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Technical Note

Intraoperative demonstration of reduced distal spinal cord stiffness following untethering of the spinal cord using ultrasound shear wave elastography (SWE)

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1. Introduction

Tethered cord syndrome (TCS) was initially described in 1976 by Hoffman and colleagues after investigating a group of children who improved neurologically following spinal cord release by dividing a thick Filum Terminal.[1] TCS can be considered when the lower end of the spinal cord (LESC) level is low-laying or below normal (below L1-2 disc space level or L2 vertebral body) and if the filum terminal thickness is over 2 mm. TCS may be congenital (or primary) as a result of a caudal neuralization defect, which promotes various spinal dysraphism entities such as myelomeningocele and thickened filum terminal. Secondary (or acquired) TCS is thought to affect adults due to a disease process like traumatic injuries, tumors, infections, or previous childhood surgery.[6,7]

The classification systems in the literature on TCS are numerous, heterogeneous, and conflicting.[8] TCS could be associated with multiple neurological, urological, gastrointestinal, and orthopedic signs and symptoms.[2] Presumably, symptoms associated with TCS were attributed to traction and increased tension on the spinal cord and conus medullaris, which affected its function.[1,4] According to some, even a spinal cord with a “normal” level and appearance on MRI may require untethering due to filum terminal histologic properties that cause elasticity reduction and, eventually, tethering.[3] However, little knowledge is available on the elasticity of LESC. The current paper aims at evaluating the elasticity of the LESC using intraoperative shear wave elastography (SWE) ultrasound before and after spinal cord untethering. This may improve our understanding of the disease process’s effect on the distal LESC and may have future implications for TCS diagnosis and management.

2. Details of the case and technique illustration

2.1. Clinical and imaging data

A 45-year-old male presented with progressive bilateral lower limb radicular pain, right-sided foot drop, intermittent sphincter dysfunction, and occasional back pain for one year. Physical examination showed a lumbar spine area para-midline skin dimple, lower limb muscle wasting, weakness of the right lower limb (ankle dorsiflexion 3/5, big toe extension 4/5, plantar flexion 4/5), a positive straight leg raising test, intact sensations, and intact reflexes.

Magnetic resonance imaging (MRI) and computed tomography (CT) of the lumbar spine showed high-grade spondylolisthesis of L5/S1, bilateral L5 pars interarticularis defects, and significant wedging of the left side of the L5 vertebral body and the right side of S1. In addition, imaging showed a low-lying spinal cord, terminating at S1 level (tethered spinal cord), associated with an intraspinal lipoma opposite S3 and S4 levels (measured at 3 cm in the craniocaudal dimension), and a split cord malformation at L3 level with no associated bony bar. A bony defect involving the posterior elements of the lower lumbar and sacral regions was also observed, with a dorsal dermal sinus extending from the skin surface to the thecal sac at the L3–4 levels (Fig. 1).

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Abbreviations

shear wave elastography (SWE)
tethered cord syndrome (TCS)
lower end of the spinal cord (LESC)

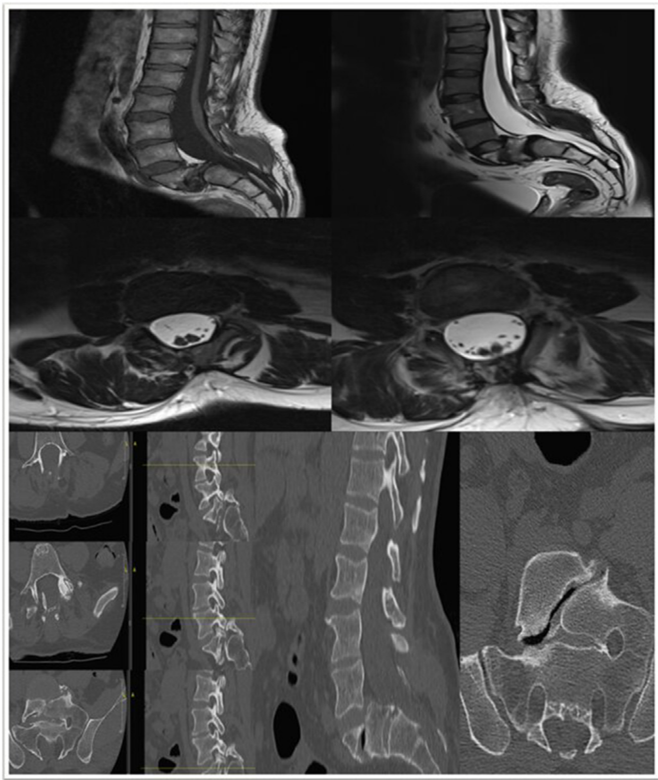

Fig. 1. CT/MRI lumbosacral spine. Shows high-grade spondylolisthesis of L5/S1, bilateral pars interarticularis defects at L5, significant wedging of the left side of L5 vertebral body and right side of S1, low lying spinal cord terminating at S1 level (tethered spinal cord) associated with intraspinal lipoma opposite S3 and S4 levels measuring 3 cm in craniocaudal dimension, split cord malformation at L3 level with no associated bony bar, bony defect of the posterior elements of the lower lumbar and sacral region with a dorsal dermal sinus extending from the skin surface to the thecal sac at L3-4 level.

2.2. Material and methods-Surgical intervention and intraoperative ultrasound evaluation

The surgical intervention included lumbar laminectomy (L 3–5), excision of a dermal sinus tract, and untethering of the spinal cord with the aid of neurophysiology monitoring. Following bone removal and before untethering the spinal cord, intraoperative ultrasound was performed. We used both B-mode and SWE provided by the SuperSonic Aixplorer system (SuperSonic Imagine SA) with a linear transducer array (2–15 MHz, SuperLinear 15–4) and an IOUS frequency of 2 MHz. Details of the technique were similar to previously published literature.[11,12] The field was carefully irrigated to avoid air bubbles that could interfere with the ultrasound examination. The evaluation started with a B-mode ultrasound, followed by SWE. Random regional points of interest (ROI; 2 mm) within the spinal cord were selected, and the average was calculated. This was compared before and after the untethering of the spinal cord. Before untethering, the mean elasticity of the distal end of the spinal cord (SCE) was 20–24 kPa (Fig. 2).

The dura was then freed from surrounding adhesions. The fistula connection was disconnected. The dura and arachnoid were opened at the midline, and the fistula was found to be adherent to the cord. In addition, multiple thick fibrous bands and extensive arachnoid adhesions were noted, tethering the cord to the inner surface of the dura (Fig. 3).

The fistula and the filum were stimulated up to 10 mA without a CMAP response. Also, the right L5 CMAP was seen at 1.8 mA (positive for nerve root attachment to the filum). All the bands were safely released. The dural opening was extended in a craniocaudal fashion to confirm no further adhesions. Following anatomic confirmation of the detethering process, SCE evaluation was repeated (Fig. 4), and it showed a significant drop in the mean SCE (<11 kPa at L4 level), indicating a softer spinal cord and, possibly, lower tension following untethering.

Intraoperative neurophysiology assessment showed no changes in SSEP, EMG, or MEP compared to baseline except for moderate transient tibialis anterior improvement. It was decided that spondylolisthesis would be treated in a second stage of surgery, if necessary. The dura was closed watertight. The fistula connection was circumferentially dissected and excised completely to the skin. Postoperatively, right lower limb ankle dorsiflexion, and big toe extension weakness improved, and the radicular pain disappeared.

3. Discussion

The pathophysiology of TCS is complex. Tethering causes tension, a reduction/cessation of blood flow, and, as a result, neurological dysfunction/deficit. In the pediatric age group, the filum is usually under higher tension (down stretched) compared to adults. Accordingly, adults usually have delayed presentations.[5] Pang et al categorized the pathophysiology and precipitating factors into three mechanisms, including transitory movement/stretching of the spinal cord, spinal trauma, and progressive tightening of the spinal canal.[5]

Two patterns of symptomatology models have been postulated. The threshold model suggests that in the presence of below-threshold tension, any further movement, lengthening, or other provoking factors/events will surpass the threshold level. Crossing the threshold level will affect blood flow and cause neurological dysfunction to develop. On the other hand, in repetitive (cumulative) theory, the symptomatology is slow and progressive, appearing after long years of tethering process progression that worsens tension in addition to the repetitive daily movements that ultimately lead to micro-ischemic insults over time.[5] In our illustrated case, in the presence of tethering processes including dermal sinus, lipoma, extensive adhesions, and a low-lying spinal cord, there are multiple provoking factors such as lumbar spondylosis and spondylolisthesis.

The mainstay of management of symptomatic TCS is aggressive and safe detethering of the spinal cord from all its attachments to reduce or eliminate tension.[4,6] Thereafter, it precludes further damage, alleviates pain, and improves neurological function.[1] Early detection of the condition promotes complete surgical detethering by disconnecting the filum and is subsequently associated with a favorable prognosis in symptomatic adults with TCS.[14] Moreover, the TCS type largely affects the outcome. For instance, lipoma type and adhesion type are generally associated with lower efficacy rates compared to the other types due to the difficulty associated with releasing the adherent nerves and a subsequently higher rate of re-tethering, recurrence of symptoms, and reoperations.[8] Re-tethering is generally common in the pediatric age group and remarkably low among adults.[910]

Intraoperative ultrasound elastography is an ultrasonic-based imaging modality that is a considerably new and evolving technology.[12] It analyzes the inherent biomechanical characteristics of tissue as well as changes caused by various pathologies.[13,14] While strain or pressure ultrasound elastography is qualitative, shear wave elastography (SWE) is a quantitative method to assess tissue elasticity.[13] In SWE, a force impulse from an acoustic radiofrequency wind is generated, and a secondary shear wave propagates transversely and perpendicularly. The shear wave speed (SWS) is used to calculate the wave spread over the surrounding

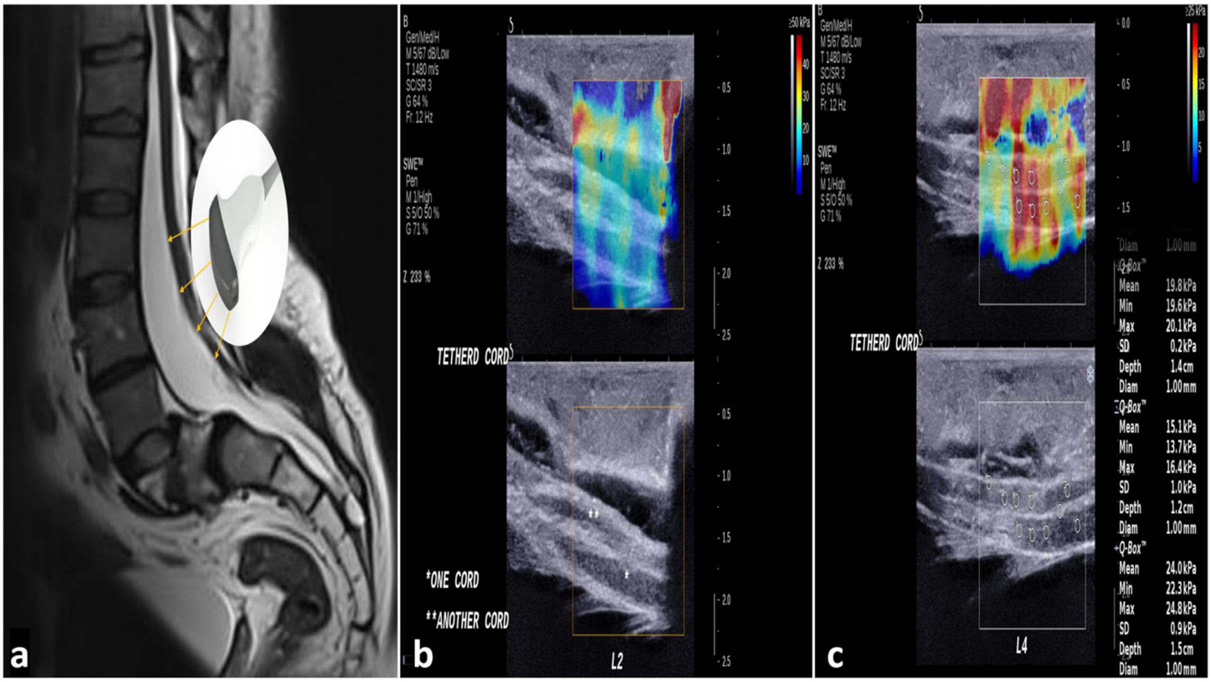

Fig. 2. IOUS/SWE pre-detethering of the spinal cord. Spinal cord sagittal view ultrasound image of lumbar spinal levels (L2 and L4). (**a**) The area of interest is defined by an ultrasound probe positioned posteriorly epidurally on a lumbosacral sagittal spine MRI. The lower half of both views (**b** and **c**) represents the B mode image, while the upper half represents the superimposed elasticity map. The selected region of interest (ROI) is represented by a Q-Box. Each Q-Box represents 2 mm. Each Q-Box is labeled with a particular symbol. The details of each Q-Box are seen on the right side. A color scale is posted in the right upper corner of each level. All the numbers are represented in kilopascals (kPa). There was an elevated SCE (image b) indicating increased spinal cord stiffness and tension inflicted by the tethering processes. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

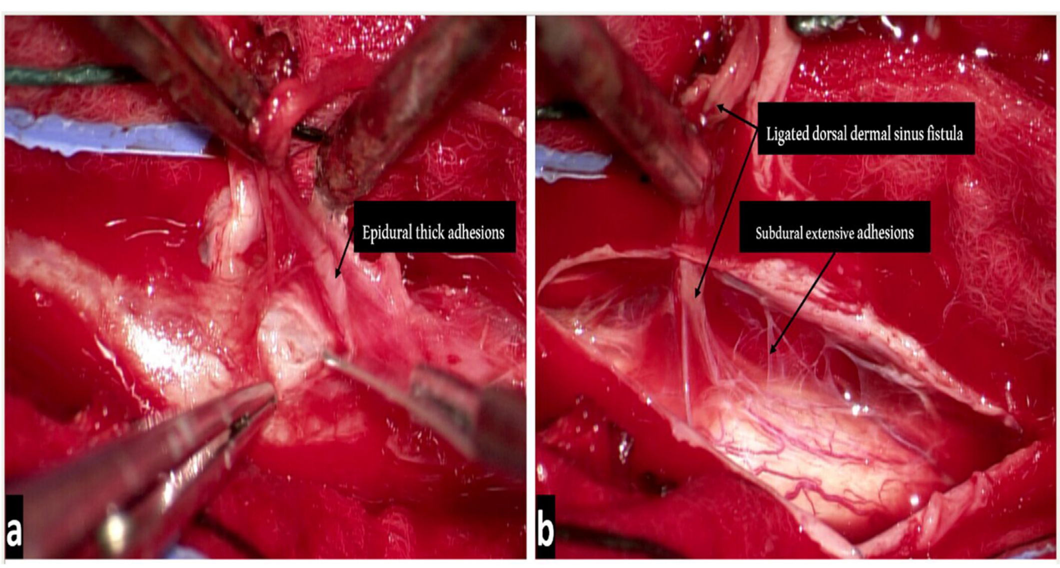

Fig. 3. Intraoperative. a An extensive epidural thickening and adhesions along with a disconnected dermal sinus fistula. **b** dermal sinus extending to the cord and extensive subdural adhesions.

tissue, and Young's (elastic/shear) modulus or tissue elasticity (kPa) is measured.[13,14] Subsequently, a real-time intraoperative US image and colored elasticity/stiffness map are acquired and superimposed on the B mode US image. The higher the kilopascal (kPa) values, the stiffer the tissue and the darker the red color becomes. In contrast, the lower the kilopascal (kPa) values, the softer the tissue, and the darker the blue color.[15]

There has been an evolutionary growth of ultrasound elastography imaging techniques' clinical applications over the years. An encouraging result for many organ pathologies, including but not limited to the liver, breast, thyroid, prostate, lymph nodes, and musculoskeletal system,[13,16] heart, lung, spleen, and uterus.[11] Central nervous system (CNS) sonography is still at its infancy level. The CNS clinical applications widened substantially over recent years.[17] The spinal cord (SC) applications are extremely limited when compared to the brain applications11. Spinal cord elasticity has been examined in a few cadaveric and animal studies. A single intraoperative human study investigated the effect of different compressive lesions on spinal cord elasticity (SCE) before and after

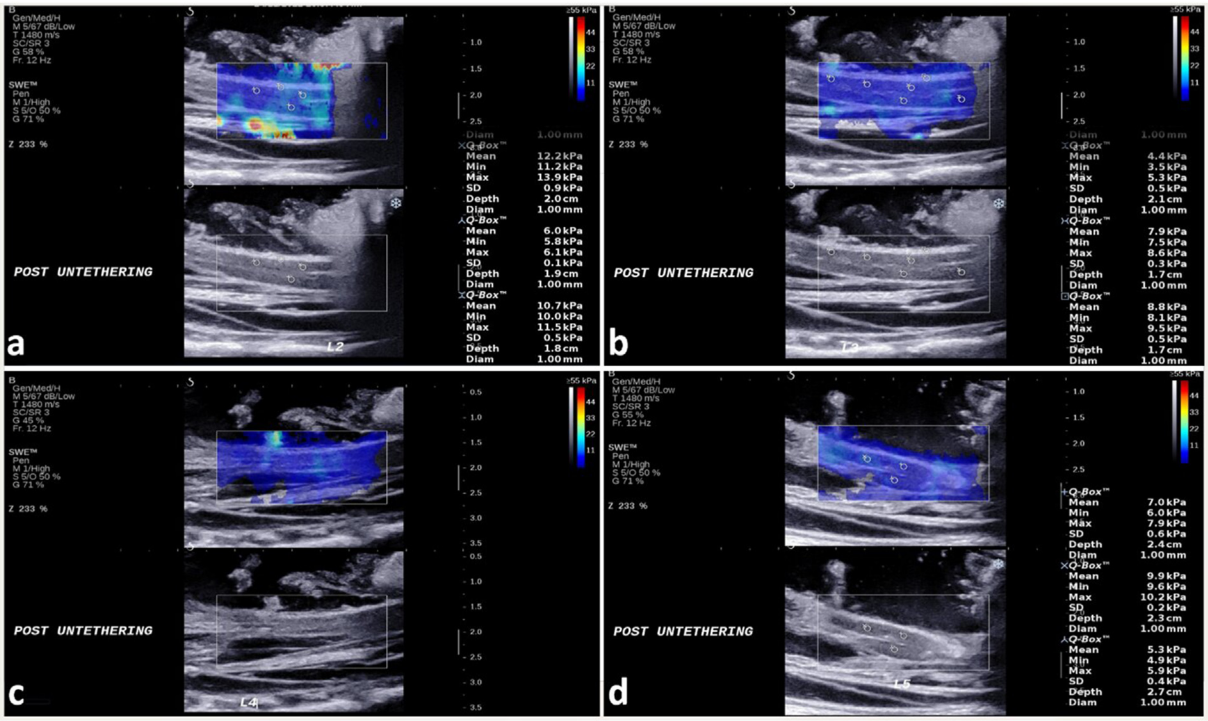

Fig. 4. IOUS/SWE Post-detethering of the spinal cord. Spinal cord sagittal view ultrasound image of lumbar spinal levels (L2–L5). At each level, the lower half represents the B mode image, and the upper half represents the B mode image overlapping with the elasticity map. **a** L2, **b** L3, **c** L4, and **d** L5. The selected region of interest (ROI) is represented by a Q-Box. Each Q-Box represents 1 mm. Each Q-Box is labeled with a particular symbol. The details of each Q-Box are seen on each level's right side of each picture. Color scales are posted in the right upper corner of each level. All numbers are represented in kilopascals (kPa). The picture collectively shows a significant drop in the SCE map to the least levels (dark blue) across all imaged spinal cord levels indicating a softer spinal cord and low tension in response to detethering. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

decompression. They found a significant drop in spinal cord elasticity after decompression from a median of 93.84 (75.27–121.75 kPa) to a median of 9.35 (6.95–11.22 kPa).11 The spinal cord elasticity value reported appears to be in line with the elasticity demonstrated in the current illustrated case (kPa < 11)). Huan Wee Chan et al reported a median of 14.9 kPa elasticity for the normal brain parenchyma which was considerably lower than that for all tumors (median 33.5 kPa).[14]

Nowadays, many promising and rapidly evolving applications for a variety of brain disorders including traumatic brain injury (TBI), brain tumors, ischemic stroke, Neonatal Hypoxic Ischemic Encephalopathy, epilepsy surgery, dementia, normal pressure hydrocephalus, Parkinson's disease, and multiple sclerosis.[14,17,18]

In the current case, reduced stiffness following untethering intraoperatively was a useful marker of a satisfactory untethering process. Moreover, in the presence of a multi-tethering process triggering the patient's symptoms including adhesions, dermal sinus, low-laying filum terminal, and spondylolisthesis, the use of ultrasound SWE established the point in time by which the spinal cord became completely untethered without the need to further address other tethering factors. In addition, the impact of each tethering factor on the spinal cord may be estimated with repetitive SWE scanning.

Conclusion: The elasticity of the tethered spinal cord was never clearly demonstrated. To our knowledge, this is the first demonstration of reduced stiffness (elasticity value) of the LESC before and following untethering using intraoperative SWE ultrasound. Intraoperative use of ultrasound shear wave elastography (SWE) for tethered spinal cord cases has academic value and encouraging practical implications. Further research is needed to evaluate the elasticity of the spinal cord in TSC cases and correlate it with clinical findings.

CRediT authorship contribution statement

Fawaz S. Almotairi: Conceptualization, Methodology, Software, Validation, Visualization, Writing – review & editing. **Ali A. Basalamah:** Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Validation, Visualization, Writing – original draft, Writing – review & editing. **Amr Amir:** Data curation, Investigation, Resources, Software, Validation, Visualization. **Amro F. Al-Habib:** Conceptualization, Data curation, Methodology, Software, Supervision, Validation, Visualization, Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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