological monitoring, surgical complications were rarely observed in 4 children (5.0%). Of them, two showed cere-
Surgical treatment of TCS showed promising outcome in young children with short duration.

Surgical treatment of TCS showed promising outcome in young children with short duration and two cases with central nervous system infection were observed.

Factors Which Benefit from Surgical Intervention

Univariate analysis showed that patients of less than one year of age received significant surgical intervention benefits, including normal lipoma location and elimination of cutaneous abnormalities after a period of 3 months. Multivariate analysis considered age and duration as independent protective factors (Table IV).

Discussion

TCS is an important health issue for the pediatric population. Since the length of the spine grows with age, tethering leads to ischemia as a result of

Table I. Clinical characteristics.

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>No. (%) n=80</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sex</td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>43 (53.8)</td>
</tr>
<tr>
<td>Female</td>
<td>37 (46.2)</td>
</tr>
<tr>
<td>Age</td>
<td></td>
</tr>
<tr>
<td>&lt; 1 years</td>
<td>34 (42.5)</td>
</tr>
<tr>
<td>1-5 years</td>
<td>37 (46.2)</td>
</tr>
<tr>
<td>&gt; 5 years</td>
<td>9 (11.3)</td>
</tr>
<tr>
<td>Post-term birth</td>
<td>13 (16.2)</td>
</tr>
<tr>
<td>Symptoms and signs</td>
<td></td>
</tr>
<tr>
<td>Cutaneous abnormalities</td>
<td>58 (72.5)</td>
</tr>
<tr>
<td>Bladder dysfunction</td>
<td>35 (43.8)</td>
</tr>
<tr>
<td>Bowel dysfunction</td>
<td>24 (30.0)</td>
</tr>
<tr>
<td>Leg weakness</td>
<td>25 (31.3)</td>
</tr>
<tr>
<td>Abnormal gait</td>
<td>10 (12.5)</td>
</tr>
<tr>
<td>Hyperreflexia</td>
<td>9 (11.3)</td>
</tr>
<tr>
<td>Back pain</td>
<td>3 (3.8)</td>
</tr>
<tr>
<td>Pathological type</td>
<td></td>
</tr>
<tr>
<td>Lipoma</td>
<td>36 (45.0)</td>
</tr>
<tr>
<td>Thickened filum terminale</td>
<td>2 (2.5)</td>
</tr>
<tr>
<td>Mixed type</td>
<td>42 (52.5)</td>
</tr>
<tr>
<td>Lipoma location</td>
<td></td>
</tr>
<tr>
<td>Transitional lipoma</td>
<td>16 (20.0)</td>
</tr>
<tr>
<td>Dorsal lipoma</td>
<td>9 (11.3)</td>
</tr>
<tr>
<td>Filar lipoma</td>
<td>9 (11.3)</td>
</tr>
<tr>
<td>Caudal lipoma</td>
<td>8 (10.0)</td>
</tr>
<tr>
<td>Radiographic sign</td>
<td></td>
</tr>
<tr>
<td>Low-lying conus</td>
<td>63 (78.8)</td>
</tr>
<tr>
<td>Lipomatous infiltration</td>
<td>35 (43.8)</td>
</tr>
<tr>
<td>Thickened filum terminale</td>
<td>2 (2.5)</td>
</tr>
<tr>
<td>M (min-max)</td>
<td>7 (0-43)</td>
</tr>
</tbody>
</table>

SBNS: Spina Bifida Neurological Scale.
In most TCS cases, early surgical intervention leads to stabilization or improvement of neurological dysfunctions.15 Surgical treatment may significantly prevent the irreversible neurological impairments in children with TCS if at early ages.16 Our study was conducted on children with the age of ≤3 months and results documented the benefit of surgical intervention since a normal spinal is essential for normal neurological function.

Spinal lipoma accounts for 70% of tethering cases.17 Symptoms in TCS children with lipomyelocele were relatively severe because dissociating and completely removing the fat surrounding filum terminale was difficult and was also accompanied by subsidiary-injury risks and a poor prognosis for improvement in TCS patients.18 The location of filar lipoma was independently associated with improved neurosurgical symptoms when treated with surgery in this study, indicating that the filum terminale was relatively easy to dissociate and completely remove in filar lipoma type.

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Prophylactic surgery for TCS children is also controversial. The rate of shift from asymptomatic to symptomatic TCS is reported to be 3%-4% per year, with a hazard ratio of 40% in 10 years.19 Based on previous research,20 neurological manifestations in asymptomatic TCS can be disappeared for 7 years after the initial surgery, suggesting that in terms of asymptomatic TCS, prophylactic surgery would be effective in a certain period. In this study, 74% of asymptomatic TCS children who took the surgery were free of neurological symptoms in the follow up, indicat-
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Informed Consent
The guardians of the study participants gave written informed consent for their respective minors to participate in the study.

Ethics Approval
This research complied with the guidelines for human studies and was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. The study was approved by the Medical Ethics Committee of Wuhan Children's Hospital, Tongji Medical College, Huazhong University of Science and Technology (approval No. 2022R109-E01), which is equivalent to an IRB committee.

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Availability of Data and Material
All data generated or analyzed during this study are included in this published article.

Authors’ Contributions
H.D. and W.Y.D. designed the research and wrote the manuscript; M.Y.L. collected and analyzed the data; Y.Y.L, Y.Z.H. and L.Y.H. contributed to the analysis of the study; Y.Y.L and Y.Z.H. supervised the study and contributed to the writing of the manuscript.

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Limitations
Nonetheless, our study had also some limitations. Aside from surgical skills, neurophysiological instruments and infection prevention strategies were required for a reconsideration. Moreover, we were not able to extend the time and sample size of our study, which is an important issue when speaking about the advantages of surgical intervention. On the other hand, we hadn’t access to homogenous samples from TCS cases for a more detailed study while for valid results this is a must-do. Consequently, more randomized trial studies are required with larger samples in future’s studies.

Conclusions
This study showed that surgical intervention is remarkably efficient for decreasing the rate of TCS cases. Moreover, electrophysiological monitoring is documented as an effective approach to decreasing the rate of spinal damage and to having a safe and well-oriented surgery. This study also concluded that such a surgical intervention would be extremely effective at early ages and decrease the likelihood of future CNS-related disorders in TCS patients. These were independent protective factors for surgical intervention.


