Spinal arachnoid web-a distinct entity of focal arachnopathy with favorable long-term outcome after surgical resection. Analysis of a multicenter patient population

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Abstract: BACKGROUND CONTEXT Spinal arachnoid web (SAW) is a rare condition characterized by focal thickening of the arachnoid membrane causing displacement and compression of the spinal cord with progressive symptoms and neurological deficits. Recent reports and clinical experience suggest that SAW is a distinct entity with specific radiological findings and treatment strategies distinguishable from other arachnopathies and potential differential diagnoses. PURPOSE To better define the diagnostic and clinical features, treatment options and outcomes of surgically treated SAW. STUDY DESIGN Multicentric retrospective cohort study. PATIENT SAMPLE Twelve cases of SAW surgically treated at three different centers. OUTCOME MEASURES Self-reported and neurological outcome measurements (pain, sensory-motor deficits, vegetative dysfunctions) were assessed at follow-up timepoints. METHODS Retrospective review of prospectively collected data on all patients surgically treated for SAW from three participating neurosurgical centers between 2014 and 2020. Clinicopathological data, including neurological presentation, radiological and histological findings and outcome data were analyzed. RESULTS Twelve radiologically and surgically confirmed cases of SAW were analyzed. Mean patient age was 54.7 [±12.7], 67% were male. All SAWs were located in the posterior thoracic dural sac. On magnetic resonance imaging (MRI), the "scalpel sign" - a characteristic focal dorsal indentation of the spinal cord resembling a scalpel blade - was identified in all patients. A focal intramedullary syrinx was present in 83%. Preoperative clinical symptoms included signs of myelopathy, pain, weakness and sensory loss, most commonly affecting the trunk/upper back or lower extremities. Laminectomy or laminoplasty with intradural excision of the SAW was the surgical treatment of choice in all cases. Intraoperative ultrasound was valuable to visualize the cerebrospinal fluid (CSF) flow obstruction, confirm the SAW location before dura incision and to control adequacy of resection. After surgery, sensory loss and weakness in particular showed significant improvement. CONCLUSIONS The present study comprises the largest series of surgically treated SAW, underscoring the unique clinical, radiographic, histopathological, and surgical findings. We want to emphasize SAW being a distinct entity of spinal arachnopathy with a favorable long-term outcome if diagnosed correctly and treated surgically. Intraoperative ultrasound aids visualizing the SAW before dural incision, as well as verifying restored CSF flow after resection.

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Clinical Study

Spinal arachnoid web—a distinct entity of focal arachnopathy with favorable long-term outcome after surgical resection. Analysis of a multicenter patient population

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Abstract

BACKGROUND CONTEXT: Spinal arachnoid web (SAW) is a rare condition characterized by focal thickening of the arachnoid membrane causing displacement and compression of the spinal cord with progressive symptoms and neurological deficits. Recent reports and clinical experience suggest that SAW is a distinct entity with specific radiological findings and treatment strategies distinguishable from other arachnopathies and potential differential diagnoses.

PURPOSE: To better define the diagnostic and clinical features, treatment options and outcomes of surgically treated SAW.

STUDY DESIGN: Multicentric retrospective cohort study.

PATIENT SAMPLE: Twelve cases of SAW surgically treated at three different centers.

OUTCOME MEASURES: Self-reported and neurological outcome measurements (pain, sensorymotor deficits, vegetative dysfunctions) were assessed at follow-up timepoints.

METHODS: Retrospective review of prospectively collected data on all patients surgically treated for SAW from three participating neurosurgical centers between 2014 and 2020. Clinicopathological data, including neurological presentation, radiological and histological findings and outcome data were analyzed.

RESULTS: Twelve radiologically and surgically confirmed cases of SAW were analyzed. Mean patient age was 54.7 [±12.7], 67% were male. All SAWs were located in the posterior thoracic dural sac. On magnetic resonance imaging (MRI), the "scalpel sign" - a characteristic focal dorsal indentation of the spinal cord resembling a scalpel blade - was identified in all patients. A focal intramedullary syrinx was present in 83%. Preoperative clinical symptoms included signs of myelopathy, pain, weakness and sensory loss, most commonly affecting the trunk/upper back or lower

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CONCLUSIONS: The present study comprises the largest series of surgically treated SAW, underscoring the unique clinical, radiographic, histopathological, and surgical findings. We want to emphasize SAW being a distinct entity of spinal arachnopathy with a favorable long-term outcome if diagnosed correctly and treated surgically. Intraoperative ultrasound aids visualizing the SAW before dural incision, as well as verifying restored CSF flow after resection.

Keywords: Arachnoid membrane; Intradural; Intraoperative ultrasound; Operative video; SAW; Spinal arachnoid web; Spine surgery

Introduction

Spinal arachnoid web (SAW) is a rare pathology causing spinal cord compression due to the formation of thickened arachnoid tissue [1]. It can lead to spinal cord compression or focal syrinx formation causing symptoms of myelopathy such as progressive (neuropathic) pain, weakness, sensory deficits or other less common neurological findings. Syringomyelia is frequent and caused by mass effect of the SAW or obstruction of cerebrospinal fluid (CSF) flow [2,3].

Clinical management is challenging since the spinal magnetic resonance imaging (MRI) findings are often subtle and easily overseen. In addition, microsurgical treatment requires a high degree of skill and experience. Only 45 individual cases of surgically confirmed SAW have been described in the literature [4–6]. Current data indicate that SAW is an extremely rare form of intradural spinal pathology, raising the possibility of under- or even misdiagnosis. The latter is attributed to a variety of overlapping pathologies described in the literature (eg, idiopathic syringomyelia) [7,8], which also present with syringomyelia. However, these cases lack the characteristic scalpel sign seen on conventional MRI studies and there is no associated CSF flow obstruction. Furthermore, a small focal intramedullary syrinx or focal signs of myelomalacia subsequent to focal cord compression may be misdiagnosed as an intramedullary tumor.

As diagnostic and surgical treatment of SAW are evolving and are currently mainly performed in specialized centers, our study aimed on summarizing the cumulative experiences of three centers with SAW and raising awareness on this specific and difficult to diagnose condition. We therefore gathered diagnostic features, accompanied by radiological images and videos of intraoperative ultrasound (ioUS) and microsurgical techniques to illustrate that SAW should be considered as a distinct form of spinal arachnopathy. In the present study, we retrospectively analyzed 12 surgically treated SAWs from three different neurosurgical centers and compared the multicentric experience regarding diagnosis, surgical treatment and clinical outcome with the available literature.

Methods

Study population

In the present study, we retrospectively identified all patients with SAW diagnosed between 2014 and 2020, who were surgically treated at three different neurosurgical centers (Department of Neurosurgery University Hospital Zurich, Division of Neurosurgery - St. Michael's Hospital University of Toronto and Department of Neurosurgery University Hospital Geneva). Patients' characteristics, neurological symptoms, radiological findings, operative reports, video sequences of ioUS and microsurgical procedures, histopathological sections, and outcome characteristics were retrieved from patient records. Most recent follow-up consultations were partly carried out by telephone due to the 2020/2021 pandemic.

Ethical considerations

Data extraction and analysis performed in this study was done in accordance with the ethical standards of the

Table 1
Summary of patient and spinal arachnoid web characteristics

Characteristics	No. of patients	Distribution
Sex		
Male	8	67%
Female	4	33%
Age		54.7 [±12.7]
Spine region		
Thoracic	12	100%
Axial orientation		
Dorsal	12	100%
Scalpel sign present	12	100%
Syrinx		
none	2	17%
above	4	33%
same level	3	25%
below	3	25%
History		
Traumatic spine injury	6	50%

Age is presented as mean ± standard deviation (SD).

Table 2
Clinical and MR-imaging information of the 12 patients with spinal arachnoid web

		MRI findings				Medical history		
	Age/sex	Lvl	Scalpel sign	Myelopathy	Synrinx	Sympt. duration	Spinal trauma	Operation
1	67/m	T4–5	T5	no	T5–T6	12 m	no	T3–6 LE
2	79/m	T5–6	T6	yes	T6–T8	36 m	no	T4–7 LE
3	60/m	T3–4	T3	no	T1–T3	48 m	yes	T2–4 LE
4	49/m	T5–6	T5	no	T5	30 m	yes	T4–7 LE
5	41/m	T2–3	T3	yes	C5–T3	27 m	no	T2–4 LE
6	50/f	T3–4	T3	yes	T2–T3	23 m	yes	T2–4 LE
7	35/m	T5–6	T5	no	T4–T9	19 m	yes	T5–6 LE
8	60/m	T5–9	T7	no	T7–T8	0.5 m	yes	T6–7 LE
9	51/m	T7–9	T8	no	T8–T9	60 m	no	T7–8 LE
10	39/f	T4–5	T5	no	no	unclear	no	T3–6 LP
11	55/f	T1	T1	yes	C7–T1	24 m	yes	T1 LE
12	70/f	T3–4	T4	no	no	2 m	no	T3–4 HLE

m, male; f, female; m, months; LE, laminectomy (with intradural excision); LP, laminoplasty (with intradural excision); HLE, hemilaminectomy (with intradural excision).

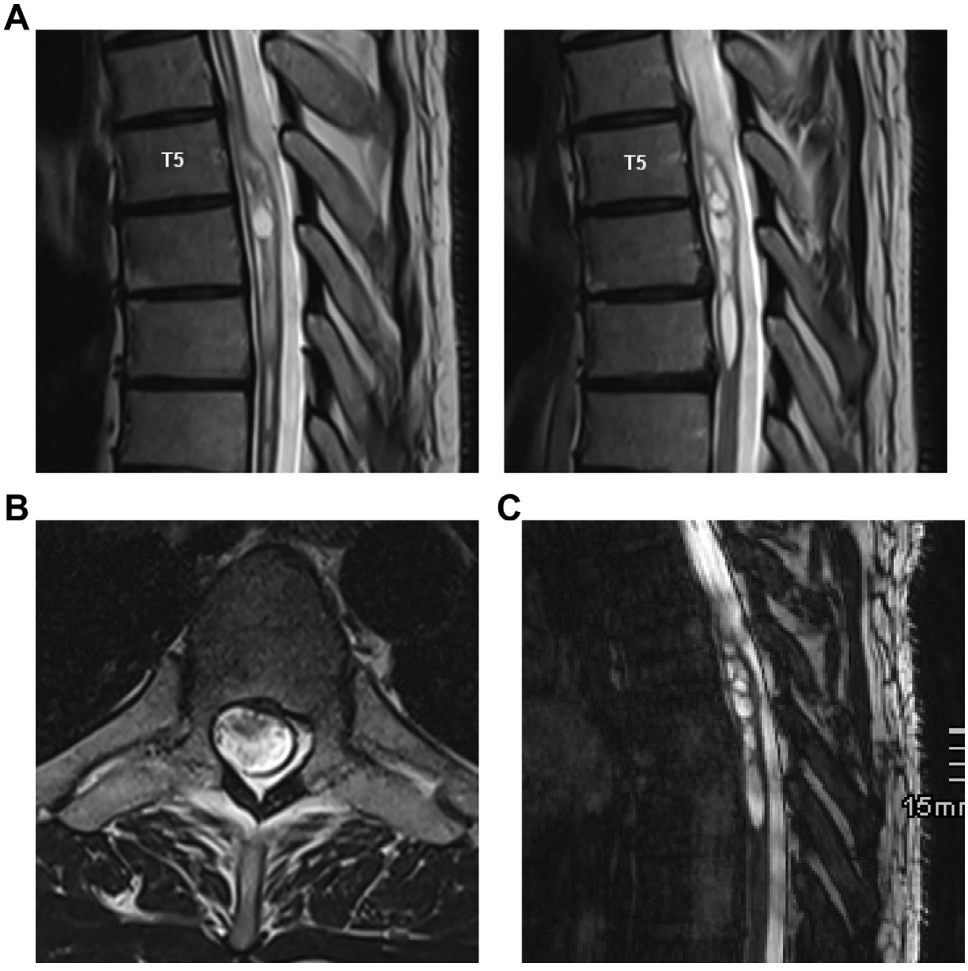

Fig. 1. Exemplary preoperative presentation of spinal arachnoid web in MRI. (A & B) T2-weighted sagittal images showing the scalpel sign (left) and the syrinx (right), axial section shows the ventrally displaced spinal cord. (C) Sagittal constructive interference in steady-state (CISS) sequence of preoperative arachnoid trabeculae.

institutional research committees and approval by the local ethical review boards of the participating neurosurgical centers. Informed consent was obtained from each patient.

Data analysis

Due to the small patient number, descriptive statistical analysis was performed.

Results

General patient characteristics

The contributing centers identified 12 patients with surgically treated SAW. The majority of cases were male (n = 8; 67%) with a mean age of 54.7 years (±12.7 SD); range 35 to 79 years (Table 1).

Clinical presentation and course

Most patients presented with progressive pain (75%, 9/12), which most frequently affected the trunk/back or lower extremities (Table 3). The majority of patients experienced subjective weakness of the lower extremities (75%, 9/12), which correlated with mild paresis on neurological examination. Additionally, 67% (8/12) had a partial sensory loss, mostly in the lower extremities or portions of the trunk/back, both contributing to gait instability or disturbances. On neurological examination, all but two patients revealed upper motoneuron (UMNS) or pyramidal signs (PMS) of the lower extremities. Distinct vegetative dysfunction was only present in one patient (impaired bladder emptying and sexual dysfunction). Whole spinal axis and cranial imaging were performed to rule out other causes for the neurological symptoms. Mean preoperative symptom duration was 25.6 months (median = 24 months, range: 2 weeks to 5 years). In six patients (50%), assessment of the medical history revealed blunt trauma to the thoracic spine, such as falls (1–3 m), without known fractures at the time of trauma. Furthermore, there were no cases with previous spinal surgery or spinal cord injury at the same level as the SAW.

Imaging findings

In all cases, the SAW was localized in the upper and mid thoracic spine (83%, 10/12 involving Th 3-6), with the mass effect located in the dorsal aspect of the dural sac, compressing the spinal cord from posterior (Tables 1 and 2). MRI revealed the presence of the previously described "scalpel sign" in all 12 cases (Figs. 1A, B & 2). In some cases constructive interference in steady-state (CISS) or cardiac-gated phase-contrast cine mode MRI sequences were used to visualize the adhesive arachnoid membranes (Fig. 1C) or the CSF flow obstruction (Supplementary Fig. 1). Furthermore, a syrinx of varying extent (ranging from 1 to 6 levels, median = 2; see Table 2 for individual case characteristics), was present in all but two cases. The syrinx was found to extend in roughly equal proportions above, below or at the level of the SAW (Table 1 & Fig. 1).

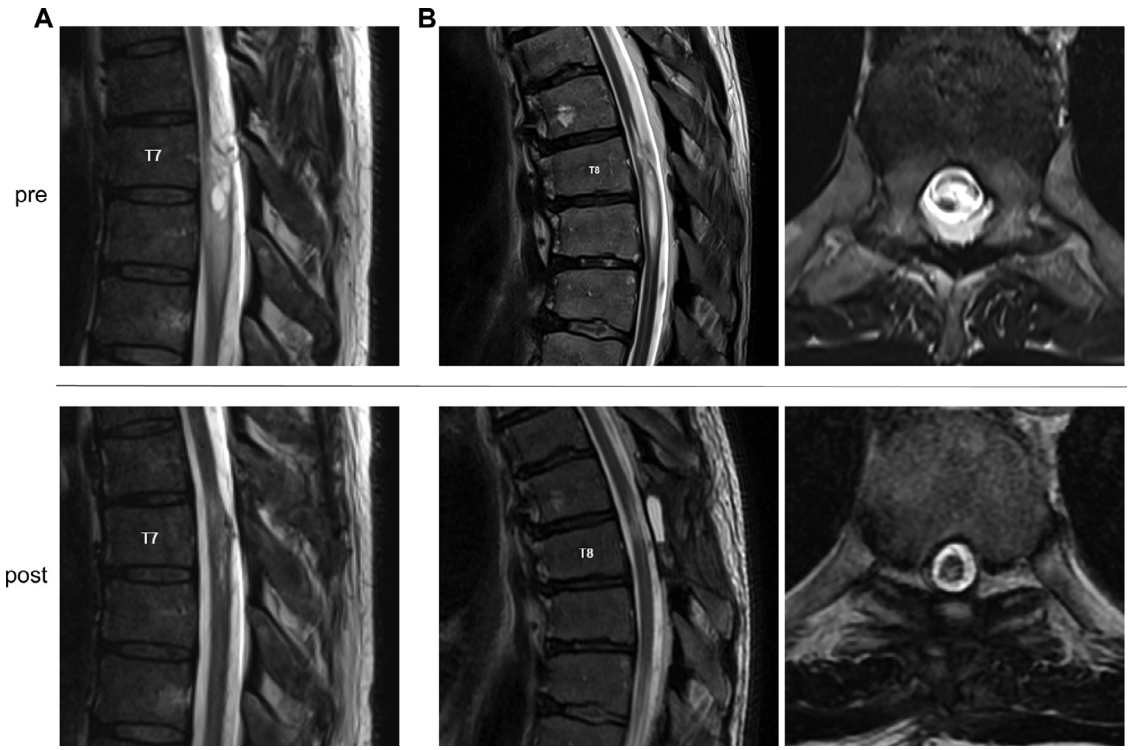

Fig. 2. Illustrative examples of two cases (A & B) with pre- (upper) and postoperative (lower row) MRI images. pre, preoperative; post, postoperative.

Surgical treatment and intraoperative presentation

In all cases, the surgical approach was microsurgical laminectomy (hemilaminectomy and laminoplasty in one case each) at the affected level with intradural excision of the SAW (Table 2 & Fig. 4). Prior to opening of the dural sac, ioUS was used to visualize the pathology, confirm the correct level and verify the suspected diagnosis (Fig. 3 & Video 1). After dural opening, the SAW was identifiable as a thickened arachnoid membrane (Fig. 4). Microsurgical excision of the SAW is demonstrated in Video 1. After completion, the restored CSF flow and the complete excision were again visualized by ioUS. A representative case is demonstrated in Fig. 3 and Video 1. The video illustrates the preoperative pulse-synchronous movement of the arachnoid-membrane in a caudo-rostral fashion with associated, impaired CSF flow. After intradural excision of the SAW, the postoperative ioUS control showed restored CSF flow (Fig. 3 & Video 1).

Histopathological characteristics

Histologically, the resected membrane (specimens of three patients available) consisted of fibrovascular connective tissue as well as small nests of meningothelial cells (Fig. 5). Immunohistochemical preparations showed reactivity for epithelial membrane antigen (EMA) in the meningothelial cells, confirming arachnoid origin.

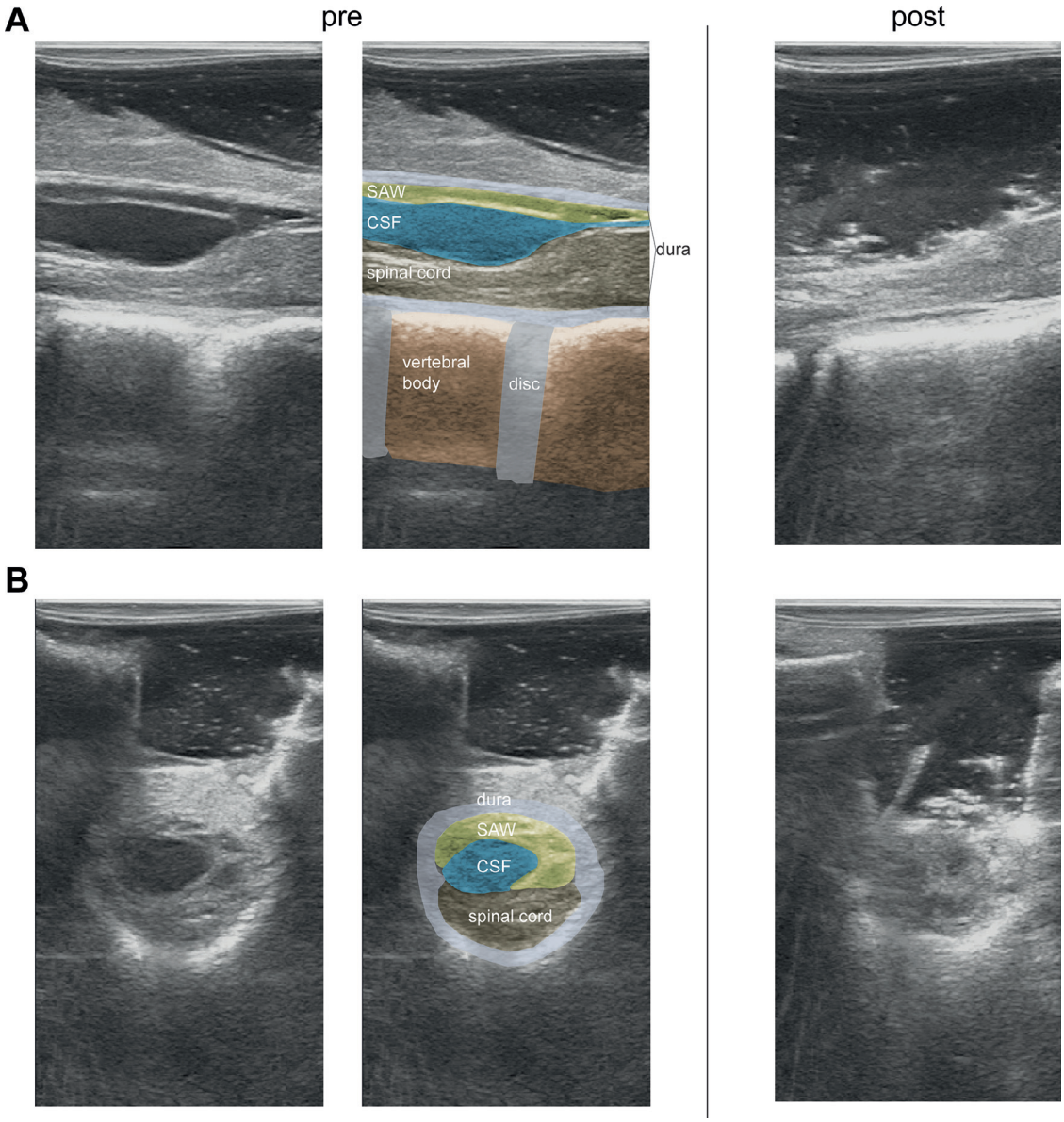

Fig. 3. Intraoperative ultrasound before dural opening: Visualizing the arachnoid web as a focal thickening of arachnoid membrane with subsequently impaired CSF flow. Pre- (left column row) and postoperative (right column) ultrasonic features are shown in sagittal (A) and axial (B) view. pre, preoperative; post, postoperative; SAW, spinal arachnoid web; CSF, cerebrospinal fluid.

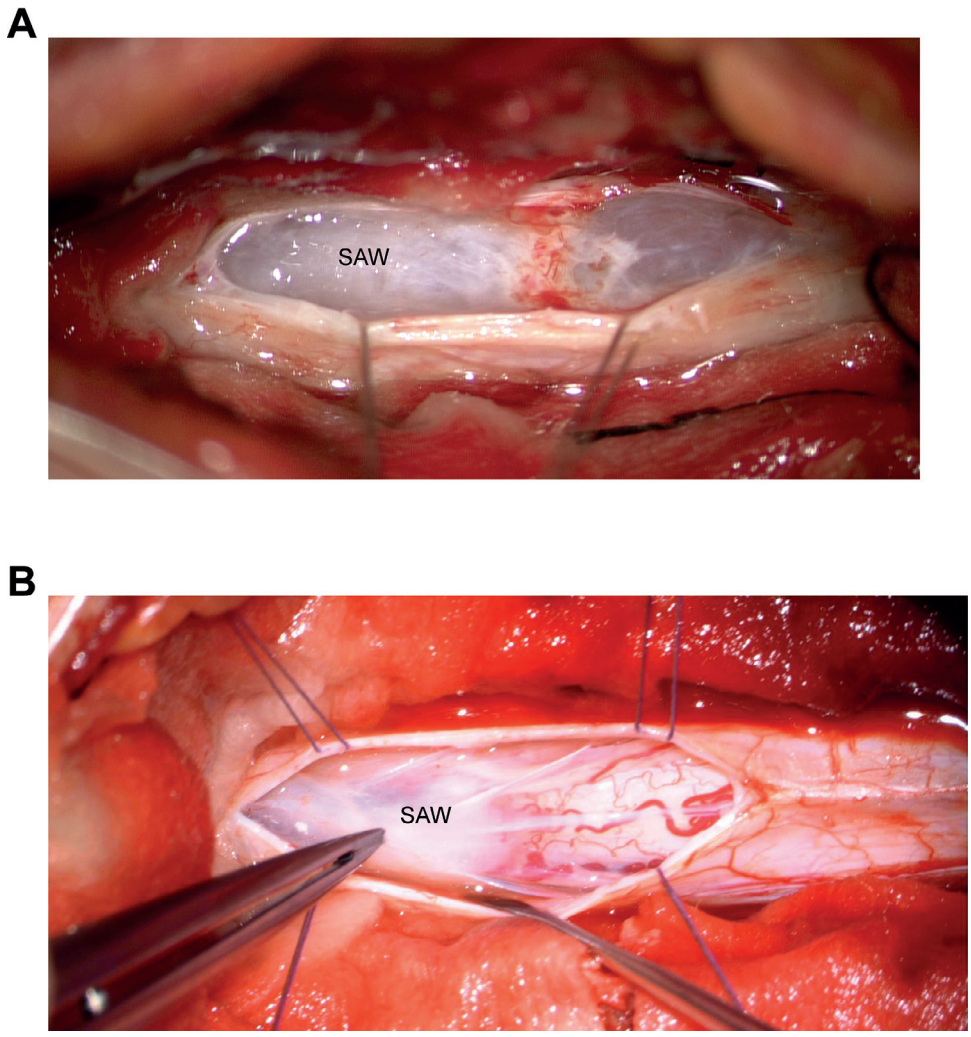

Fig. 4. Intraoperative view of two exemplary cases (A & B): After opening the dura, the SAW can be identified as a thickened arachnoid membrane. SAW, spinal arachnoid web; CSF, cerebrospinal fluid.

Clinical outcome

Mean follow-up time after operation was 1169 days (3.2 years; Table 3). Postoperatively, all but one patient experienced significant symptom improvement. In particular, there was substantial recovery from weakness and sensory loss (Table 3). In the single case with preoperative vegetative dysfunction, the bladder emptying impairment improved to an extent that the patient no longer required self-catheterization. Medical history review of the only patient (#10), who did not show postsurgical clinical improvement revealed a long history of chronic pain for which various diagnoses were considered (eg, cervicobrachialgia with cervical disc herniation and subsequent surgery, among others). After surgical resection of the SAW, the postoperative MRI showed sufficient decompression of the spinal cord without any surgical complications or spinal cord edema or trauma. However, there was no improvement of symptoms.

Discussion

In the current study, we evaluate the largest series of surgically confirmed SAW and illustrate the distinctive radiographic, histopathological and intraoperative features of this unique form of focal spinal arachnopathy. In addition, we document the favorable clinical course after microsurgical excision of the arachnoid web.

The largest series to date, published by Mallucci et al. in 1997 [9], includes 9 patients treated surgically for confirmed SAW. They describe a favorable outcome when SAW was surgically resected rather than shunting the syrinx. Since the initial description in 1997, less than 50 surgically confirmed cases of SAW have been reported in the literature [4–6].

It is now considered to represent a distinct pathological subtype of focal spinal arachnopathy. Our cases showed an overall strong improvement of clinical symptoms after surgery. Importantly, motor function recovered significantly

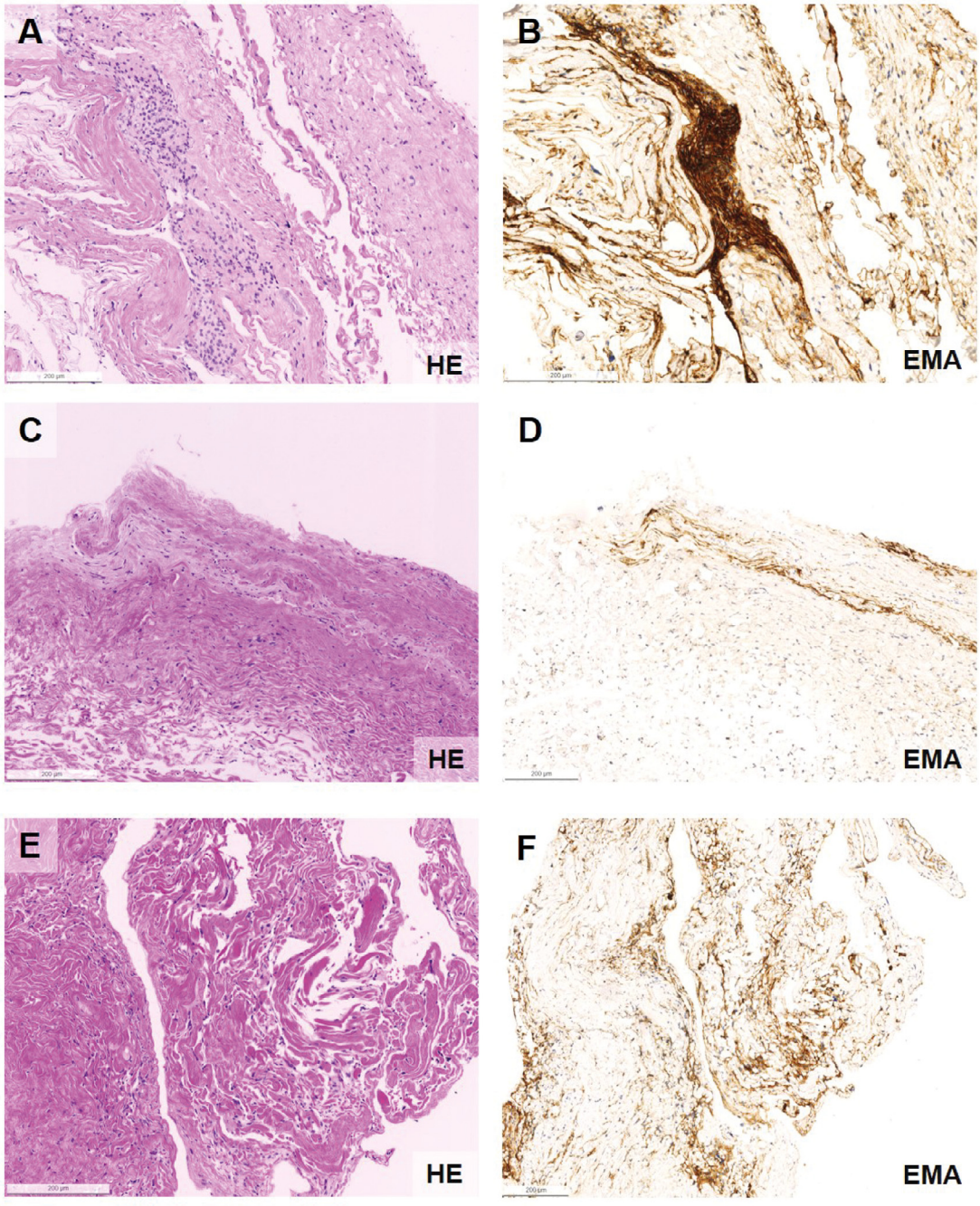

Fig. 5. Representative histology of three patients (patient 1 A-B, patient 2 C-D, patient 3 E-F). Histologically, the resected membrane consisted of delicate fibrovascular connective tissue with small nests of meningothelial cells (HE staining, A, C, E). Immunohistochemistry showed reactivity for EMA in the meningothelial cells. HE, hematoxylin and eosin staining; EMA, epithelial membrane antigen.

after decompression and intradural excision of the SAW. Return of sensory function and significant pain relief was also observed. Our findings with over 91% of patients with postoperative symptom improvement and one patient with a history of chronic pain and stable symptoms after surgery are in line with the previously reported cases in the literature (clinical improvement in 91% of all operated SAW reported in the literature, 41/45). Therefore, our cases support surgery as the treatment of choice for symptomatic SAW [4–6,10]. Surgical treatment leads to favorable clinical outcome and, as our study and the current literature suggests, there is good long-term symptom control with rarely reported symptom recurrence [4], especially compared to unspecific secondary arachnopathies. In contrast to SAWs, secondary arachnopathies after, eg, spinal surgery with arachnoid scarring, infections, or spinal hemorrhage are often multifocal and diffusely distributed over different spinal levels. In these cases, insufficient symptom amelioration

Table 3
Preoperative clinical characteristics, follow-up time in days and postoperative outcome of each patient

	Preoperative					Follow-up (days)	Postoperative course			
	Pain	Sens. loss	Weak.	Veg. dysf.	UMNS/PMS		Pain	Sens. loss	Weak.	Veg. dysf.
1	LE/TB	no	LE	no	yes	699	=		=	
2	no	LE	LE	no	yes	769		=	+	
3	UE/LE	UE/LE	LE	no	yes	1452	+	=	+	
4	TB	no	LE	no	yes	1986	+		+	
5	TB	UE	UE	no	no	2309	=	++	++	
6	TB	no	LE	no	yes	2400	=		++	
7	TB/LE	TB/LE	LE	no	yes	801	+	++	++	
8	no	TB/LE	LE	no	yes	378		++	++	
9	no	TB/LE	LE	yes	yes	519		+	++	+
10	TB/LE	no	no	no	no	760	=			
11	TB/UE/LE	UE	UE/LE	no	yes	1837	++	++	++	
12	TB	TB	no	no	yes	114	+	++	=	

Sens. loss, Sensory loss; Weakn., Weakness; Veg. dysf., Vegetative dysfunction; UMNS, upper motoneuron sign; PMS, pyramidal sign; LE, lower extremity; UE, upper extremity; TB, trunk/back.

+ indicates improvement, ++ strong improvement, = no improvement. Of note, no worsening of symptoms was evident in any patient (-).

and a high recurrence rate after surgical treatment by adhesiolysis and/or shunting of the associated syrinx is reported [11–17]. Although our study strongly suggests a favorable long-term outcome with a mean follow-up period of more than 3 years, subsequent studies and continued follow-ups of the so far treated patients are necessary to capture the total rate of recurrences or sequalae.

Accurate diagnosis, in particular, the differentiation from other arachnopathies is crucial, as progression can result in severe disability in untreated patients. In conventional MRI studies, diagnostic criteria rely on the so-called “scalpel sign” [18–22] which is a characteristic focal dorsal indentation of the spinal cord resembling a scalpel blade. This finding, first described by Reardon et al. in 2013 [22], is considered pathognomonic of SAW. However, SAWs are often difficult to visualize in MRI sequences, due to their small size. The scalpel sign was present in all cases of the current study and best visualized in T2-weighted sagittal image series. Similarly, a syrinx was noted in all but one of our cases compared to 2/3 in the literature [4]. Because of the syrinx, other subtle features can be overlooked, and possibly misdiagnosed as idiopathic syringomyelia or spinal intramedullary tumors. Therefore, careful inspection of MRI images for ventral displacement of the spinal cord and/or the presence of the “scalpel sign,” in particular in cases with syringomyelia without any readily identifiable course, is essential for the diagnosis of SAW.

Moreover, there is considerable ambiguity regarding the nomenclature of other spinal arachnopathies: Spinal arachnoiditis is used as a generic term for various inflammatory conditions of the arachnoid membrane, including those observed after previous spinal trauma or surgical intervention [17,23,24]. Currently, SAW is not clearly considered as distinct pathological entity, but sometimes considered as a variant of spinal arachnoid cysts [1,25]. However, spinal arachnoid cysts harbor distinct characteristics like the usually well-defined cyst borders, a slower filling of the cyst in CT-myelography or MRI CSF flow imaging and a less focal distortion of the spinal cord without a scalpel sign [1,25–27].

Intraoperative visualization reveals a focally thickened arachnoid membrane [4,20,22,25,26,28,29]. In the vast majority of the described cases, surgical treatment consisting of intradural excision of the thickened arachnoid membrane significantly improved symptoms. Only a few previous reports have applied ioUS for SAW resection [2,3]. Our study showed the useful applicability of ioUS to visualize the web intraoperatively before dural incision and verify the restored CSF flow before closing. The surgeon can directly visualize the increased dynamic motion of spinal cord pulsations after membrane excision.

The present study supports SAW as a distinct focal arachnopathy. It shows distinct diagnostic and clinical characteristics that facilitate distinction from unspecific arachnopathies after intradural bleedings or surgeries. Another condition which can mimic imaging findings of SAW is the idiopathic spinal cord herniation (SCH) which is therefore an important differential diagnosis: While both conditions share common clinical features, subtle MRI-findings can be used to distinguish SAW from SCH. While the scalpel sign is considered pathognomic for SAW, a ventral displacement of the spinal cord with an interrupted ventral subarachnoid space and a C-shaped dorsal cord surface is characteristic for SCH [4,5,29,30]. While both entities can be treated surgically, operative SCH treatment requires the closing of the ventrally located dural defect the spinal cord herniates through [30].

Arachnoiditis ossificans is another important differential diagnosis of focal arachnopathy. It is a rare calcification of the arachnoid membrane with characteristic histopathological characteristics. It causes—similar to SAW—spinal cord compression and is mostly located in the thoracic spine with corresponding clinical symptoms [31]. Associated syringomyelia is described in a subset of these cases, but is not the norm as in

SAW [31]. Diagnostic modalities such as conventional MRI and computed tomography (CT) show an intradural, extramedullary calcified lesion compressing the spinal cord [31,32]. Similar to SAW, a history of prior spinal procedures is frequently reported in patients with this pathology [33–35]. Surgical approaches rely mainly on decompressive laminectomy with or without intradural lesion excision, which usually leads to clinical improvement [31,33,36].

Conclusion

By summarizing the cardinal features and illustrating them with radiological images and videos of ioUS and microsurgical techniques, we want to emphasize SAW being a distinct entity of focal spinal arachnopathy. Its early recognition in association with careful evaluation of radiological findings is essential to avoid delay in diagnosis. Microsurgical resection is often associated with symptom improvement and favorable long-term outcome. The routine use of ioUS helps to visualize the SAW before dural incision, as well as verifying restored CSF flow after resection.

Declarations of competing interests

The authors have no competing interests or financial disclosures in relation to this trial.

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Supplementary materials

Supplementary material associated with this article can be found in the online version at https://doi.org/10.1016/j.spinee.2021.06.018.

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