



## Case Report

# Type I split cord malformation and tethered cord syndrome in an adult patient: A case report and literature review

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## ABSTRACT

**Background:** In a split cord malformation (SCM), the spinal cord is divided longitudinally into two distinct hemicords that later rejoin. This can result in a tethered cord syndrome (TCS). Rarely, TCS secondary to SCM presents in adulthood. Here, we present an adult female with Type I SCM resulting in TCS and a review of literature.

**Case Description:** A 57-year-old female with a history of spina bifida occulta presented with a 2-year history of worsening back and left leg pain, difficulty with ambulation, and intermittent urinary incontinence; she had not responded to conservative therapy. Magnetic resonance imaging (MRI) revealed a tethered cord secondary to lumbar type I SCM. The patient underwent an L1-S1 laminectomy for resection of the bony septum with cord detethering. At 2-month follow-up, the patient had improvement in her motor symptoms and less pain. In literature, 25 cases of adult-onset surgically managed SCM with TCS were identified (between 1936 and 2018). Patients averaged 37 years of age at the time of diagnosis, and 56% were female.

**Conclusion:** TCS can present secondary to SCM in adulthood and is characterized predominantly by back and leg pain.

**Keywords:** Adult, Diastematomyelia, Split cord, Tethered cord

## INTRODUCTION

With routine use of folic acid supplementation, spinal dysraphism is rare in Western countries.<sup>[21]</sup> Split cord malformation (SCM), or diastematomyelia, describes a spinal cord divided longitudinally into two distinct hemicords that later rejoin. Symptoms typically reflect traction on the cord and stretch-induced ischemia.<sup>[1-7,9-11,13-15,18,20-28,30,32-35]</sup> TC in adults presents somewhat differently than in children, with predominant symptoms that include pain (81.6%), weakness (72.3%), and sensory disturbances (78.7%).<sup>[2]</sup> There are often cutaneous stigmata, foot deformity, or scoliosis not previously diagnosed in childhood. SCM has been categorized into two types: Type I SCM consists of two hemicords, each contained within its own dural tube and separated by a dura-sheathed rigid osseocartilaginous median septum [Figure 1].

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Type II SCM consists of two hemicords housed in a single dural tube separated by a nonrigid, fibrous median septum.<sup>[8]</sup> Here, we present a 57-year-old female with Type I SCM and TC who underwent bony septum resection, and detethering with an adequate outcome, while also reviewing literature.

## CASE REPORT

A 57-year-old female with a history of spina bifida occulta and a left-sided clubfoot presented at age 56 with a 2-year history of worsening back and left leg pain, difficulty with ambulation, and intermittent urinary incontinence. On neurologic examination, she had 4+/5 strength in her LLE (iliopsoas, knee extension, knee flexion, dorsiflexion, and plantar flexion) with decreased distal left lower extremity strength (everters absent/loss EHL and decreased sensation). Deep tendon reflexes were 2+ throughout, and she had Babinski sign present on the left. MRI revealed a TC with SCM starting in the upper lumbar spine, becoming two separate dural sacs with a bony septum, and reconstituting into one dural sac/grouping of nerve roots ending in the sacrum [Figure 2]. The patient underwent an L1–S1 laminectomy utilizing neuromonitoring. The bony septum was excised, with spinal cord detethering/transsection of the filum terminale. The patient was discharged 5 days after surgery to an acute rehabilitation hospital. 5 months postoperatively, she had significant improvement in dorsiflexion and plantar flexion, minimal back pain, no leg pain, and sphincter incontinence.

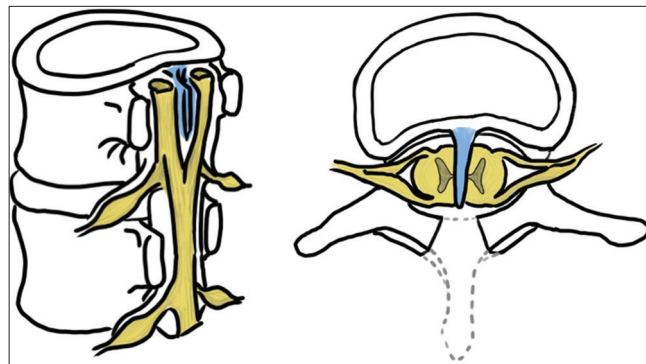
## Operative procedure

Under total intravenous anesthesia, with electromyograph (EMG) and somatosensory evoked potential (SEP) monitoring, the patient underwent an L1–S1 laminectomy. Under an operating microscope, the bony septum was removed followed by a durotomy caudal to the split in the thecal sac. No expansile duraplasty was performed. The split cord occurred well before the division in the thecal sac and so was under no tension or compression.

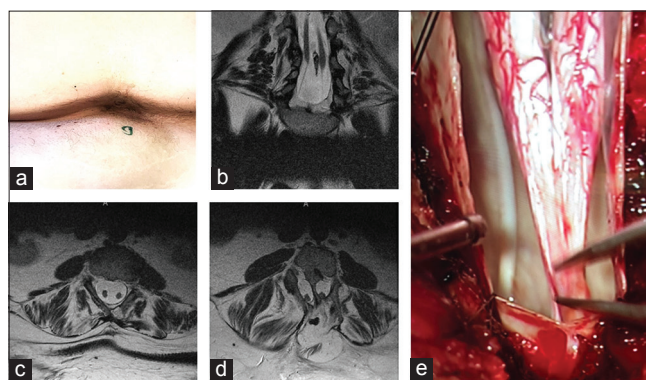
Examination of the cord revealed fat and ectopic ganglion cells and nerve roots extending to the filum. These were resected, and the filum was detached, detethering the cord. The durotomy was closed with a running suture and a Valsalva was performed to verify a watertight closure. Intraoperative monitoring revealed just transient but nonsignificant changes in EMG and SEP.

## DISCUSSION

A total of 24 case reports from 20 studies between 1936 and 2018 described SCM and TC diagnosed and surgically treated in adulthood. We included patients over the age of 18 years of age with symptomatic TC/SCM and who underwent surgical detethering. Preoperative status, surgical procedures, and outcomes



**Figure 1:** Type 1 diastematomyelia, with bony septum highlighted in blue and absent spinous process shown by dotted line. Illustration by Erin D'Agostino, not previously published, permission granted.



**Figure 2:** Split cord radiographic and operative images. (a) Preoperative photograph of an area of hypertrichosis over the lower lumbar spine. (b) Sagittal T2-weighted magnetic resonance imaging (MRI) showing the division of the spinal cord above the dural division and bony septum. (c) T2-weighted axial MRI showed the split cord within a single dural sac. (d) Split cord within divided thecal sac. (e) Intraoperative imaging showing the split cord with associated fat and ectopic ganglion cells from the intradural lesion.

are summarized [Table 1]. A more complete table reviewing the relevant literature was also generated [Table 2]. Surgical procedures included laminectomy, resection of a septum if present, and cord detethering. Patients were followed for an average of 1.61 years (standard deviation 1.77, range 0.03–7). For those who were initially symptomatic, postoperatively, 78.5% experienced reduced back pain (11/14), 69.2% reduced leg pain (9/13), 91.6% improved motor weakness (11/12), 80% improved sensation (9/11), and 66.7% improved bowel, bladder, or sexual dysfunction (4/6). Two of 19 patients with documented complications had a superficial infection and a cerebrospinal fluid leak.

Infrequently, tethered cord syndrome (TCS) secondary to SCM presents in adulthood.

SCM is reported to account for 10%–38% of adult TC diagnoses.<sup>[16,17,19,21,26,27,31]</sup> Two subsets of patients with SCM present in adulthood – those with SCM diagnosed in childhood who experience

**Table 1:** A summary of demographics, presenting signs and symptoms, and surgical outcomes for patients with SCM and TC diagnosed and surgically treated in adulthood.

Pre-operative clinical factors	Mean (SD)	n (%)	Range
Demographics			
Age at diagnosis (years)	37.24 (15.26)		18–73
Female		14/25 (56)	
Symptom duration (years)	2.62 (2.49)		0.08–9
Pre-operative signs and symptoms			
Leg pain		18/25 (72)	
Back pain		19/25 (76)	
Motor weakness		15/25 (60)	
Sensory dysfunction		15/25 (60)	
Bowel/bladder/sexual dysfunction		9/25 (36)	
Cutaneous stigmata		14/25 (56)	
Type I SCM		9/22 (40.9)	
Location of SCM			
Lumbar		16/20 (80)	
Thoracolumbar		1/20 (5)	
Thoracic		1/20 (5)	
Cervicothoracic		1/20 (5)	
Cervical		1/20 (5)	
Post-operative clinical outcome			
Follow-up (years)	1.68 (1.79)		0.03–7
Improved back pain		11/14 (78.5)	
Improved leg pain		9/13 (69.2)	
Improved motor weakness		11/12 (91.6)	
Improved sensation		9/11 (80)	
Improved bowel/bladder/sexual dysfunction		5/7 (71.4)	
Complications (infection, CSF leak)		2/19 (10.5)	

SCM: Split cord malformation, CSF: Cerebrospinal fluid

recrudescence symptoms and those who are first diagnosed with SCM in adulthood.<sup>[26]</sup> Our patient falls into this second category. In adulthood, presentation differs somewhat, with predominance of back and leg pain rather than with overt skeletal abnormalities or with progressive urological or orthopedic dysfunction.<sup>[12,15,26,32,34]</sup> Our patient presented primarily due to back and leg pain. In literature, 76% of patients present with back pain and 72% with leg pain. Our patient was also noted to have intermittent incontinence. In literature, 36% of patients experience bowel, bladder, or sexual dysfunction. Our patient presented with cutaneous stigmata, much like 56% of patients in literature. On imaging, our patient had a Type I SCM in the lumbar spine. In literature, 41% of patients with SCM present with Type I SCM and 80% of SCMs are found in the lumbar spine.

### Adult TC surgical results

In the adult population, the general consensus is that surgery is appropriate if neurologic deficits exist or symptoms are progressive.<sup>[21]</sup> In one review of 368 adult patients with TC

who were operated (67 of whom had SCM), 83% experienced improvement in pain symptoms, 43% improvement in sensory deficits, 59% improvement in motor deficits, and 46% improvement in incontinence.<sup>[2]</sup> Predictors of poor outcome in the review included lipomyelomeningocele, previous surgery, rapid motor deterioration before operation, long delay to diagnosis, and, notably, SCM.<sup>[2]</sup> A smaller study of adults with TCS without SCM demonstrated similar improvements but with greater improvement in leg weakness.<sup>[12]</sup> One study found that of 24 patients with SCM and neurologic deficits who underwent surgery, 23 demonstrated neurologic improvement.<sup>[29]</sup>

### Complications of adult detethering procedures

Adult patients who undergo detethering appear to have higher complication rates than pediatric patients but lower rates of retethering,<sup>[17]</sup> with 16% of adult patients demonstrating retethering over 8 years compared to 52% of pediatric patients within 5 years.<sup>[16]</sup> In our review of adult-onset SCM with TCS,

**Table 2:** Literature review demonstrating presenting signs and symptoms, imaging findings, operative procedures, and follow-up status of patients with adult presentation of diastematomyelia with tethered cord syndrome who underwent operative intervention.

Case reports	Presenting signs and symptoms	Imaging findings	Surgical procedure	Clinical outcome
Current patient	A 57-year-old female with lumbar hypertrichosis and 2-year history of back and leg pain, intermittent incontinence, and sensory dysfunction	Upper lumbar Type I SCM with bony septum and sacral tethered cord	L1-S1 laminectomy, resection of bony septum, detethering. Complicated by superficial cellulitis	At 2-month follow-up improved back and leg pain, no incontinence
Viswanathan, 2018 <sup>[32]</sup>	A 67-year-old female with a 6-month history of back and radicular leg pain, paresthesia, neurogenic claudication	Fibrous lumbar Type I SCM, conus at L4, syrinx at T1	L2-L3 laminectomy, fibrous septum resection, detethering. No complications	At 5-year follow-up resolved leg pain, improved but moderate residual back pain
Akay, 2004 <sup>[11]</sup>	A 53-year-old female with lumbar hypertrichosis and a 5-year history of progressive back pain, leg weakness, and numbness	Type 1 SCM at L1-L2 with a midline osseous spur and conus at L3	L1-L2 laminectomies, cord detethering, resection of bony septum. No complications	At 1-year follow-up resolved leg pain, mild residual back pain
	A 28-year-old male with lumbar hypertrichosis and a 5-year history of radicular lower back pain and neurogenic detrusor	Type II SCM at L3, conus at L3	Untethering and filum terminale release. No complications	At 44-month follow-up reported as "better."
Wolf, 1987 <sup>[34]</sup>	A 23-year-old male with hypertrichosis and a 2-year history of radicular lower back pain and impotence	Myelomeningocele, spina bifida, Type II SCM at L3, conus at S2	Untethering and filum terminale release. No complications	At 39-month follow-up reported as "better."
	A 20-year-old male with hypertrichosis and >1 year of radicular lower back pain	L3 hemivertebra, Type II SCM at L3, conus at L4	Untethering and filum terminale release. No complications	At 17-month follow-up reported as "better."
	A 27-year-old male with Klippel-Feil syndrome and 1-year history (starting after fall off ladder) of spastic quadriplegia and sensory alteration	Klippel-Feil C2-C3, spina bifida C1-C3, meningocele C2, tethered cord at C4, Type II SCM C3-C4	Detethering. No complications	10 days after surgery weakness and sensation improved, spasticity unchanged
Kim, 2013 <sup>[20]</sup>	A 35-year-old female with a 2-month history of neck pain, back pain, numbness, and recurrent bilateral leg weakness	Type II SCM C7-T11, conus at L5	Laminectomy T8-T11, L5-S1 laminectomy for detethering. No complications	7 years after surgery normal examination with no paraparesis or back pain
Elmaci, 2001 <sup>[7]</sup>	A 42-year-old male with a 1-month history of leg pain, distal leg weakness, and neurogenic claudication	Intradural lumbar teratoma and Type I SCM at L5	Lumbar laminectomy, removal of bone spur and cystic mass, sectioning of terminal filum. No complications	At 1-year follow-up, claudication resolved with residual leg weakness
Sheehan, 2002 <sup>[30]</sup>	A 38-year-old female with a 7-month history of leg weakness and pain, distal paresthesia	Type I SCM at T1-T3, T2-4 intradural epidermoid cyst, and C6-7 syrinx, tethered cord	T1-3 laminectomies with resection of fibrous intradural septum, untethering, hemicord mass excised. No complications reported	At 9-month follow-up, improved weakness, sensation. Rare radicular pain
Lewandrowski, 2004 <sup>[22]</sup>	A 44-year-old female with a history of multiple remote spinal surgeries (including for scoliosis) presents with 4-year history (starting after car accident) of bilateral leg pain, unilateral weakness, and sphincteric dyssynergia	Type I SCM at L1-L3 with bony spur, conus at L5/S1, spina bifida occulta S1	Two procedures: Harrington rod partially removed T11-S1, bony spike resected. Detethering Complicated by CSF leak	At 1-year follow-up, complete resolution of leg symptoms. Mild residual lower back pain
Hamby, 1936 <sup>[14]</sup>	A 20-year-old female with cutaneous stigmata and scoliosis presents with a 9-year history of back pain and motor and sensory deficits	Not reported	SCM (unclear type), tethered cord, Neurenteric cyst/dermoid cyst/hamartoma/"intradural mass." No complications	Improvement of motor and sensory deficit. Unchanged pain

(Contd...)



**Table 2:** (Continued)

Case reports	Presenting signs and symptoms	Imaging findings	Surgical procedure	Clinical outcome
Cliffon, 1947 <sup>[5]</sup>	A 22-year-old male with hypertrichosis and a 6-month history of back and leg pain	Spina bifida occulta of sacrum. Type II SCM at L5-S1. Conus at L4	L4-L5 laminectomy, detethering. No complications	At 5-month follow-up resolved leg and back pain
Freeman, 1961 <sup>[9]</sup>	A 43-year-old female with leg length discrepancy and lumbar hypertrichosis who presented with a 3.5-year history of leg pain	Spina bifida L2-L5, Type II SCM with bony spicule at L2-L3	Septum resection, detethering. No complications	At 1-year follow-up improved pain with some residual sensory alteration
Fulcher, 1970 <sup>[10]</sup>	A 38-year-old female with a history of scoliosis presents with an 8-year history of leg and back pain, motor deficits, sensory deficits	SCM (unclear type), fibrous adhesions, tethered cord, spina bifida	Not reported	At follow-up reported to have worse pain, worse motor follow functioning, and worse bladder/bowel functioning
Garcia, 1980 <sup>[11]</sup>	A 37-year-old female with a 3-year history of pain, motor deficits, sensory deficits, and bowel/bladder dysfunction	Spina bifida, scoliosis, SCM (unclear type)	Not reported	Improvement in pain and bowel/bladder function. No improvement in weakness or sensation
Warder, 1993 <sup>[33]</sup>	A 53-year-old female with lumbosacral hemangioma, leg length discrepancy presents with spasticity and hyperreflexia, back and leg pain, and paresthesia	Bifid vertebrae, intradural lipoma, SCM (unclear type), fatty filum	Not reported	At follow-up resolved back pain and paresthesia. Spasticity, hyperreflexia, and leg pain unchanged
Quinones-Hinojosa, 2004 <sup>[28]</sup>	A 73-year-old female with childhood history of scoliosis and late ambulation milestones with a 2-year history of progressive difficulty walking, back pain, and weakness	Type II SCM T12-L3 with conus at L3	T12-L3 laminectomies, soft tissue septum resected, detethering. No complications	At 6-week follow-up decreased back pain, improved ambulation. Strength mildly improved
Aufschnaiter, 2008 <sup>[2]</sup>	A 52-year-old female with sacral dermal sinus and a 3-year history of progressive back and bilateral leg pain, weakness, and urinary incontinence	Incomplete spina bifida S2-S4. Type II SCM and syringomyelia at L2 with tethered cord. Arachnoid cyst S4-S5	Laminectomy S2-S4, detethering. No complications	At 28-month follow-up, urinary and sensorimotor deficits resolved. Moderate pain continues
Kaminker, 2000 <sup>[18]</sup>	A 38-year-old male with bilateral forefoot cavus deformity and 1.5-year history of bilateral leg pain and neurogenic claudication	Multilevel occult spina bifida L2-coccyx. Type I SCM L2-L3/L4 with bony septum. Conus tethered at L4	Laminectomy L2-L3, septum resected, detethering. No complications	At 2-year follow-up, complete resolution of leg symptoms with mild residual lower back pain
Chehrizi, 1985 <sup>[4]</sup>	A 32-year-old male with lumbar hypertrichosis and a 3-year history of bladder fullness, sexual dysfunction, constipation, and reduced sensation	Sacral spina bifida, Type II SCM at L1, conus at L2/L3	L3-TII laminectomies, detethering. No complications	At 1-year follow-up, 10-15% improvement in bladder and sexual follow-up function. Almost normal sensation. Significant bowel function improvement
Duz, 2008 <sup>[6]</sup>	A 21-year-old male with hypertrichosis presenting with altered sensation and back pain	Type II SCM, conus at L5, dermal sinus	Detethering, resection of dermal sinus and fibrous septum. No complications reported	At approximately 10-month follow-up, no improvement - continued back pain
	A 21-year-old male with urgency, sexual dysfunction, weakness, altered sensation, back pain, and leg pain	Type II SCM, conus at L4-5	Detethering, resection of fibrous septum. No complications reported	At approximately 10-month follow-up, no back and leg pain, no weakness, improved bladder dysfunction

(Contd...)

**Table 2:** (Continued)

Case reports	Presenting signs and symptoms	Imaging findings	Surgical procedure	Clinical outcome
Maiti, 2010 <sup>[24]</sup>	An 18-year-old male with lumbar hypertrichosis and a 1-year history of back pain, urinary symptoms, bilateral leg paresthesia	Type II SCM at L1, conus at L4. Heterogeneous lesion (teratoma) L2-L3. Syring proximal to SCM	L1-L3 laminectomy, partial decompression of lesion, excision of fibrous band, detethering. No complications	At 6-month follow-up, paresthesia improved, no motor deficits. Bladder symptoms persist
Maroun, 1982 <sup>[25]</sup>	A 29-year-old female with lumbar hypertrichosis and leg length discrepancy with several week history of worsening back and leg pain and hypalgesia	Spina bifida L3-sacrum, Type I SCM with bony and cartilaginous spur L3-L4. Conus at L5-S1	Two procedures: Laminectomy with spur removal. Detethering with cyst removal. No complications	At 7-month follow-up, no clinical symptoms

SCM: Split cord malformation, CSF: Cerebrospinal fluid

surgery – which comprised laminectomy, resection of a septum if present, and detethering with sectioning of the filum terminale – resulted in improved weakness (92%), sensory dysfunction (80%), back pain (79%), leg pain (69%), and bladder, bowel, or sexual dysfunction (67%). Our patient underwent an uncomplicated L1–S1 laminectomy for the resection of bony septum and cord detethering, and at 2-month and 5-month follow-up reported improved motor symptoms and reduced pain.

## CONCLUSION

TCS secondary to SCM rarely presents in adulthood. The adult presentation is primarily characterized by back and leg pain. Trauma on a macro/micro scale may result in increased traction on the cord, and progressive vascular compromise resulting in the symptom onset in adulthood. Surgical treatment is appropriate and effective when neurological symptoms are present.

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## Conflicts of interest

There are no conflicts of interest.

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