

Unveiling the Current Understanding of Idiopathic Spinal Cord Herniation: A Systematic Review

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Abstract:

Background: Idiopathic spinal cord herniation (ISCH) is a rare condition that is characterized by ventral herniation of the spinal cord through a defect in the dura mater into the epidural space, with no identifiable cause. ISCH is frequently underdiagnosed, and the information available in case reports is limited. To provide an overview of the clinical manifestations and diagnosis of this condition, this study aims to conduct a review of reported cases of ISCH.

Methods: A literature review was carried out using seven databases. The search was conducted using the keywords “Idiopathic spinal cord herniation” OR “Idiopathic Ventral Spinal Cord Herniation” AND “Case report” OR “case series.”

Results: A total of 92 relevant papers reporting 224 cases, besides the index case, were determined. Of the cases, 58.5% were females and the mean age was 50.7 (SD 13.2) years. Symptoms, diagnoses, and outcomes were similar between genders. The most common clinical signs included motor symptoms (82.6%), instability (61.3%), hypoesthesia (59.2%), and disturbance of thermal sensitivity (47.3%). Brown-Séquard syndrome was observed in 27.2% of the cases, and surgical treatment was employed in 89.7% of the cases.

Conclusions: ISCH is a pathology that is principally treated with surgical approach. This study provides valuable insights into the clinical manifestations and diagnosis of ISCH, which can aid in the early recognition and treatment of this rare condition.

Keywords:

Systematic review, Idiopathic spinal cord herniation, Neurosurgery, Spine, Spinal cord diseases

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Introduction

ISCH has been a condition described since 1974 by Wortzman et al.¹. ISCH is characterized by the ventral herniation of the spinal cord through a defect in the dura mater into the epidural space, which results in symptoms of progressive thoracic myelopathy. With the increasing use of magnetic resonance imaging (MRI), the case reports of ISCH have increased in number, reaching approximately 200 reports in the literature².

Middle-aged adults are commonly more prone to ISCH, with a higher prevalence among females. Up to 50% of the cases are associated with Brown-Séquard syndrome, which

is characterized by paralysis of the ipsilateral extremity, loss of the sense of vibration and position, and contralateral loss of pain and temperature sensation, among other symptoms. ISCH is typically observed at the thoracic level, between the T3 and T7 segments, although isolated cases have been reported at the cervical and lumbar levels².

The pathogenesis of ISCH is not well understood, and non-specific symptoms may contribute to delayed diagnosis and treatment, which can lead to a poorer prognosis and a lower rate of symptom resolution. Timely surgical intervention is therefore crucial for optimal outcomes in patients with ISCH³.

Clinical symptoms, principal care, and treatment out-

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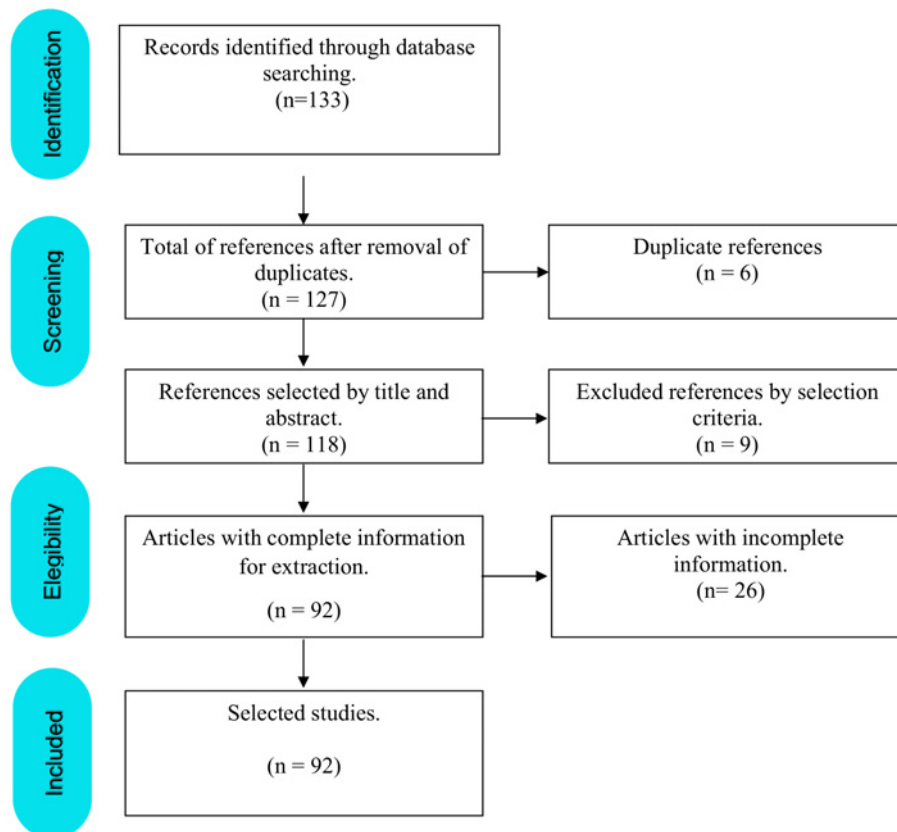


Figure 1. Flowchart illustrating the literature search process.

comes are important for figuring out how to give appropriate care. However, the scientific literature is limited, and there is a broad number of case reports. Using a comprehensive review of reported cases on ISCH, we aim to emphasize the prevalent symptoms, diagnosis, principal treatment, and postsurgical outcomes.

Materials and Methods

Literature review

Using the search strategy (“Idiopathic spinal cord herniation” OR “Idiopathic Ventral Spinal Cord Herniation”) AND (“Case report” OR “Case series”), a systematic literature review was conducted following the PRISMA statement. Seven databases (PubMed, Scopus, Embase, SciELO, ProQuest, and Springer) were searched. Two researchers independently screened the titles and abstracts for eligibility, and then, full-text articles were reviewed. Relevant data such as age, comorbidities, symptoms, diagnosis, treatment, and outcomes were extracted from the included studies. To ensure transparency and minimize bias, the study protocol was registered in PROSPERO (CRD42023367011).

Study selection

The inclusion criteria for the study were as follows: articles related to ISCH, articles in all languages, and articles with no publication date limit. The exclusion criteria for the

study were as follows: case reports with incomplete information, spinal hernias with an identifiable cause, and cases where ISCH was not the primary diagnosis but rather a differential diagnosis.

Assessing the quality of the selected articles

To ensure the quality of the included case reports, a critical appraisal will be conducted using the Joanna Briggs Institute critical appraisal tool for case reports. This tool includes nine items that assess the methodological quality of the case report, such as the clarity of the case presentation, the appropriateness of the diagnostic criteria, and the description of the intervention and outcomes. Each item will be scored as “yes,” “no,” “unclear,” or “not applicable” according to the information provided in the case report. Two independent reviewers will evaluate the quality of each case report, and any disagreements will be resolved through discussion or consultation with a third reviewer.

Statistical analysis

Descriptive and inferential statistical analyses were employed to synthesize the literature on ISCH. Using the Shapiro-Wilk test, the normality of quantitative variables was tested. Quantitative variables were described using mean, median, standard deviation, and interquartile range, whereas categorical variables were described using frequencies and percentages. Categorical variables such as Brown-Séquard syndrome (absent/present) and surgical outcome

Table 1. Summary of Case Reports.

	Total 224 n (%)
Mean age, years	50.7 (13.2)
Sex, male/female	93/131
Symptom duration, months	49.9 (57.9)
Comorbidities	
None	208 (93.7)
Diabetes mellitus	2 (0.9)
Immune disease	5 (2.3)
Paget's syndrome	1 (0.5)
Arterial hypertension	4 (1.8)
Precedent of cancer	1 (0.5)
COPD	1 (0.5)
Pain	74 (33)
Location:	
Lower limbs	38 (52.1)
Column	31 (42.5)
Thoracic	4 (5.5)
Emesis	1 (0.5)
Sensitive symptoms	
None	39 (17)
Hypoesthesia	132 (59.2)
Hyperesthesia	13 (5.8)
Anesthesia	9 (4)
Dysesthesia	8 (3.6)
Paresthesia	23 (10.3)
Motor symptoms	185 (82.6)
Unilateral	110 (59.4)
Bilateral	75 (40.6)
Spasticity	36 (16.4)
Alteration in proprioception	10 (4.6)
Hemosiderosis	1 (0.5)
Decreased thermal sensitivity	105 (47.3)
Gait disturbances	
None	82 (36.9)
Instability	136 (61.3)
Ataxia	4 (1.8)
Romberg	26 (12.3)
Urinary incontinence	42 (18.8)
Urinary retention	3 (1.3)
Bowel incontinence	9 (4.1)
Extensor plantar reflex	36 (16.7)
Reflex	
Normal	143 (63.8)
Hyporeflexia	17 (7.5)
Hyperreflexia	55 (24.5)
Clonus	9 (4.2)
Brown-Séquard	59 (26.6)
CSF fistula	14 (6.3)

Table 1. Summary of Case Reports (continued).

	Total 224 n (%)
Hernia	
Anterior	221 (98.7)
Posterior	3 (1.3)
Hernia position	
Cervical	4 (1.8)
Thoracic	217 (96.9)
Lumbar	3 (1.3)
Treatment	
Conservative	23 (10.3)
Surgical	201 (89.7)
Surgical approach	
Laminectomy	193 (96)
Discectomy	1 (0.5)
Pediculotomy	1 (0.5)
Costotransversectomy	5 (2.5)
Anterior approach	1 (0.5)
Surgical outcome	
Favorable	176 (78.5)
Unfavorable	18 (8)
Equal	30 (13.5)
Differential diagnosis	
None	201 (89.7)
Subarachnoid cyst	20 (9)
Herniated disk	2 (0.9)
Tarlov's cysts	1 (0.4)

(improve/non-improve) were compared using the Chi² test or Fisher's test. Quantitative variables were compared using Student's *t*-test or Kruskal-Wallis test. The statistical analyses were conducted using STATA 13.

Results

The initial search yielded a total of 118 articles. After duplicate removal, title and abstract screening, and full-text assessment, a total of 92 case reports¹⁻⁹²⁾ met the inclusion/exclusion criteria and were included in the final analysis (Fig. 1). The remaining 26 cases were excluded because of inadequate documentation or lack of important diagnostic information.

The 92 case reports included a total of 224 patients and a mean age of 50.7±13.2 years, with 58.5% being female. The average duration between symptom onset and diagnosis was 49.9±57.9 months, with no significant differences observed between genders (*p*>0.05) in terms of symptoms, diagnosis, and outcomes (Table 1). As shown in Fig. 2, motor symptoms, instability, hypoesthesia, and disturbance of thermal sensitivity were the most prevalent signs among patients.

No significant antecedents related to the manifestation of ISCH were found in the patients. However, the thoracic spine level was the most common location for ISCH, with

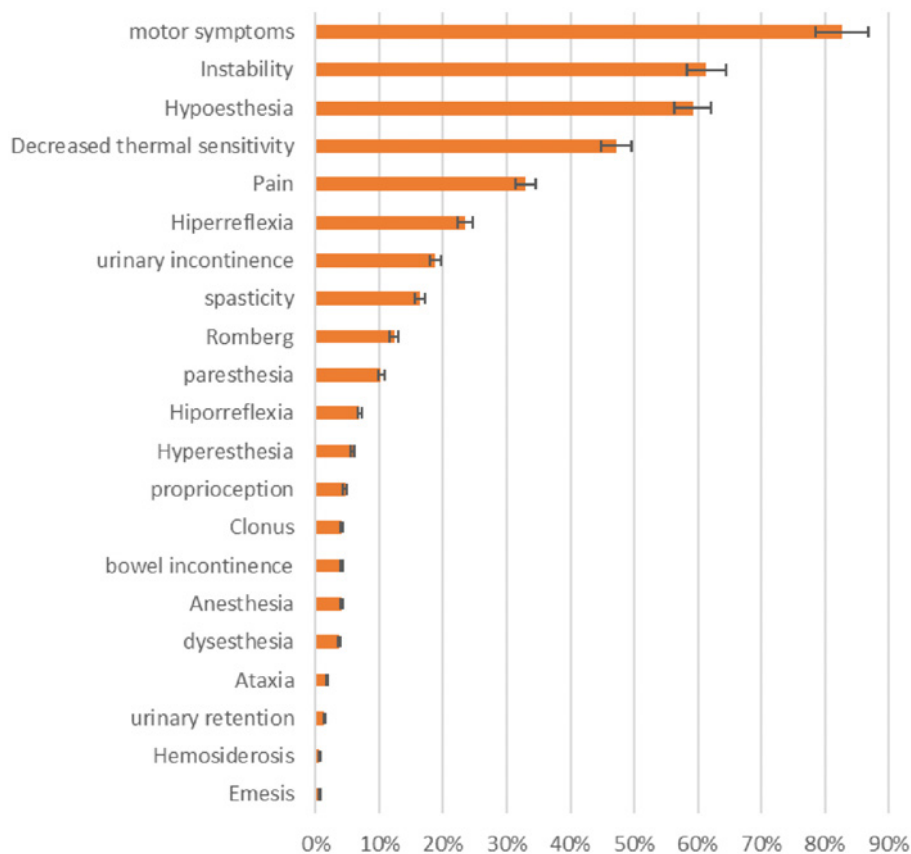


Figure 2. Prevalence of signs and symptoms in cases.

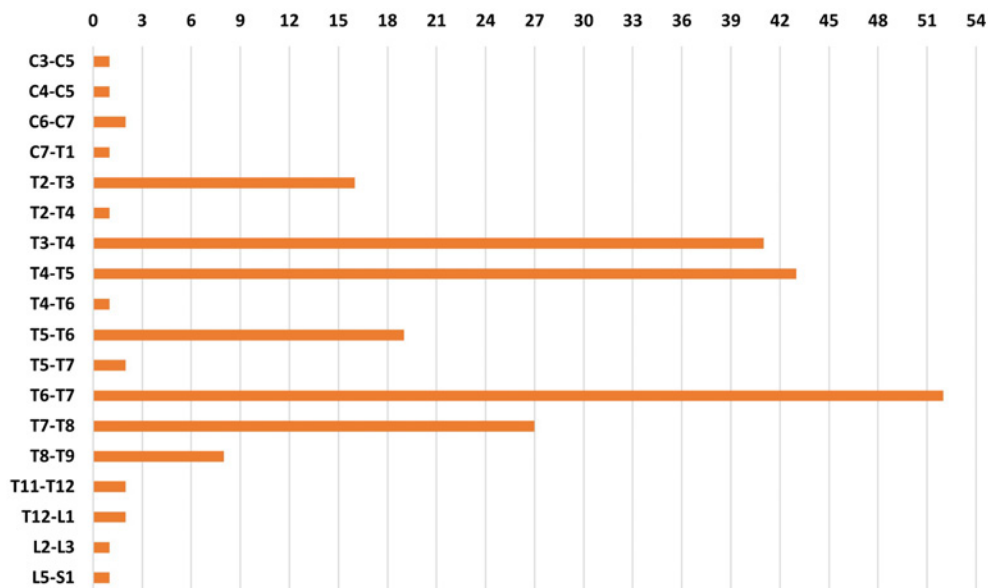


Figure 3. Prevalence of Idiopathic spinal cord herniation (ISCH) by spinal cord segments.

the T6-T7 segment being the most affected (22%) (Fig. 3). Furthermore, a higher percentage of hernias were located at the ventral level (98.2%).

Among the patients, 27.2% exhibited Brown-Séquard syndrome and 89.7% required surgery, with laminectomy being the most common procedure (96%).

The patients who presented with Brown-Séquard syndrome had ISCH located at the thoracic level and were pri-

marily selected for surgical intervention (96.7%) compared to those patients with an unclear clinical presentation where the presence of Brown-Séquard syndrome was not described (87.2%). Consequently, this resulted in a higher proportion of symptom resolution (85.2%), as shown in Table 2.

Conversely, regarding symptom resolution, a favorable outcome was observed in most patients (78%), especially among those who underwent surgical treatment. The evi-

Table 2. Summary of Case Reports by Brown–Séguard Syndrome Manifestation.

	Present n 61 (27.2%)	Absent n 163 (72.8%)	<i>p</i> value
Gender, Male/female: n	21/40	71/92	0.287
Age, years: median [IQR]	51 [44–60]	51 [41–60]	0.903 a
Duration of symptom, months: median [IQR]	36 [24–60]	24 [12–60]	0.137 a
Hernia: n (%)			
Anterior	61 (100)	160 (98.2)	
Posterior	0	3 (1.8)	0.567 b
Hernia position, n (%)			
Cervical	0	4 (2.5)	
Thoracic	61 (100)	156 (95.7)	
Lumbar	0	3 (1.8)	0.222 b
Treatment, n (%)			
Conservative	2 (3.3)	21 (12.8)	
Surgical	59 (96.7)	142 (87.2)	0.05 c
Surgical approach, n (%)			
Laminectomy	57 (96.6)	136 (95.7)	
Discectomy	0	1 (0.7)	
Pediculotomy	0	1 (0.7)	
Costotransversectomy	2 (3.4)	3 (2.1)	
Anterior approach	0	1 (0.8)	0.713 b
Surgical outcome, n (%)			
Favorable	52 (85.2)	124 (76)	
Unfavorable	4 (6.5)	14 (8.5)	
Equal	5 (8.3)	25 (15.5)	0.331 c
Time until the surgical outcome, days, median [IQR]	0 [0–3]	0 [0–5]	0.797 a
Differential diagnosis, n (%)			
None	52 (85.2)	149 (91.4)	
Subarachnoid cyst	9 (14.8)	11 (6.7)	
Herniated disk	0	2 (1.2)	
Tarlov’s cysts	0	1 (0.7)	NC

IQR: interquartile range; a Kruskal–Wallis test; b Fisher’s exact test; c Chi² test.

dence indicates a slight predominance of surgical management among those who showed improvement in comparison to those who did not. Additionally, the data from Table 3 revealed that costotransversectomy was more frequently performed in patients who did not experience improvement.

Table 4 presents the key findings obtained from MRI to facilitate the imaging recognition of ISCH and differentiate it from other similar conditions, like “spinal arachnoid web.” Notably, evidence of anterior herniation of the spinal cord is highly indicative of ISCH, as well as distortion of cerebrospinal fluid flow without complete interruption of the flow. Moreover, the highest diagnostic value in MRI is observed in high-resolution T2-weighted images, where changes in the epidural space or cerebrospinal fluid flow can be clearly visualized.

Discussion

Generally, ISCH is a rare and frequently overlooked cause of slowly progressive myelopathy. ISCH is characterized by the displacement of the spinal cord through a dural defect, typically found in the ventral region; however, a smaller percentage of cases may develop in the posterior region. In this review, it was observed that more than half of the ISCH patients are women, which is consistent with the existing literature that reports that approximately two-thirds of cases occur in women⁹⁰. The exact etiology of ISCH remains incompletely understood and is often regarded as idiopathic. Nevertheless, some studies suggest that it may be triggered by trauma, disk protrusion, arachnoid cyst pressure, or other factors⁹¹.

ISCH manifests as a slowly progressive myelopathy, characterized by symptoms and signs that indicate dysfunction

Table 3. Summary of Case Reports by Surgical Outcome.

	Improved 175	Not improved 49	<i>p</i> value
Gender, Male/female: n	72/103	19/30	721
Age, years: median [IQR]	50 [43–58]	52 [44–63]	0.249 a
Duration of symptoms, months: median [IQR]	36 [12–60]	24 [14–60]	0.994 a
Hernia: n (%)			
Anterior	173 (98.8)	48 (98)	
Posterior	2 (1.2)	1 (2)	0.529 b
Hernia position: n (%)			
Cervical	3 (1.7)	1 (2)	
Thoracic	169 (96.5)	48 (98)	
Lumbar	3 (1.7)	0	0.967
Treatment: n (%)			
Conservative	16 (9.3)	7 (14.3)	
Surgical	159 (90.7)	42 (85.7)	0.307 c
Surgical approach: n (%)			
Laminectomy	155 (97.6)	38 (90.4)	
Discectomy	1 (0.6)	0	
Pediculotomy	1 (0.6)	0	
Costotransversectomy	1 (0.6)	4 (9.6)	
Anterior approach	1 (0.6)	0	0.291
Time until the surgical outcome, days: median [IQR]	0 [0–5]	0 [0–1]	0.232 a
Differential diagnosis: n (%)			
None	156 (89)	45 (91.8)	
Subarachnoid cyst	17 (9.8)	3 (6.1)	
Herniated disk	2 (1.2)	0	
Tarlov's cysts	0	1 (2.1)	0.913

IQR: interquartile range; a Kruskal–Wallis test; b Fisher's exact test; c Chi² test.

Table 4. Main Findings in MRI.

- An abrupt anterior twist of the spinal cord can be detected by observing the scalpel sign.
- Ventral displacement and angulation of the spinal cord can be observed.
- High-resolution T2-weighted images show the hernia as a subtle signal change in the ventral epidural space.
- High-resolution T2-weighted images can assist in demonstrating the distortion of CSF flow.
- Nuclear trail sign: linear area of hyper-attenuation at CT or signal hyperintensity at MRI due to the calcification of nucleus pulposus leakage from a herniated disk.
- The absence or presence of CSF ventral to the spinal cord can help to differentiate between a web and cord herniation (generally in SAW, there is an absence of CSF flow).

CT: computed tomography CSF: cerebrospinal fluid MRI: magnetic resonance imaging SAW: spinal arachnoid web

Table Compiled from "Differentiation of Idiopathic Spinal Cord Herniation from Dorsal Arachnoid Membranes on Magnetic Resonance Myelography and Computed Tomography"⁹³⁾ and "Idiopathic Spinal Cord Hernia."⁹²⁾

of the spinal cord, such as paresis, hypoesthesia in the lower limbs, and thermal abnormalities. Loss of sphincter control may also be observed⁹²⁾. In the present review, motor symptoms, instability, hypoesthesia, and disturbance of thermal sensitivity were the most observed signs among patients, which is consistent with the typical clinical manifestations of myelopathy.

Brown-Séquard syndrome, characterized by ipsilateral motor function loss and contralateral pain and temperature

sensation loss, is a frequently observed presentation in patients with ISCH. Studies have reported varying occurrences of Brown-Séquard syndrome in ISCH, which range from 26% to 50%⁹¹⁾. Nevertheless, the present review identified a lower prevalence. This disparity in findings may be attributed to the fact that many of the cases reported in the literature do not explicitly mention the presence of Brown-Séquard syndrome but rather focus on the most prominent symptoms.

The diagnosis of ISCH can be established through MRI visualization of the spinal cord herniation, along with the presence of the “scalpel sign.” The imaging features of ISCH generally appear as a mass with a signal or density like that of the spinal cord. ISCH can be differentiated from other lesions, such as subarachnoid cysts, on MRI via the visualization of nerve roots traversing the subarachnoid space, as subarachnoid cysts displace nerve roots to the periphery (Table 4). MRI or high-resolution computed tomography with myelography are more sensitive imaging modalities for diagnosing ISCH⁹³). The diagnosis of ISCH is often challenging and can take approximately 49.9 months, emphasizing the difficulty in accurately identifying the condition.

Notably, there is a significant prevalence of hernias in the thoracic spine, specifically at the T6-T7 level, which is known to be susceptible to mechanical stress and trauma. These observations align with the existing literature, which identifies the T6-T7 segment as the most affected area, possibly due to the inherent kyphosis and limited mobility of this spinal segment. Nevertheless, there have also been reported cases of ISCH occurring in the cervical and lumbar regions^{42,58}).

The management of ISCH typically involves surgical intervention aimed at repositioning the spinal cord and repairing the dural defect, utilizing various surgical techniques. Primary closure of the dura mater with sutures poses a high risk of recurrence, hence the preference for using a dural patch. Among the surgical options available, laminectomy coupled with surgical reduction of the herniated spinal tissue using a dural patch has been identified as the most recommended approach²). The reviewed cases provide evidence that supports the recommendation that the majority of patients with ISCH were primarily treated surgically, with laminectomy being the most commonly performed procedure.

The postoperative outcome of ISCH patients is multifactorial, depending on various patient-specific factors, including age, severity and duration of neurological symptoms, and comorbidities. Improvement in symptoms is typically observed in the immediate postoperative period, with motor symptoms that show the most notable improvement¹²).

Strengths

The strengths of the presented study include the detailed clinical characterization of patients with ISCH, such as their demographics, clinical manifestations, and surgical management. The study highlights the importance of early recognition and diagnosis of ISCH since delayed diagnosis can lead to worsened outcomes. The high proportion of female patients and the predominance of hernias at the thoracic spine level, particularly at T6-T7, provide valuable insights into the epidemiology and clinical presentation of ISCH. The study also highlights the significance of surgical management for ISCH, with laminectomy being the predominant procedure. Moreover, the study underscores the varied clinical

manifestations of ISCH, with the incidence of Brown-Séquard syndrome in approximately 27.2% of patients accentuating the need for individualized treatment strategies.

Limitations

Some limitations of this study include the fact that it is based on case reports, which are subject to selection bias and reporting bias. Moreover, the study only includes patients with a confirmed diagnosis of ISCH, and it is possible that cases with milder symptoms or different clinical presentations were not identified. The study also does not provide information on the long-term outcomes of patients with ISCH, considering that most of the cases included had relatively short follow-up periods. Finally, the study does not explore potential risk factors or causes for ISCH, which may limit the generalizability of the findings to other populations or settings.

Conclusion

The current standard of care for treating ISCH involves the use of a patch to correct the dural defect. Nevertheless, the success of the postoperative outcome heavily relies on the early diagnosis of ISCH. As such, timely recognition of this condition is crucial for successful treatment and improved patient outcomes. With ongoing research efforts and advancements in diagnostic techniques and surgical management, it is hoped that patients will continue to have improved results in the future.

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Ethical Approval: Institutional approval was not required for this systematic review of the literature.

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