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RESEARCH

Composite type split cord malformation: a two-center study and analysis of a series of 25 cases

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Abstract

Introduction Split cord malformation (SCM) is a type of congenital spinal dysraphism characterized by the development of a split spinal cord separated by either a bony septum or a membranous/fibrous septum. The presence of two distinct SCMs in the same patient, with segments of normal cord interposed between them, is referred to as a composite-type SCM. This study aims to evaluate the variations of composite-type SCMs, which have been reported only at the case-report level in the literature, and to present the challenges encountered in surgical technique.

Results The study included 25 patients who underwent surgery for composite-type SCM at two centers. The mean age was 37.48 months. Sixty-four percent of the patients were female. The most common presenting findings were cutaneous stigmata, kyphoscoliosis, and gait disturbances.The most frequent location was the thoracolumbar region. Among the patients, 20% had Type 1 + Type 2 SCM, 12% had Type 1 + Type 1.5, and 68% had Type 1 + Type 1. The most commonly affected vertebrae were between T8 and T11.

Conclusion The demographic characteristics of composite SCMs were found to be similar to those of classical SCMs. However, cutaneous findings and kyphoscoliosis were more frequently observed in composite SCMs. In terms of localization, composite SCMs tended to occur at higher spinal levels and were more commonly found in the thoracic-thoracolumbar regions compared to classical SCMs. Intraoperative complications related to prolonged operative time and increased risk of bleeding should be anticipated and addressed in the preoperative period. Surgical planning should prioritize starting from the upper SCM, with the final step being the transection of the filum terminale.

Keywords Composite split cord malformation · Spina bifida occulta · Split cord malformation · Spinal dysraphism

Introduction

Split cord malformation (SCM) is a type of congenital spinal dysraphism characterized by the development of a split spinal cord separated by either a bony or membranous/fibrous septum. Depending on its type, one or two dural sacs may be formed, and SCM is frequently associated with spina bifida.

The condition was first described in 1837 by Ollivier, who used the term diastematomyelia to refer to a bifurcated spinal cord [1]. In 1906, Bruce introduced the term diplomyelia to describe cases with two hemicords lacking a bony septum [2]. Due to the confusion arising from various terminologies, Pang et al. proposed the unified term "split cord malformation" in 1992. In their classification, diastematomyelia (two hemicords separated by a bony septum) corresponds to Type 1 SCM, while diplomyelia (two hemicords separated by a membranous/fibrous septum) corresponds to Type 2 SCM [3].

In 2019, Meena et al. described a newly identified type of split cord anomaly characterized by ventral or dorsal placement, not occupying the entire spinal canal, and enclosed within a single dural sac. This group was defined as Type 1.5 split cord malformation, with dorsally located cases classified as Type 1.5a and ventrally located ones as Type 1.5b

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[4]. All these classifications have been proposed to aid in anatomical description, anticipate surgical challenges, and predict prognosis.

Although split cord malformation (SCM) is rare, it is frequently associated with additional anomalies, such as meningomyelocele, myeloschisis, kyphoscoliosis, hemivertebra, lipoma/lipomyelomeningocele, dermoid cysts, and others. Both the type of SCM and the accompanying anomalies can influence the surgical course and prognosis. Patients with SCM may present with diverse clinical symptoms and varying neurological deficits. Notably, untreated Type I SCM is known to result in progressive neurological deterioration [5].

The presence of two distinct SCMs in a single patient, separated by an intervening segment of normal spinal cord, is referred to as a composite-type SCM [6], Fig. 1]. In light of this information, composite-type SCM—being the rarest form of SCM-has been reported in the literature only in the form of isolated case reports or with a few cases in broader SCM series. Its surgical management is considerably more complex than other types. This study aims to assess the specific characteristics of composite-type SCM and to highlight the challenges encountered in its surgical treatment.

Materials and methods

This study was approved by the institutional ethics committee for non-interventional clinical research of our university under the approval number 304 and date 18.06.2025 and was conducted in accordance with the principles of the Declaration of Helsinki.

A total of 25 patients who underwent surgery for composite-type SCM between 2011 and 2025 at two medical centers (Dicle University and Kocaeli University) were included in the study.

The following variables were recorded: age (in months), sex, presence of cutaneous markers, clinical presentation, lesion localization, number of affected vertebrae, neurological status, presence of tethered cord, kyphoscoliosis, spina

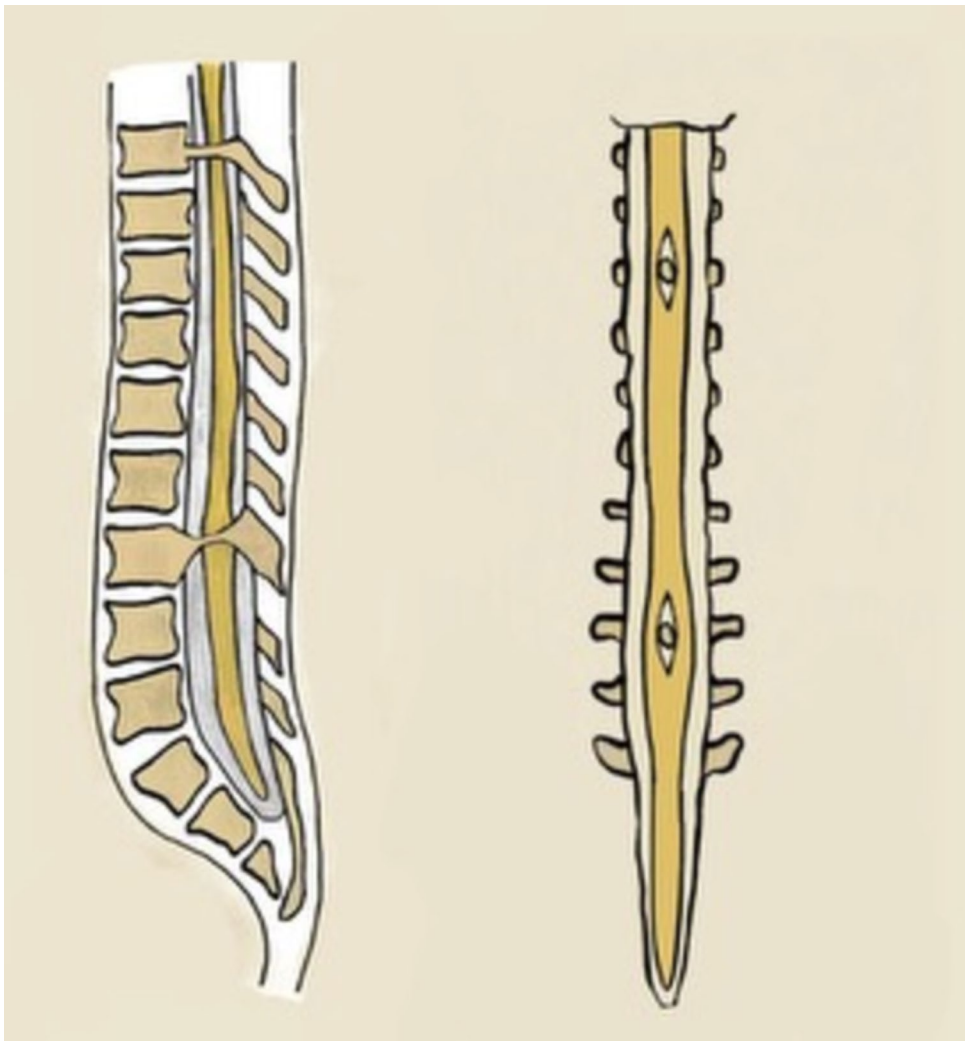


Fig. 1 Diagram of composite type split cord malformation

bifida aperta, Chiari malformation, syringomyelia, posterior fusion defects, hydrocephalus, and additional anomalies.

Radiological evaluations of all patients were performed using magnetic resonance imaging (MRI) and computed tomography (CT). Surgical planning, technique, and operative challenges were also analyzed.

Statistical analysis

SPSS for Windows (version 20.0) was used for statistical analysis. Data were evaluated in terms of mean values, counts and percentages, and minimum and maximum values.

Results

A total of 25 patients who underwent surgery for composite-type SCM were included in the study. The mean age at diagnosis was 37.480 months (range: 1–108 months]. Of the patients, 64% were female and 36% were male.

At presentation, 64% had spinal curvature (kyphoscoliosis), 36% had gait disturbances, 88% exhibited cutaneous markers (dimple-dermal sinus tract, hyperpigmentation, increased hair growth on the back), and 8% presented with a dorsal mass.

Neurologically, 36% of patients had lower extremity paresis, and 8% had plegia, while the remaining patients had no motor deficits in the lower extremities. Newly developed urinary retention was present in 16% of cases. Paraplegic patients were operated to prevent the progression of kyphoscoliosis.

In terms of SCM localization, 44% had thoracic, 36% thoracolumbar, 16% lumbar, and 4% lumbosacral involvement. No cervical SCMs were observed. Regarding SCM types, 20% had Type 1 + Type 2, 12% had Type 1 + Type 1.5, and 68% had Type 1 + Type 1 SCM (Fig. 2). The highest level affected was T3, and the lowest was L4. The most frequently affected vertebral levels were between T8 and T11. The average number of affected vertebrae was 3.32 (range: 2–5).

Radiological findings revealed kyphoscoliosis in 72%, Chiari malformation in 32%, ventricular dilation in 12 patients, and tethered cord in all cases. Syringomyelia was present in 44% of patients. When localization was examined, it was lumbar in 36.4%, thoracolumbar in 18.2%, thoracic in 36.4%, and cervicothoracic in 9%. In postoperative follow-up, syringomyelia was observed to disappear in 18.2% of patients, to shrink in 45.4%, and to remain the same in 36.4%.

Regarding additional anomalies, 24% of patients had renal anomalies, and 16% had cardiac anomalies.

Surgical technique

Although the surgical technique is fundamentally similar to that used in classical SCM cases, multi-level SCM presents distinct challenges. Due to the extended surgical exposure required for multi-level incisions, the risk of hypothermia and hemorrhage is higher, especially in younger patients. Therefore, complications associated with prolonged operative time and increased bleeding risk should be anticipated and addressed during the preoperative period.

In cases of SCM accompanied by tethered cord syndrome, it is generally recommended in the literature first to perform anatomical reconstruction and cord untethering before transecting the filum terminale (Fig. 3). This approach aims to prevent neurological deficits that may occur due to the sudden upward shift of the spinal cord during filum sectioning [7]. Although no standard protocol has been established specifically for composite SCM due to its rarity, the surgical approach described—starting from the upper SCM, performing reconstruction at each level, and concluding with filum terminale transection [8]—was adopted and implemented in all cases in this study.

In all patients, surgery was initiated at the upper SCM level. In cases where the incisions were close together, a single incision was used; for more widely spaced lesions, each level was accessed sequentially, starting from the uppermost, and incisions were closed one by one upon completion. This method aimed to minimize the risk of infection, hypothermia, and bleeding.

Intraoperative monitoring was used throughout the surgery. During surgery, controlled resection was achieved by taking into account electromyelographic changes during fibrotic band/bone spur resection and dissection of roots attached to the filum during filum release.

None of the patients experienced postoperative neurological deterioration compared to their preoperative status. Cerebrospinal fluid leakage from the surgical site was observed in two patients and was successfully managed with dressing on postoperative days 2 and 6, with no need for additional intervention.

One patient required intraoperative and another postoperative transfusion. No postoperative neurological worsening was observed in any patient. All patients were discharged without complications and followed up thereafter.

Discussion

Split cord malformations (SCMs] are a rare form of spinal dysraphism, and their terminology has evolved over time. With these changes in terminology, surgical approaches have also shifted, leading to the development of various classifications [7]. While Pang et al. classified SCM based on the

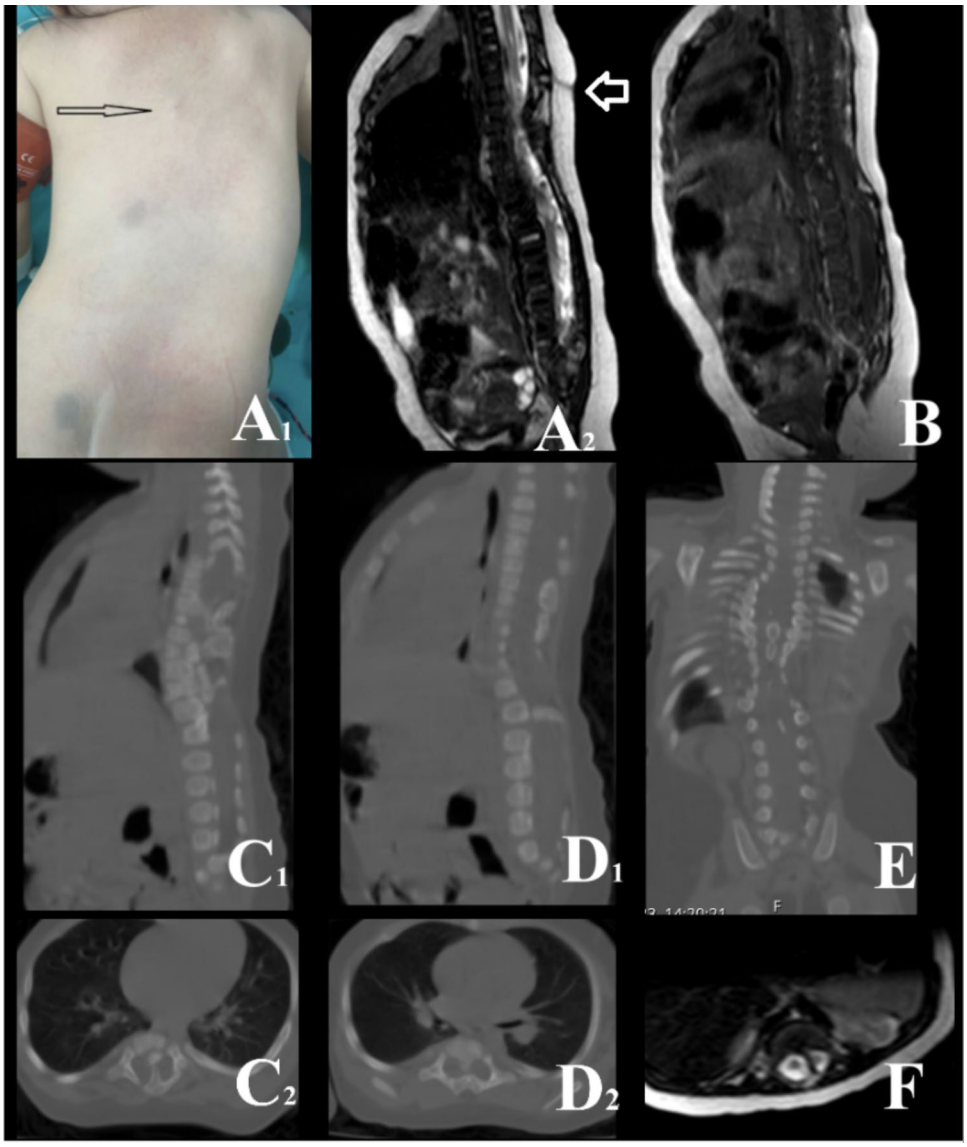


Fig. 2 A1. 6 month old girl, scoliosis (+), neurological examination normal. Black arrow indicates dermal sinus tract. **A2.** White arrow marks the dermal sinus tract at T2W MRI. The syrinx cavity is seen at the upper level. **B.**T1W MRI shows tethered cord. **C1**. CT image of upper SCM, multilevel Type 1 SCM.**C2.** Axial CT view of the upper SCM. **D1.** CT image of lower SCM, multilevel Type 1 SCM. **D2.** Axial CT view of the lower SCM. **E.** Two separate bony septum appearances on coronal CT. **F**. Two hemicord views on T2W MRI

presence of a bony septum or fibrous band [3], Meena et al. introduced an intermediate category, Type 1.5, representing cases with partial bony septum formation [4]. These classifications were primarily designed to aid surgical planning and predict prognosis. However, composite-type SCMs—unmentioned in these classifications and reported only as isolated case studies in the literature—stand out as a distinct group due to their intraoperative differences and therefore warrant separate consideration.

A search conducted in June 2025 using the term "Composite type split cord malformation" on PubMed and Google Scholar yielded 17 studies. Of these, nine were found to be unrelated to the topic, and one was later revised and republished. A total of seven cases (five case reports) were presented as individual case reports [8–12], and two original articles included two and six composite SCM cases, respectively, within broader SCM cohorts [13, 14]. However, to our knowledge, no case series specifically focused on composite-type SCM has been reported in the literature. Thus, we designed this study to highlight both the differences of composite-type SCMs from classical forms and the perioperative challenges involved.

During the third week of embryonic development, the ectoderm, mesoderm, and endoderm form in a stage known as gastrulation [15]. If the notochord fails to fuse properly and an accessory neuroenteric canal persists, an endomesenchymal tract develops [3]. If this tract contains a bony septum, Type I SCM forms; if it contains a fibrous band, Type II SCM results [16]. Although the presence of multiple accessory neuroenteric canals has been proposed to explain the formation of composite SCMs, the exact mechanisms underlying the development of multiple neuroenteric canals and different malformations at different levels remain unclear [17].

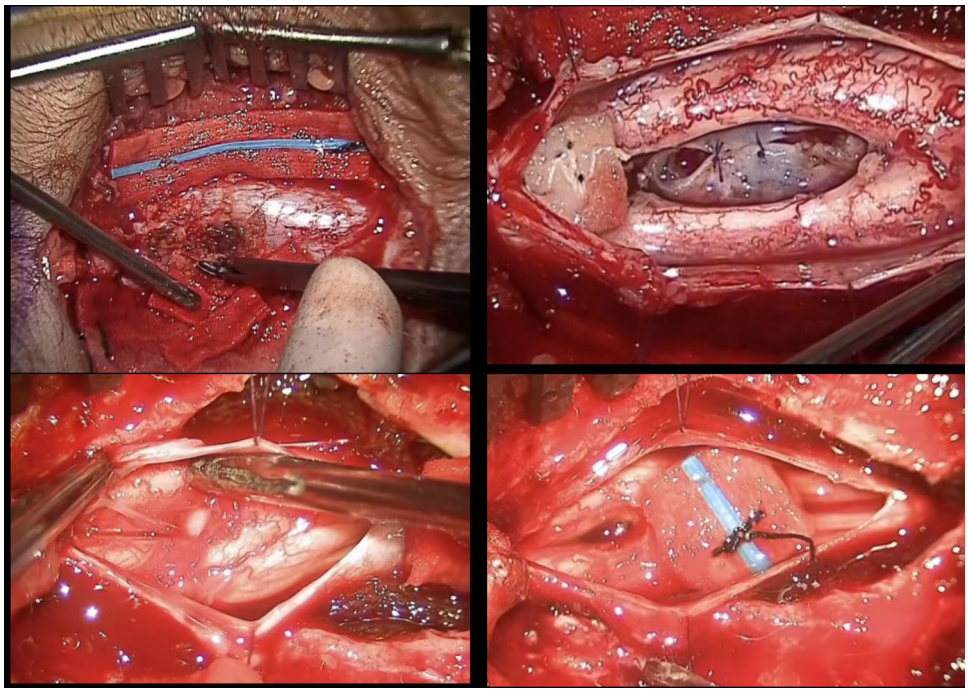


Fig. 3 Intraoperative images of Type 1 and Type 2 SCM before and after resection

In a 2013 study by Ersahin involving 131 patients, 73% were female, and the mean age at diagnosis was 16.5 months [14]. Similarly, our study demonstrated a female predominance, with a mean age of diagnosis of 22.4 months. These findings indicate that the demographic characteristics of composite SCMs are comparable to those of classical SCMs.

In a 2020 study by Alnefaie et al. involving 25 patients, scoliosis/kyphoscoliosis was reported in 64%, cutaneous markers in 48%, and neurological findings in 56% of cases [5]. In Ersahin's study, 58% had cutaneous markers, 38% had spina bifida cystica, 19% had paraparesis, 24% had unilateral leg atrophy and motor weakness, and 13% had bladder or bowel dysfunction [14]. In our study, cutaneous findings and kyphoscoliosis were observed at higher rates. Neurological functions were comparable to those in classical SCM cases. Notably, spina bifida cystica was frequently reported in Ersahin's series, while it was identified in only two patients in ours.

In classical SCM cases, Type I lesions are more commonly located in the lumbar region [7, 18]. While Type II lesions can be found throughout the spinal column, they are most frequently seen in the thoracolumbar and lumbar regions [5]. In our study, only 14.3% of cases had Type 1 + Type 2 SCM, indicating that all patients had a bony septum, which typically suggests lumbar localization. However, in contrast to classical SCM cases, our series revealed a predominance of thoracic and thoracolumbar involvement, which is a noteworthy finding.

Conclusion

The demographic features of composite-type SCMs were found to be similar to those of classical SCMs; however, cutaneous findings and kyphoscoliosis were more frequently observed in composite SCMs. Additionally, in terms of anatomical localization, composite SCMs tended to occur at higher levels in the spinal column, particularly in the thoracic and thoracolumbar regions.

In the perioperative setting, the distinguishing factor between composite and classical SCMs lies in the number of levels involved. The extended duration of surgery required for multi-level incisions increases the risk of hypothermia and hemorrhage, especially in younger patients. Therefore, potential complications due to prolonged surgical time and increased bleeding should be carefully addressed during the preoperative planning phase. Surgical intervention should proceed from the upper SCM level downwards, with transection of the filum terminale performed as the final step.

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Data availability Data and materials can be provided upon request.

Code availability Not applicable.

Declarations

Ethics approval The study was approved by the Dicle University (Approval number; 304, 18.06.2025).

Consent to participate The study was conducted in accordance with the Declaration of Helsinki, and informed written consent was obtained from the parents of all patients.

Consent for publication The authors have consented for publication.

Conflicts of interest The authors declare no competing interests.

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