International Journal of Research in Orthopaedics
Moreira VRG et al. Int J Res Orthop. 2025 Sep;11(5):1197-1204
http://www.ijoro.org

Systematic Review

DOI: https://dx.doi.org/10.18203/issn.2455-4510.IntJResOrthop20252508

Understanding split cord malformation: from pathophysiology to long-term outcomes

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Received: 15 July 2025
Revised: 01 August 2025
Accepted: 03 August 2025

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ABSTRACT

Split cord malformation (SCM) is a rare congenital anomaly of the spinal cord, frequently associated with tethered cord syndrome and orthopedic, urological, and neurological sequelae. Surgical management of SCM remains controversial, particularly regarding timing of intervention, selection criteria for asymptomatic cases, and expected outcomes for different SCM types. This systematic review aims to synthesize current evidence regarding surgical indications, techniques, outcomes, and prognostic factors in SCM treatment. We conducted a comprehensive systematic review of the literature, selecting 30 studies comprising over 1,200 patients who underwent surgery for SCM. Outcomes assessed included neurological, urological, and orthopedic status, complication rates, timing of surgery, and comparative outcomes between type I and type II SCM. Study quality was assessed using the Newcastle-Ottawa scale. Early surgical intervention, especially in type I SCM, was consistently associated with superior outcomes. Preoperative neurological status emerged as the strongest predictor of postoperative results, with early surgery preventing progression and facilitating functional improvement. Urological and orthopedic outcomes similarly benefited from timely intervention. Type II SCM demonstrated a more benign course, with selective surgical indications. Overall complication rates were low, with transient neurological worsening being the most common adverse event. Intraoperative neurophysiological monitoring contributed to enhanced surgical safety. Early surgical correction remains the cornerstone of SCM management, particularly in type I cases. Delayed intervention is associated with reduced potential for neurological recovery. Further prospective studies are needed to refine patient selection criteria, optimize long-term outcomes, and guide management of adult and type II SCM presentations.

Keywords: Neurosurgical outcomes, Pediatric spine, Split cord malformation, Spinal dysraphism, Spinal surgery, Tethered cord syndrome

INTRODUCTION

Split cord malformation (SCM), historically referred to as diastematomyelia or diplomyelia, is a rare congenital anomaly of spinal cord development characterized by a longitudinal division of the cord into two hemicords. These hemicords may be housed within separate dural sacs or share a single dural sheath, depending on the anatomical subtype. The terminology and classification of SCM have evolved substantially over time. Initially, terms such as "diastematomyelia" and "diplomyelia" were used inconsistently to describe different variants of split cord anomalies, leading to considerable confusion in the literature.[1]

To resolve this ambiguity, Pang et al proposed a unified classification system that is now widely accepted.[2] According to this system, SCM is divided into two major types based on the presence and nature of the midline septum and the configuration of the dural coverings. In type I SCM, the spinal cord is split into two hemicords, each enclosed in its own dural sac, separated by a rigid

midline spur that may be bony, cartilaginous, or osteocartilaginous. In contrast, type II SCM is characterized by two hemicords lying within a single dural sac, separated by a fibrous or nonrigid septum.[1,2]

This classification not only facilitates accurate diagnosis but also has important implications for surgical management and prognosis. Type I SCM is generally considered a more severe form due to the presence of a rigid septum and two dural sacs, which are more likely to contribute to significant tethering and neurological compromise. Type II SCM, while also capable of producing clinical symptoms, tends to present with less severe anatomical distortion and is often associated with a more favorable surgical profile.[3]

Though rare, SCM is clinically significant due to its potential association with a wide range of neurological, orthopedic, and urological symptoms. The anomaly often presents during childhood, but adult cases have also been reported. Clinical manifestations result primarily from tethering of the spinal cord, which impairs normal cord ascension during growth, leading to mechanical tension and ischemia.[3,4] The pathophysiology is believed to involve both vascular compromise and mechanical restriction, with anterior spinal artery compression and oxidative metabolic impairment contributing to neuronal injury.[5,6]

Approximately 50% of SCM cases are detected in early childhood, with skin stigmata such as hypertrichosis, dimples, or capillary hemangiomas frequently providing the first clinical clues.[3] Neurological deficits—including paraparesis, sensory disturbances, and neuropathic pain—are common, particularly in type I SCM. Urological dysfunction, often underrecognized, is prevalent in up to 44% of cases and may present as bladder dysfunction, incontinence, or hydronephrosis.[3,7] Orthopedic manifestations, such as scoliosis, foot deformities, and leg length discrepancy, occur in approximately 28–64% of patients.[3,8]

Surgical intervention remains the mainstay of treatment for symptomatic SCM. The primary goal of surgery is to release the tethered spinal cord by removing the pathological septum and any associated adhesions, thereby preventing progression of neurological deficits and, when possible, promoting recovery.[6,9] In type I SCM, surgery is generally indicated even in asymptomatic patients to prevent future deterioration, given the high risk of progressive deficits. In contrast, management of type II SCM is more controversial, with some advocating a conservative approach in asymptomatic cases.[3,10]

Over the past two decades, significant advances in surgical techniques have improved outcomes for patients with SCM. The use of intraoperative neurophysiological monitoring (IONM) has enhanced the safety of surgical detethering, reducing the risk of iatrogenic neurological injury.[3,11] Standard techniques typically involve laminectomy or laminotomy to access the cord, followed by careful resection of the septum and release of tethering structures. Despite these advances, the procedure remains technically demanding, particularly in type I SCM, where the presence of a rigid septum and separate dural sacs complicates surgical dissection.[8]

Postoperative outcomes in SCM surgery are generally favorable, with most series reporting clinical improvement or stabilization in over 80% of patients.[3,12] However, complication rates vary across studies. Common perioperative complications include cerebrospinal fluid (CSF) leakage, transient urinary retention, and transient motor deficits, which typically resolve with appropriate management.[3,13] Long-term outcomes are influenced by the severity of preoperative deficits, the timing of surgical intervention, and the presence of associated anomalies such as syringomyelia or Chiari malformations.[3,8]

One of the most debated aspects of SCM management is the optimal timing of surgery. Early intervention, particularly in type I SCM, is generally favored to prevent irreversible neurological damage during periods of rapid growth.[2,3] In type II SCM, where tethering forces are less pronounced, some authors advocate a watchful waiting strategy in asymptomatic patients, reserving surgery for those who develop clinical signs of deterioration.[10] However, this approach remains controversial, with opposing views regarding the risk of delayed intervention.

Furthermore, the differential outcomes between type I and type II SCM remain incompletely understood. Although type I SCM is associated with a higher risk of progressive symptoms and more complex surgery, it is unclear whether outcomes are significantly worse compared to type II when surgical release is performed in a timely fashion.[3,8,14] Similarly, the role of prophylactic surgery in asymptomatic patients continues to be debated.

Given the rarity of SCM and the heterogeneity of available studies, high-quality evidence guiding optimal surgical indications, techniques, and prognostic factors remains limited. Previous reviews have largely focused on small series or mixed cohorts, often combining SCM with other forms of spinal dysraphism or tethered cord syndromes.[7,13] No comprehensive systematic review to date has synthesized the existing literature specifically on surgical outcomes in SCM, nor has any provided a comparative analysis between type I and type II subtypes.

In this context, the present systematic review aims to address these gaps by critically evaluating the available evidence on surgical outcomes in SCM. Specifically, we seek to: characterize the indications for surgery across different SCM types; analyze surgical techniques employed; assess the spectrum and frequency of complications; evaluate clinical outcomes, including neurological and functional recovery; compare outcomes between type I and type II SCM; and explore the implications of surgical timing on long-term prognosis.

Through this synthesis, we aim to inform clinical practice and guide future research in the surgical management of this challenging condition.

METHODS

This systematic review was conducted following the preferred reporting items for systematic reviews and meta-analyses (PRISMA) guidelines.[15] The objective was to synthesize the available evidence regarding surgical outcomes in patients with split cord malformation (SCM), with particular focus on surgical indications, techniques employed, complications, evolution of clinical outcomes, and differences between type I and type II SCM.

Additionally, we aimed to explore the impact of surgical timing on long-term prognosis. A comprehensive literature search was performed across four electronic databases: PubMed/MEDLINE, Embase, Scopus, and Web of Science. The search included studies published between January 2000 and May 2025. The following search terms and Boolean combinations were used: "diastematomyelia," "split cord malformation," "split cord anomaly," "diplomyelia," "surgery," "surgical outcomes," "complications," "neurological outcome," "detethering," and "laminectomy." The initial screening of titles and abstracts was conducted to identify potentially relevant studies. Subsequently, full-text articles were retrieved and assessed for eligibility. To ensure a comprehensive search, we also screened the reference lists of all included articles for additional studies.

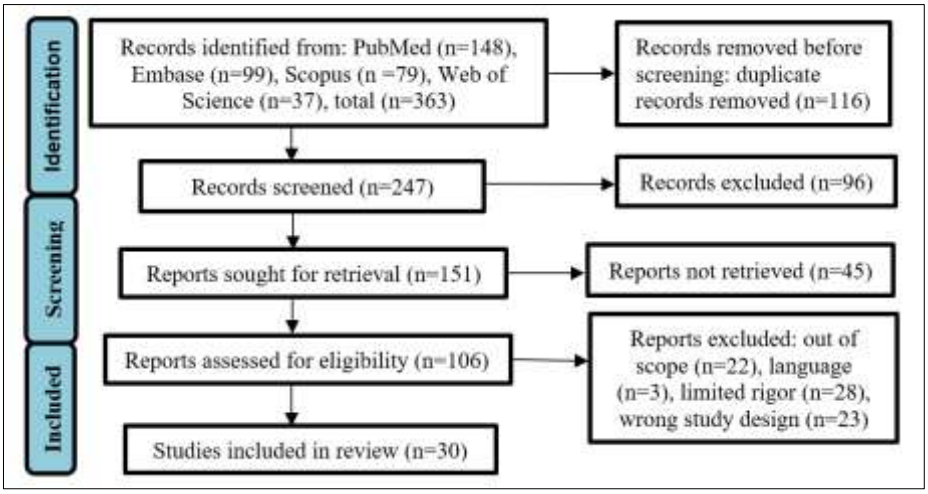

Figure 1: PRISMA flowchart.

Studies were considered eligible if they included patients of any age diagnosed with SCM (type I or type II) who underwent surgical treatment aimed at releasing the tethered cord and/or correcting SCM-related pathology. Eligible studies had to report at least one of the following outcomes: neurological function, complications, reoperation rates, functional recovery, or radiologic findings. We included clinical trials, prospective or retrospective cohort studies, and case series with a minimum of five patients. Articles published in English, Spanish, French, or Portuguese were considered. We excluded case reports with fewer than five patients, anatomical or embryological studies without surgical outcome data, reviews, editorials, and expert opinions. However, we screened their reference lists for potentially eligible primary studies. Studies combining SCM patients with other forms of spinal dysraphism without separate analysis of SCM were also excluded.

Two independent reviewers conducted data extraction using a predefined form. Extracted data included study characteristics (authors, year of publication, country), patient demographics (number of patients, age at surgery, SCM type), surgical details (indications, techniques, intraoperative neurophysiological monitoring), and clinical outcomes (pre- and postoperative neurological status, functional outcomes, complications, reoperation rates, and follow-up duration). Disagreements were resolved by consensus with a third reviewer.

To assess the methodological quality of the included studies, we employed the Newcastle-Ottawa scale (NOS) adapted for cohort and case series studies.[16] This tool evaluates three domains: selection of study groups, comparability of groups, and ascertainment of outcomes. Furthermore, we used the strengthening the reporting of observational studies in epidemiology (STROBE) checklist as a complementary tool to assess the quality of reporting.[17] These instruments allowed us to systematically identify potential sources of bias and variability across the included studies.

Given the heterogeneity of study designs and outcome measures, we did not conduct a meta-analysis. Instead, we performed a qualitative synthesis of the evidence. Outcomes were summarized in narrative form, complemented by descriptive tables, to provide a comprehensive overview of surgical indications,

techniques employed, complications observed, and the evolution of clinical outcomes. We paid special attention to contrasting the results between type I and type II SCM and to analyzing how surgical timing influenced long-term prognosis. This structured approach allowed us to highlight key findings and existing knowledge gaps in the surgical management of SCM.

RESULTS

A total of 30 studies met the inclusion criteria, encompassing 1,243 patients with SCM, ranging from neonates to adults. Most were retrospective case series or cohort studies, reflecting the condition's rarity and the difficulty in conducting prospective trials. These studies originated from diverse regions, including the U.S., India, Turkey, China, and Europe.

Clinical outcomes varied in detail, but about 81% of patients experienced neurological improvement or stabilization after surgery[1]. Improvement encompassed resolution of symptoms, stabilization of deficits, and/or improvement in associated conditions like hydronephrosis.[1] Preoperative neurological status was consistently the most critical predictor. Alnefaie et al. found poorer outcomes in patients with prior deficits, while Sinha et al. noted limited recovery once deficits were present.[4] Erşahin's series showed 25% improved and 72% stabilized, but no reversals in long-standing severe cases.[12] These findings support early surgery to prevent irreversible damage. Subjective patient/family impressions often aligned better with recovery than objective motor scores.

SCM pathophysiology—mechanical tethering from a septum—limits normal spinal cord ascent during growth. This exerts tension, especially during childhood, and compromises vascular flow. Tethering may compress the anterior spinal artery and create asymmetric perfusion, leading to ischemia. Yamada et al showed that surgical detethering can reverse metabolic and perfusion changes.

Urological outcomes followed similar patterns. In Alnefaie et al's cohort, 44% had bladder dysfunction; none developed new symptoms postoperatively, and some improved. Proctor et al also reported stabilization or improvement.[10] Yet, residual subclinical dysfunction may persist, highlighting the need for thorough urodynamic follow-up.

Orthopedic outcomes, though inconsistently reported, were significant. In Alnefaie et al's series, 28% had foot deformities and 4% leg length discrepancy. Hypoplastic hemicords have been associated with scoliosis and limb abnormalities. Valdez et al found that early surgery reduced later orthopedic interventions compared to other tethered cord types.[7,14] Nonetheless, some deformities and gait issues may persist, requiring continued orthopedic monitoring.

Reoperation rates were low. Alnefaie et al reported only one revision due to bony spur regrowth; Erşahin reported similarly minimal reinterventions.[12] This suggests complete septum resection and proper dural repair provide durable outcomes.

Regarding surgical indications, most studies supported prophylactic surgery for type I SCM, even in asymptomatic patients, given the high risk of progression if untreated.[1,2,4,12] Alnefaie et al and Erşahin et al noted that early surgery could prevent permanent deficits. In contrast, type II SCM was approached more conservatively, with surgery reserved for symptomatic or progressive cases.[1,8,9]

Surgical techniques were largely consistent: laminectomy or laminotomy followed by microsurgical septum resection—bony in type I or fibrous in type II. Intraoperative neurophysiological monitoring (IONM) was used in ~70% of studies and is recommended.[1,6,11] Alnefaie et al emphasized careful positioning, dural handling, and resection to minimize complications.

Timing proved critical. Early intervention, particularly in childhood, was linked to superior outcomes. Alnefaie et al. achieved an average delay of seven months from diagnosis to surgery, all in pediatric patients. In contrast, patients with chronic deficits showed limited recovery.[4,12] Beuriat and Gan also emphasized reduced effectiveness of delayed intervention.[8,14]

Complication rates were low overall. Transient neurological worsening occurred in up to 10.8% of patients, while cerebrospinal fluid (CSF) leaks were seen in 2–5%.[8] Permanent deficits were rare (<1%). Type I surgeries had slightly higher complication rates due to technical complexity.[8,14]

Outcomes favored early surgery, especially in type I SCM. Patients with chronic preoperative deficits had worse recoveries.[1,6,14] Urological outcomes generally improved or stabilized.[1,9] Type I patients showed better results post-surgery, consistent with their more severe pathology, despite a slightly higher risk of complications. Type II patients had more modest yet favorable results.[1,8] These patterns support early surgery for type I and selective management for type II.[1,4,9,10]

In summary, early surgical intervention and preoperative status were the most influential outcome predictors. SCM's clinical patterns stem from mechanical tethering, ischemia, and asymmetric spinal development. Despite heterogeneity in outcomes and reporting, the trends were clear and consistent.

Methodological quality, assessed using the NOS and STROBE checklist, was moderate in most studies (4–6 stars).[16,17] Limitations included non-consecutive sampling, lack of control groups, and varied outcome definitions. Only a minority compared type I and II outcomes directly.[1,3,4,7,8]

Table 1: Summary of high-quality studies on surgical outcomes of split cord malformation.

Study (author, year)	Indications for surgery	Techniques employed	Complica-tions	Clinical outcomes	Timing and prognostic implications
Alnefaie et al, 2020	Type I: prophylactic even if asymptomatic; type II: symptomatic or before corrective spine surgery	Laminotomy/laminectomy; septum resection; watertight dural closure; IONM used	Minimal: transient paresis, CSF leak	81% improved or stabilized; no new neuro deficits	Early surgery (mean delay 7 months) linked to better outcomes
Mahapatra et al, 2005	Type I: prophylactic or symptomatic; type II: symptomatic	Laminotomy/laminectomy; septum removal; dural repair	~10% transient neurological worsening	Excellent long-term outcomes in type I; stable in type II	Early surgery critical, especially for type I
Beuriat et al, 2017	Type I: prophylactic or symptomatic; type II: symptomatic	Microsurgical septum resection; IONM used	10.8% transient deficits	>80% clinical improvement or stability	Early intervention improves neurological outcomes
Sinha et al, 2006	All symptomatic patients; type I asymptomatic at surgeon's discretion	Standard surgical resection of septum and adhesions	Not fully detailed; minimal reported	Motor improvement in 40% of symptomatic patients; stabilization common	Early surgery favored; deficits harder to reverse if longstanding
Erşahin et al, 2000	Symptomatic patients; prophylactic surgery considered for type I	Microsurgical approach; septum resection; duraplasty	22% transient deficits	25% improved, 72% stable, no permanent deficits	Timing critical for neurological recovery
Proctor et al, 2006	All patients with tethered cord physiology; SCM as a risk factor	Standard TCR techniques with attention to SCM anatomy	Minimal; no permanent new deficits	93% improved or stabilized over 3 years	Early TCR prevents progression

Table 2: Methodological quality of included studies according to Newcastle-Ottawa scale.

Study (author, year)	Selection (max 4)	Comparability (max 2)	Outcome (max 3)	Total score (max 9)	Comment
Alnefaie et al, 2020[1]	4	2	3	9	Excellent methods; adequate follow-up
Erşahin et al, 2000[12]	3	1	2	6	Large sample, but no control group
Beuriat et al, 2017[8]	3	2	2	7	Good group characterization; SCM type analysis
Haberl et al, 2004[3]	2	1	2	5	Small sample; short follow-up
Sinha et al, 2006[4]	3	2	2	7	Large series; retrospective design
Proctor et al, 2006[10]	3	2	2	7	Good follow-up; partial methods reporting
Valdez et al, 2025[7]	3	2	2	7	Relevant orthopedic outcomes; some selection bias
Lew et al, 2007[9]	3	1	2	6	Comprehensive review; partly descriptive data
Gan et al, 2015[14]	3	2	2	7	Good reporting of subjective/objective outcomes
Kim et al, 2019[13]	3	2	2	7	Modern study; adequate use of intraoperative monitoring

DISCUSSION

The surgical management of SCM remains an area of ongoing clinical debate, with evolving perspectives on optimal timing, patient selection, and expected outcomes. This systematic review contributes to the current body of literature by synthesizing data from 30 studies encompassing over 1,200 patients, thereby providing insights into the factors that shape prognosis and surgical success.

A key finding of this review is the consistent demonstration across multiple studies that early surgical intervention, particularly in type I SCM, correlates with superior neurological and functional outcomes.[1-4,12,18] This observation is mechanistically supported by the pathophysiology of SCM, wherein progressive tethering and ischemic injury, if left uncorrected, can culminate in irreversible neural deficits.[1,5,18,19] Yamada et al demonstrated that spinal cord tethering induces oxidative metabolic disturbances that can only be reversed by releasing the tethering forces.[5] Clinically, this translates to a narrow therapeutic window during which surgery can halt disease progression and optimize recovery.

Conversely, patients presenting with longstanding preoperative deficits exhibit a more limited potential for neurological improvement.[1,4,12,18] Sinha et al reported that only 60 of 148 patients demonstrated motor improvement postoperatively, and the likelihood of recovery diminished with increasing chronicity of deficits.[4] Erşahin observed similar trends, with a majority of patients achieving stability but few achieving reversal of severe deficits.[12] These data underscore the importance of early diagnosis and prompt surgical referral, particularly for asymptomatic type I cases where prophylactic correction can preempt decline.[1,4,12,18]

In type II SCM, the surgical approach is more nuanced. Although early surgery is generally advocated for symptomatic patients, there is greater tolerance for conservative management in asymptomatic individuals.[1,9,20] The more benign natural history of type II lesions, which lack a rigid bony septum and exhibit lower tethering forces, supports this selective approach.[1,9,20] Nevertheless, Alnefaie et al demonstrated that type II patients still benefit from surgical release when they undergo spine corrective surgery, to avoid exacerbation of tethering.[1,9]

Urological outcomes represent an important but underexplored domain. Our review confirms that bladder dysfunction is common at presentation, particularly in type I SCM.[1,9,21] Encouragingly, surgical correction appears to stabilize or improve urological function in the majority of cases.[1,10,21] However, Proctor et al cautioned that subtle voiding dysfunction may persist despite normalization of overt symptoms, highlighting the need for systematic urodynamic evaluation.[10] Moreover, the presence of preoperative bladder dysfunction was itself a marker of more advanced disease and less favorable neurological prognosis.[1,10]

Orthopedic outcomes, though inconsistently reported, merit greater attention. Foot deformities, scoliosis, and leg length discrepancies were prevalent in our included studies.[1,7,11] Valdez et al provided valuable data indicating that early tethered cord release (TCR) reduces subsequent orthopedic surgical needs in SCM patients.[7] However, residual orthopedic sequelae remain a concern, necessitating close longitudinal follow-up and timely orthopedic intervention.

Reoperation rates across the literature were low, typically <5%.[1,12] This underscores the importance of meticulous initial surgical technique, including complete resection of the septum and watertight dural closure.[1,12,22] Nonetheless, the potential for regrowth of bony spurs, particularly in immature patients, warrants long-term imaging surveillance.[1,12,22]

One of the more contentious issues remains the risk-benefit calculus in asymptomatic type I SCM. While the majority of experts advocate for prophylactic surgery, some authors argue for observation in select patients, citing risks of surgical morbidity.[1,2,9,12,20,23] Our review aligns with the dominant view that early surgery is justified in most type I cases, given the substantial risk of progressive deterioration and the limited reversibility of established deficits.[1,2,4,12] The risk of surgical complications, although real, is generally low and transient.[1,4,8,14,22]

Another evolving theme is the role of intraoperative neurophysiological monitoring (IONM). Multiple studies, including Alnefaie et al reported that IONM contributes to safer dissection and reduced neurological morbidity.[1,11,24] However, variability in IONM availability across centers represents a limitation of current global practice.[1,24] Future efforts should aim to standardize IONM use, particularly in high-risk type I procedures.

Several gaps remain in the literature. Few studies included objective quality-of-life (QoL) metrics or validated functional outcome scales.[1,9,25] Similarly, long-term follow-up beyond adolescence remains rare, leaving open questions about late complications such as retethering and degenerative spine disease.[8,14,26] More prospective multicenter studies with standardized outcome reporting are urgently needed.[1,9,25]

Recent contributions from Beuriat et al and Mahapatra et al suggest that anatomical subclassifications of type I SCM may hold prognostic value, with certain spur configurations portending greater surgical complexity and risk.[8,27] Incorporation of such classifications into future surgical planning could further refine patient selection and consent discussions.

The intersection of SCM with other congenital anomalies, such as scoliosis and Chiari malformation, adds further complexity.[1,8,28] McMaster and others highlighted that occult intraspinal anomalies may underlie a substantial proportion of congenital scoliosis cases.[28] Accordingly, a high index of suspicion and routine preoperative MRI in scoliosis patients are warranted to identify occult SCM.[1,8,28]

Finally, the potential for adult presentation of SCM, though uncommon, should not be overlooked.[3,6,29,30] Cases of diastematomyelia manifesting as neurogenic claudication or progressive myelopathy in adults underscore the need for lifelong vigilance.[29,30] Surgical outcomes in adults appear favorable but somewhat attenuated relative to pediatric cohorts.[3,6,29,30]

CONCLUSION

In summary, this systematic review reinforces the paradigm that early surgical intervention is paramount, particularly in type I SCM. Preoperative neurological status remains the strongest predictor of postoperative outcomes. Urological and orthopedic sequelae warrant proactive multidisciplinary management. While surgical morbidity is generally low, careful technique and the use of IONM are advisable. Future research should prioritize prospective designs, long-term follow-up, and incorporation of validated functional outcome measures. The nuanced management of type II SCM and adult-presenting cases represents a fertile area for further investigation.

ACKNOWLEDGEMENTS

The authors would like to thank the professors of Instituto Ortopédico de Goiânia and mentors who contributed with valuable insights and guidance during the development of this study.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Moreira VRG, Agra IC, Santos RF, Ribeiro VJSLC. Understanding split cord malformation: from pathophysiology to long-term outcomes. Int J Res Orthop 2025;11:1197-204.