

SURGICAL MANAGEMENT OF TETHERED CORD SYNDROME

By

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ABSTRACT

Background: Tethered cord syndrome is a progressive anomaly resulting in neurological, orthopedic and urological dysfunction caused by the anchoring of the spinal cord by deferent pathologies. The underlying pathophysiological processes include decreased blood flow impaired oxidative metabolism and abnormal glucose metabolism.

Objective: To evaluate clinical outcome of surgical detethering for cases of tethered cord syndrome at Al-Azhar University Hospitals.

Patients and Methods: We worked on 25 cases Adults and children with symptomatic tethered cord syndrome operated by microscopic detethering and treatment of the associated pathology using intraoperative neurophysiologic monitoring at Al-Azhar university hospitals between February 2017 and October 2020 and follow up was done for at least 6 months postoperatively. All cases were subjected to History, clinical examination and MRI LSS pre and postoperatively.

Results: Untethering procedures were performed in 25 patients (age range, 3 months-26 years), 13 males and 12 females. The most common preoperative sign or symptom was pain (94%), followed by motor deficits (94%), sphincter affection (62.5%), and foot deformity (32%). The level of conus in the preoperative MRI was low lying in 24 patients (96%). After detethering, pain was the most responsive to surgery with 86.7% improvement, followed by weakness (66.7%), then sphincters (50%). Foot deformity showed no spontaneous improvement but was stationary in 62.5% of cases, and still progressive in 37.5% of cases.

Conclusion: The surgical outcome was excellent for the resolution of pain and good for motor deficits, but disappointing for bladder dysfunction. So, Early diagnosis and adequate surgical release might be the keys to a successful outcome in school aged children, adolescents, and young adults with TCS.

Key words: tethered cord, evaluation of surgical outcome, neurophysiological monitoring, thickened filum terminale and back lipoma.

INTRODUCTION

In the healthy spine, the spinal cord moves unimpeded with spinal fluid pulsation in the rostral and caudal directions. When a portion of the spinal cord becomes attached to lesions within the spinal column, excess strain can cause

signs and symptoms such as pain, motor deficits, sensory deficits, bladder dysfunction, and bowel dysfunction. This condition is termed tethered cord syndrome. (O'Connor et al., 2020). Patients also may be asymptomatic, which does not exclude the diagnosis of TCS (Sanchez et al., 2014). TCS was described

in 1976 by Hoffman *et al.* after observing that the spinal cord was tethered via a thickened filum terminale to the sacral bones in 31 children and that there was a noticeable neurological improvement following the release of the cord (*Abdallah et al., 2018*). Spine MRI demonstrates elongation and caudal descent of the conus medullaris (below L2 vertebral level) and a fatty filum terminale (usually >2 mm in diameter) (*Malek et al., 2020*). Myelomeningocele, lipoma, lipomyelomeningocele, diastematomyelia, dermal sinus tract, and dermoid sinus are the usual associated causes (*Shukla et al., 2018*). Traction and elasticity of the spinal cord were fundamental factors underlying the pathophysiology of the disease. Increase in the traction forces on the spinal cord reduces blood flow to the cord and spinal evoked potentials demonstrated patterns consistent with ischemia. A decrease in the diameter of the lumen of spinal vessels, due to traction, substantially reduced the total spinal blood flow, causing local ischemic insult (*Filippidis et al., 2010*). The classic definition of the TCS involves the presence of a thickened filum terminale and/or a low-lying conus medullaris in a patient with neurological deficits. Currently, a more accepted diagnosis of TCS is defined as a pathological fixation of the spinal cord in an abnormally lying position.

The data, derived from the pathophysiology of the syndrome, indicate that mechanical tension of the caudal spinal cord, vascular compromise, and hypoxia result in metabolic derangements and neurological impairment. Although the radiological evidence of a low-lying conus are the key factors in the diagnosis

of TCS, a clinical picture consistent with TCS can also be present in a group of patients—accounting for 14–18% of various published series with a normal anatomical position of the conus (*Filippidis et al., 2010*). Surgical treatment is the only effective method to relieve occupying, loose adhesions, and compression, its main purpose is to lift the tethered to reduce the stretching of the taper tension, and thus to control further development of symptoms and to reduce further damage to the nerve function (*Gao et al., 2016*). Intraoperative neurophysiological monitoring (IONM) has been widely used as a tool to improve surgical results concerning safety (prevention of neurological morbidity) and efficacy (lasting effect of detethering). Tethered spinal cord surgery with the use of IONM seems to be long-term effective on the neurological, urological, and pain domains (*Dulfer et al., 2017*).

The present work aimed to evaluate the clinical outcome of surgical detethering for cases of tethered cord syndrome at Al-Azhar University Hospitals.

PATIENTS AND METHODS

This study included 25 cases with symptomatic tethered cord syndrome operated at Al-Azhar University Hospitals in Cairo 13 males (52%) and 12 females (48%). The present work included children and adults between 3 months and 26 years old with main age of 6.25 years, 22 child (88%) younger than 18 yrs. old, and 3 adults (12%). All cases were operated with microscopic surgical detethering using intraoperative neurophysiological monitoring between February 2017 and October 2020 and

follow up was done for at least 6 months postoperatively. We excluded from our study asymptomatic patients of tethered cord syndrome, and patients with associated infected meningocele or meningomyelocel.

The following was applied for the studied cases:

History Taking: The personal history taking including name, age, sex, occupations and symptomatology including low back pain (VAS), motor, sensory, sphincter affection and associated foot deformities. This included analysis of the patient complaint was the mode of onset, the duration and the course of illness.

From the 25 cases in our study, we have 9 patients younger than 4 years old, and they could not express their low back pain and could not give accurate history about their sphincter control. Past history of surgical detethering Neurological and neurosurgical problems. **Other medical problems:** Hypertension, diabetes, tuberculosis, renal, cardiac, chest and surgery.....etc.

Examination:

1. General Examination included general appearance, pulse, temperature, blood pressure, respiratory rate, chest, heart, abdomen, uro-genital and skeletal systems examination.
2. Neurological Examination: Motor system: Motor power, tone and state of muscle. Reflexes: superficial, deep and pathological reflexes. Sensation: Superficial and deep sensations. Low back for skin stigmata of spina pifida eg. Hyperpigmented hairy skin, back lipoma, meningocele, or scar of

previous surgery. Lower limbs for any associated foot deformities

Investigations:

1. Routine laboratory investigations.
2. Images: Magnetic Resonance Imaging (MRI) was done for all cases using T1, T2 images sagittal and axial views. MRI to show the level of conus medullaris (which was low lying in all cases of our study), and the associated pathology (lipoma, thickened filum terminale, meningocele). All patients were submitted to plain X-ray lumbosacral spine including: lateral, Posterior, anterior views, or CT lumbosacral spine, to show associated spinapifida or split cord malformation. Uroflowmetry was done for some cases to assess urinary bladder control.

Informed Consent: Informed written consent was obtained from every patient or their first degree relatives.

Operative procedures: Prone position. Shaving of the skin at the operative field and proper sterilization using beta dine antiseptic solution. Connection of the electrodes of IONM. Sub periosteal separation of paravertebral muscles. Spin laminectomy at the level of conus based on MRI finding. Opening of the dura, Microscopic detethering and treatment of the cause (lipoma excision, meningocele repair,.....) aided by IONM, Good hemostasis and closure in layers

Follow up: All patients were followed up for an average of 6 months or more after detethering: Clinically by assessment of the subjective symptoms (as. Back pain and urine control) and assessment of the functional outcome by visual analog scale, power, foot deformity and sphincter

control and imaging by MRI LSS to assess adequacy of detethering, the associated cause and level of conus.

Statistical analysis:

Data were analyzed using Statistical Package for the Social Sciences (SPSS) version 24. Quantitative data were

expressed as mean \pm standard deviation (SD). Qualitative data were expressed as frequency and percentage.

RESULTS

As regard age, the mean age of all studied patients was 6.52 ± 7.8 years with minimum age of 0.1 years and maximum

age of 26 years. As regard sex, there were 13 males (52%) and 12 females (48%) in the studied patients (**Table 1**).

Table (1): Parameters of age And sex in all studied patients

| Parameters | | Studied patients (N = 25) | |
|------------|---------------|---------------------------|-----|
| Age(years) | Mean \pm SD | 6.52 \pm 7.8 | |
| | Min – Max | 0.1 – 26 | |
| Sex | Males | 13 | 52% |
| | Females | 12 | 48% |

The description of associated pathology in all studied patients. Thickened filum terminale was present in 6 patients (24%), Lipoma was present in 4 patients (16%), Spina bifida occulta was present in 1 patient (4%), Dermal sinus

tract was present in 1 patient (4%), TCS after previous meningocele repair was present in 6 patients (24%) and meningocele was present in 8 patient (32%) (**Figure 1**).

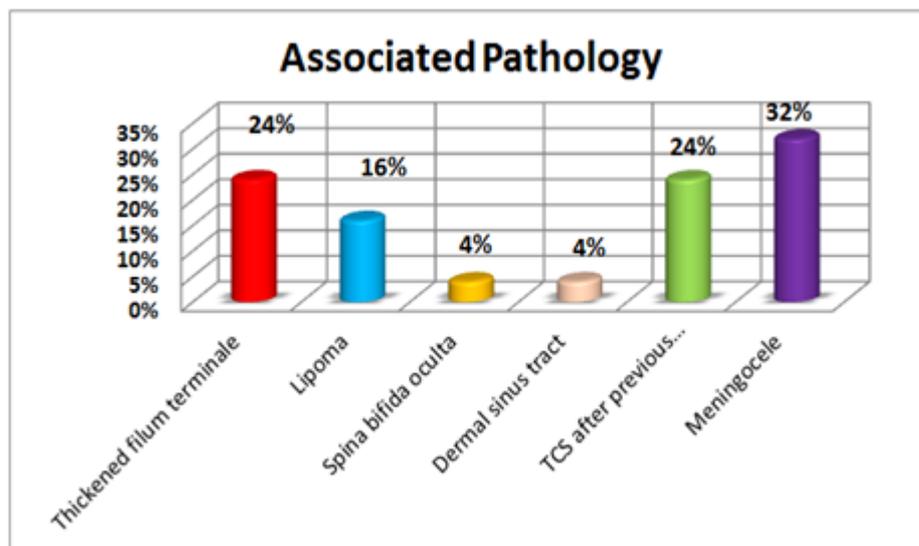


Figure (1): Associated Pathology in all studied patients

Pre-operatively, there were 15 patients (60%) with pain, 1 patient (4%) without pain, while there were 9 patients cannot be assessed because of their young age. Post-

operatively, there were 13 patients (86.7%) improved and 2 patients (13.3%) not improved (Table 2).

Table (2): Pain in all studied patients

| Parameters | | Studied patients (N = 16) | |
|---|--------------|---------------------------|-------|
| Pre-operative pain | No | 1 | 6.2% |
| | Yes | 15 | 93.8% |
| Post-operative pain assessment of 15 patients with pain | Improved | 13 | 86.7% |
| | Not Improved | 2 | 13.3% |

Pre-operatively, there were 7 patients (28%) with full power and 18 patients (72%) with power deficit. Post-

operatively, there were 12 patients (66.7%) improved and 6 patients (33.3%) not improved (Table 3).

Table (3): Power assessment in all studied patients

| Parameters | | Studied patients (N = 25) | |
|--|---------------|---------------------------|-------|
| Pre-operative power | Full power | 7 | 28% |
| | Power deficit | 18 | 72% |
| Post-operative power assessment in 18 patients | Improved | 12 | 66.7% |
| | Not Improved | 6 | 33.3% |

Pre-operatively, there were 6 patients (37.5%) with intact sphincter and 10 patients (62.5%) with sphincter deficit, while there were 9 patients cannot be

assessed because of their young age. Post-operatively, there were 5 patients (50%) improved and 5 patients (50%) not improved (Table 4).

Table (4): Sphincter assessment in all studied patients

| Parameters | | Studied patients(N = 16) | |
|--|-------------------|--------------------------|-------|
| Pre-operative sphincter | Intact | 6 | 37.5% |
| | Sphincter deficit | 10 | 62.5% |
| Post-operative sphincter assessment in 10 patients | Improved | 5 | 50% |
| | Not Improved | 5 | 50% |

The pre-operative foot deformity was reported in 8 cases (32%) and the other 17 cases have no foot deformity, and after detethering the foot deformity shows no

spontaneous improvement but was stationary in 5 cases (62.5%)and still progressive in the other 3 cases (37.5%)(Table 5).

Table (5): Skeletal deformity (pre and post-operative) in studied patients

| Parameters | | Studied patients(N = 25) | |
|--|-------------|--------------------------|-------|
| Pre-operative Skeletal deformity | No | 17 | 68% |
| | Yes | 8 | 32% |
| Post-operative skeletal deformity course in 8 patients | Stationary | 5 | 62.5% |
| | Progressive | 3 | 37.5% |

The description of previous intervention in all studied patients. VP shunt was done in 2 patients (8%), dermal sinus repair was done in 1 patient (4%), Detethering 3 times was done in 1 patient (4%), meningocele repair was done in 2 patients (8%), meningocele repair + orthodesis for talipus was done in 1 patient (4%), meningocele repair then untethering was done in 1 patient (4%),

meningocele repair then VP shunt insertion was done in 1 patient (4%), Menigomyelocele repair was done in 1 patient (4%). while 15 patient (60%) have no previous surgical intervention and 2 patients (8%) have only VP shunt without previous spine surgery, so we have 17 patient (68%)with primary tethered cord (**Table 6**).

Table (6): Previous intervention in all studied patients

| Parameters | | Studied patients (N = 25) | |
|-----------------------|---|---------------------------|-----|
| Previous intervention | No intervention | 15 | 60% |
| | VP shunt | 2 | 8% |
| | dermal sinus repair | 1 | 4% |
| | Detethering 3 times | 1 | 4% |
| | meningocele repair | 2 | 8% |
| | meningocele repair + orthodesis for talipus | 1 | 4% |
| | meningocele repair then untethering | 1 | 4% |
| | meningocele repair then VP shunt insertion | 1 | 4% |
| | Menigomyelocele repair | 1 | 4% |

The level of conus was low lying(below L1-L2 disc space) in 24 patient (96%), and was in normal level in 1 patient. we reported ascent of level of

conus post-operative in 3 cases (12%) and still at the same pre-operative level in 22 cases (**Table 7**).

Table (7): Post-operative level of Conus in all studied patients

| Parameters | | Studied patients (N = 25) | |
|-------------------------------|-----------------|---------------------------|-----|
| Post-operative level of Conus | At same level | 22 | 88% |
| | Ascent of Conus | 3 | 12% |

There were post-operative complications in 3 patients (12%), 2 patients had CSF leak and 1 patient had

wound infection. All cases respond well to conservative treatment (**Table 8**).

Table (8): Post-operative complications in all studied patients

| Parameters | Studied patients(N = 25) | | |
|---|--------------------------|----|-------|
| Post-operative complications | No | 22 | 88% |
| | Yes | 3 | 12% |
| Post-operative complications in 3 cases | CSF leak | 2 | 66.7% |
| | Wound infection | 1 | 33.3% |

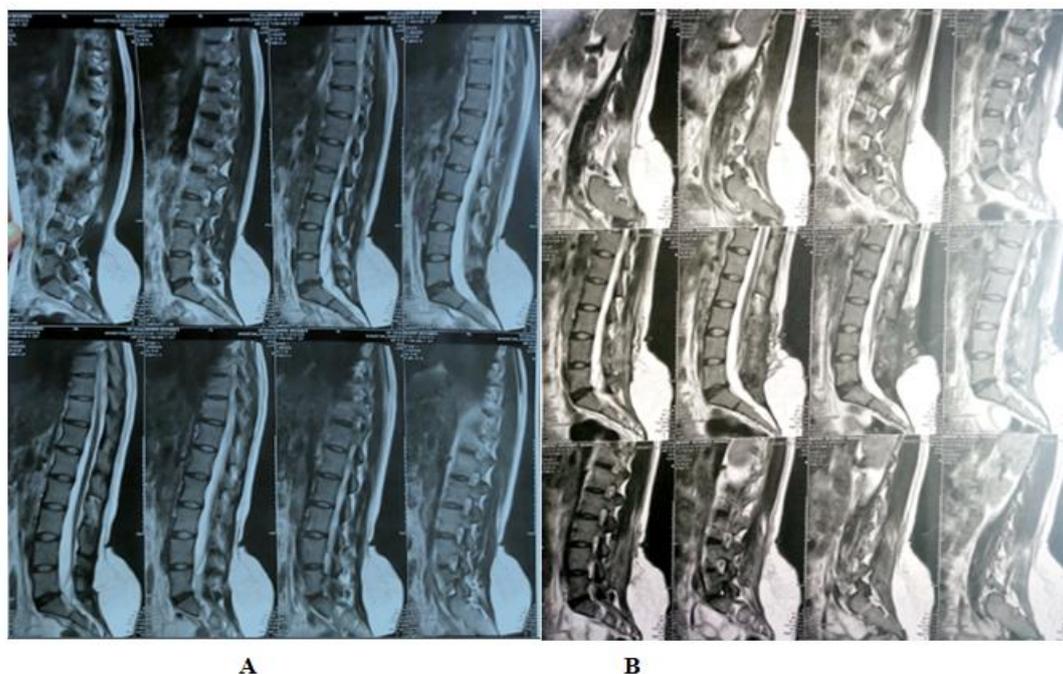


Figure (2): MRI pre (a) and post(b) operative showing spina bifida occulta and back lipoma with low lying conus medullaris (at L4-5 disc space) for female patient 22 yrs old presented with low back pain and Paraparesis. Microscopic surgical detethering was done using IONM and post-operative follow up for 1 year shows marked improvement in back pain and patient now is full power. Also MRI post after 4 months shows regression in level of conus to upper border of L4

DISCUSSION

In this study, 52% males and 48% females including children and adults between 3 months and 26 year old with main age of 6.25 year.

In the work of Kang *et al.* (2010), untethering procedures were performed, 58.3% were males and 41.6% were females with age range 7-25 years. Garg *et al.* (2014) found that 45.8% were males and 54.2% were females. Patient's age

ranged from 16 to 32 years. Thuy *et al.* (2015) mentioned that 42.6% were females and 57.4% were males. The median age at detethering surgery was 1.4 years old. Maurya *et al.* (2016) stated that 33.4% were males and 66.6 were females. The mean age of patients in this study was 5.9 - 7 years. Overall, 52% patients had deficits and 48% were asymptomatic. O'Connor *et al.* (2020) worked on a comprehensive literature review on 730 patients, 65% were females and 44% were

males and all patients were adults with an average age of 35.6 years.

In this study, the associated cause of tethering in MRI was most commonly meningocle by 32%, followed by thickened filum and tethering after previous meningocle repair by 24% for each, back lipoma was found in 16%, and there was 4% spina pifida occulta plus 4% dermal sinus tract. In the work of *Kang et al. (2010)*, the most common tethered lesions were intradural lipomas, thickened filum and fibrous band adhesions into the placode sac. In the work of *Thuyet et al. (2015)*, the most common tethering pathologies were spinal lipomas in 52.5%, filum abnormalities in 37.7%, and dermal sinus tracts in 13.1% and diastematomyelia in 11.5%. In the work of *Maurya et al. (2016)*, the most common tethering diseases were dermal sinus and fatty filum, seen in 29% each. In the work of *O'Connor et al. (2020)*, a thick and/or fatty filum terminale was the most common radiographic finding, affecting 37% of patients, followed by intradural or extradural lipoma in 29%, lipomeningocele or lipomyelomeningocele in 21%, fibrous adhesions in 8%, split cord malformation in 17%, and previous myelomeningocele repaired in 13%.

In this study, the pain was assessed only in 16 case and the other 9 cases were younger than 4 years old, and cannot express their pain. The pre-operative back and lower limb pain was reported in 15 cases (94%), and only 1 case has no back or lower limb pain and pain was the most common preoperative sign or symptom. We reported improvement of pain post-operative in 86.7%, and not improved in

13.3%, of cases which was significant. In the work of *Kang et al. (2010)*, pain was the most common presenting symptom, and was a prominent complaint in 92%. The relief of pain was 95% with significant pain became asymptomatic following surgery. *Garg et al. (2014)* stated the low backache was the most common presenting symptom in 75%, and at the time of final follow up 83.3% patients had shown improvement in backache. In the work of *Thuy et al. (2015)*, only 9.8% have back or leg pain pre-operative, and this low percent in comparison to our study is mostly because of the young age of most of the cases (the median age at detethering surgery was 1.4 years old). So, they cannot express their pain, and it appears clear that they did not take this point in consideration before calculating the percent of pre-operative pain, and 26.7% of cases have improvement or resolution of their pain post-operatively. *Maurya et al. (2016)* did not comment on pain in pre nor post-operative cases. In the work of *O'Connor et al. (2020)*, the most common pre-operative sign or symptom was pain (81%), and pain improved after detethering in 84% of cases.

In this study, the pre-operative motor deficit was reported in 94%, and the other 6% was full power, and we reported improvement in power post-operatively in 66.7% and not improved in 33.3 of cases which were considered statistically significant. *Kang et al. (2010)* stated that 75% presented with progressive leg weakness or walking difficulties. The pre-operative motor deficits improved after surgery in 66.6%. *Garg et al. (2014)* showed that pre-operative motor deficits were present in 37.5%. And post-

operative weakness improved by at least one grade in 77.8%. *Thuy et al. (2015)* showed that 18.0% had motor or gait disturbance, and improvement occurred in children with motor or gait disturbance after detethering in (36.4%). *Maurya et al. (2016)* showed that gait issues and motor deficits were the most common signs detected on neurologic examination pre-operative, noted in 38%, and this low percent in comparison to our study is mostly because they worked on symptomatic (52%) and non-symptomatic (48%) cases. Motor power improved by at least 1 grade in 6 of 8 patients (75%). *O'Connor et al. (2020)* stated that the pre-operative motor deficit was 63%, and motor deficits improved postoperatively in 61% of cases.

In this study, the pre-operative sphincter affection was assessed only in 16 cases, and the other 9 cases were younger than 4 years old, and sphincter control was not assessed. The pre-operative sphincter affection was reported in 62.5%, and there was 37.5% of patients have intact sphincter pre-operative. We reported improvement in sphincter control post-operative in 50%, and not improved in 50% which was considered statistically significant. *Kang et al. (2010)* showed that 63% had bladder symptoms. The results for sphincter dysfunction showed that 33.3% with sphincter dysfunction were helped by surgery. *Garg et al. (2014)* found that bladder involvement was recorded in 50%, and bladder symptoms improved in 50%. *Thuy et al. (2015)* showed that 18.0% sphincteric disturbance, and improvement after detethering occurred in (27.3%). *Maurya et al. (2016)* stated that sphincter disturbances were noted in 24% and this

low percent in comparison to our study was mostly because they worked on symptomatic (52%) and asymptomatic (48%) cases. Calculating the percent of power deficit from the symptomatic cases only, it was 45.5%, and improved in 3 of 5 patients (60%) who had preoperative sphincter dysfunction. *O'Connor et al. (2020)* stated that the pre-operative bladder dysfunction was 56%, and improved postoperative in 45% of patients.

In this study, the pre-operative foot deformity was reported in 32%, and the other 68.5% of cases have no foot deformity, and after detethering the foot deformity showed no spontaneous improvement, but was stationary in 62.5% and still progressive in the other 37.5%. None of our patients without a pre-existing foot deformity developed this problem after detethering. *Kang et al. (2010)* showed that 58.3% had foot deformities. *Thuy et al. (2015)* showed that 13.1% lower limb orthopedic deformities. *Maurya et al. (2016)* noted musculoskeletal abnormalities in 28.5%. But no one of these papers comment on the course of the deformity after detethering. *O'Connor et al. (2020)* showed foot deformities in 35% of preoperative patients.

In this study, the level of conus was low laying (below L1-L2 disc space) 96% of patients, and was in normal level in 1 patient. We reported ascent of level of conus post-operatively in 3 cases (12%), and still at the same pre-operative level in 88% of cases which was considered statistically non-significant. *Kang et al. (2010)* found that a low level of the conus was noted in 87.5%. But did not mention

any comment about post-operative conus level. *Kim et al. (2011)* showed postoperatively that the conus elevated in 31% of cases, and was found to be more ventrally located in 44% cases. *Garg et al. (2014)* by MRI showed evidence of low-lying (below L2) conus in all patients (100%). In the study of *Elmesallamy et al. (2019)*, conus location changed in only 7% patients at 1 year follow-up. *O'Connoret al. (2020)* did not mention any comment about conus level neither pre-nor-post-operatively.

In this study, we reported 2 cases complicated by CSF, leak and 1 case of wound infection with total 3 complicated cases (12%). All of them improved on conservative treatment. *Thuy et al. (2015)* showed that the most common complications were wound infection and cerebrospinal fluid leak. Six children (9.8%) required reoperation for wound issues. In the work of *Maurya et al. (2016)*, 10% had postoperative CSF leak from the incision. This necessitated emergency surgery to seal the site of leak. *O'Connor et al. (2020)* showed that complications occurred in 7% of patients. The most common complication was cerebrospinal fluid leak.

CONCLUSION

The surgical outcome was excellent for the resolution of pain and good for motor deficits, but disappointing for bladder dysfunction. So, Early diagnosis and adequate surgical release might be the keys to a successful outcome in school aged children, adolescents, and young adults with TCS.

Tethered cord syndrome should be included in the differential diagnosis in patients presenting with back or leg pain,

muscular weakness, sphincteric disturbance. After a definitive diagnosis is made, patients should be counseled about surgical detethering as an option. Pain is the most common presenting symptom.

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التدخل الجراحي لمناجزة متلازمة الحبل الشوكي المعلق

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خلفية البحث: تعد متلازمة الحبل الشوكي المعلق ظاهرة طبية تنتج عن تقييد الحركة التشريحية للحبل الشوكي داخل القناة الشوكية وكذلك التأثير المباشر على الإمداد الدموي له مما يؤثر على أيض الأنسجة العصبية و الوظيفة الحيوية لها. وتصنف متلازمة الحبل الشوكي المعلق إلى أولية كعيب من العيوب الخلقية، وثانوية بسبب إجراء جراحة للحبل الشوكي. ويعطينا الفهم التفصيلي الجيني لمتلازمة الحبل الشوكي المعلق القدرة على التشخيص والعلاج الأمثل لهذه الظاهرة، ويعتمد مبدأ التدخل الجراحي لمناجزة متلازمة الحبل الشوكي المعلق في الأساس على تحرير هذا التعلق بالقناة الشوكية مما يعيد الحركة التشريحية للحبل الشوكي داخل القناة الشوكية دون تقييد مما يحفظ الإمداد الدموي و الأيض للحبل الشوكي والأعصاب، فيحفظ الوظيفة الحيوية المنوطة بهذه الأعصاب. ويعتبر استخدام الميكروسكوب وأجهزة مراقبة الأعصاب أثناء إجراء التدخل الجراحي وكذلك الغلق المحكم للأغشية السحائية عوامل مهمة تؤثر بشكل كبير في النتيجة الجراحية لهذه المتلازمة.

الهدف من البحث: تقييم التطور الكلينيكي للمرضى الذين أجريت لهم عمليات جراحية لمناجزة الحبل الشوكي المعلق بمستشفيات جامعة الأزهر بالقاهرة.

المرضي وطرق البحث: اجريت الدراسة على 25 مريضا بمستشفيات جامعة الأزهر تم ادخالهم المستشفى لاجراء جراحات لمناجزة متلازمة

الحبل الشوكى المعلق، وقد اجريت الدراسة فى الفترة ما بين فبراير 2017 الى ديسمبر 2020.

نتائج البحث: أظهرت النتائج تحسن آلام المرضى بنسبة 86.7%، وتحسن فى وظائف الحركة بنسبة 66.7%، كما تحسنت القدرة على التحكم فى البول والبراز بنسبة 50%. وكانت نسبة المضاعفات 12%.

الاستنتاج: المرضى الذين يعانون من آلام أسفل الظهر والام وضعف بالطرفين السفليين أو ضعف فى التحكم فى البول أو البراز يجب تقييم حالتهم بالفحوصات اللازمة واذا تبين أنهم يعانون من متلازمة الحبل الشوكى العلق فيفضل التدخل الجراحى دون تباطوء حتى لا تتدهور حالتهم، خاصة أن التحسن بعد الجراحة فى الحركة والتحكم فى البول لا يكون بشكل كامل.

الكلمات الدالة: الحبل الشوكى المعلق، التقييم بعد جراحات مناجزة الحبل الشوكى المعلق، تضم الخيط الإنتهائى الجافوى، المراقبة الفيسيولوجية للأعصاب أثناء الجراحة.