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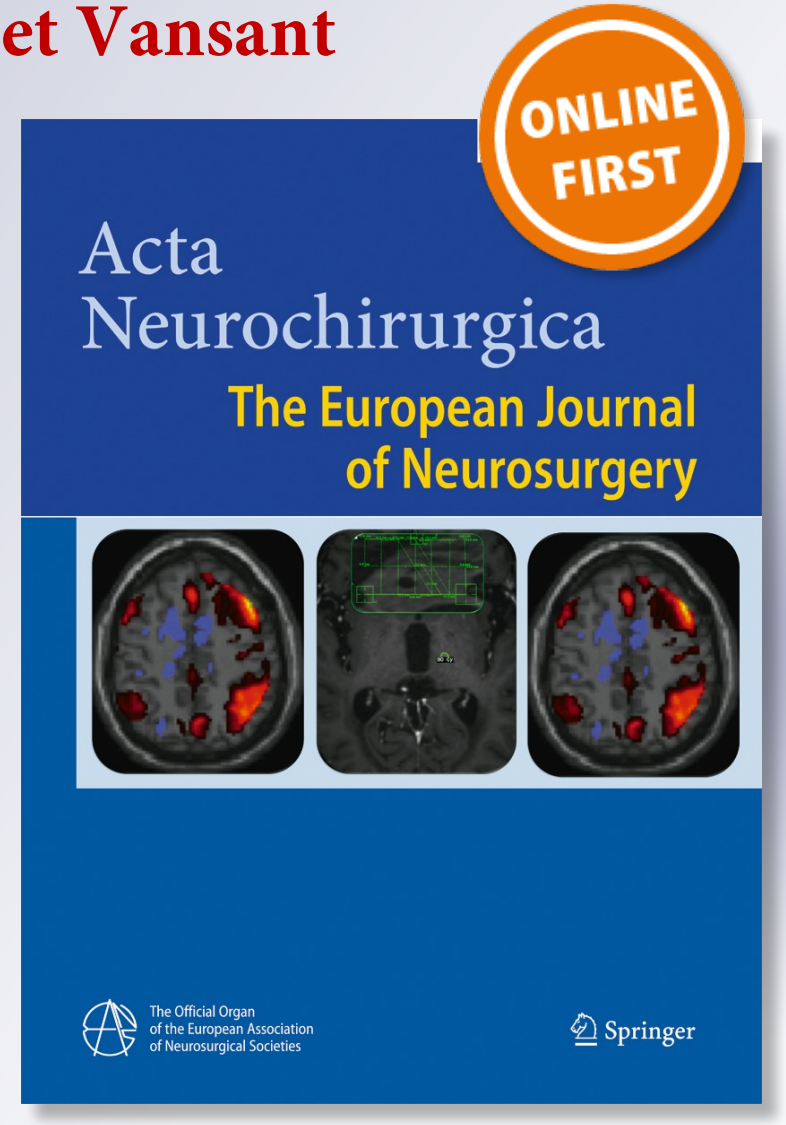

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ORIGINAL ARTICLE - SPINE

Can patients with symptomatic Tarlov cysts be differentiated from patients with specific low back pain based on comprehensive history taking?

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Abstract

Background Tarlov cysts (TCs) are expanded nerve root sheaths that occur near the dorsal root ganglion and result from increased intraspinal hydrostatic pressure. TCs most frequently affect the lumbosacral plexus and therefore may cause specific symptoms such as perineal pain and neurogenic bladder, bowel, and sphincter problems. It has been estimated that 1% of the population has symptomatic Tarlov cysts (STCs). However, STCs appear to be underdiagnosed, with the pain reported by patients commonly attributed to degenerative alterations seen on MRI. The aim of the present study is to investigate the utility of a comprehensive questionnaire for use by physicians in establishing the diagnosis of STCs.

Methods We compared questionnaire responses regarding patient history between 33 patients diagnosed with symptomatic TCs and 42 patients with chronic low back pain and sciatica due to disc problems or degenerative or inflammatory disorders. The diagnosis of STCs was confirmed using nerve conduction studies (NCS) and electromyography (EMG) of the sacral myotomes by an expert neurophysiologist.

Results The questionnaire responses revealed specific differences in perineal symptoms (perineal pain, dyspareunia, coccygodynia), bowel symptoms (constipation, diarrhea), bladder symptoms (hesitation, retention, frequency), and anal sphincter problems (anal pain, mild fecal incontinence). Additionally, sitting, walking, and straining aggravated pain more frequently in STC patients, and STC patients were more often forced to stop working and/or reduce their social activities.

Conclusions Including the above-listed items in the patient history might facilitate differentiation of low back pain and sciatica due to STCs from that due to disc problems or degenerative or inflammatory disorders.

Keywords Tarlov cysts · Perineal pain · Non-specific low back pain · Ischialgia

Introduction

Sacral TCs originate from dilating nerve root sheets and are caused by pathological increases in cerebrospinal pressure, which adds to the physiological hydrostatic pressure in the sacral nerve roots leaving the dural sac [4, 21, 22].

It has been estimated that 5% of the population harbor TCs and that 25% of these are symptomatic; thus, TCs affect 1% of the population, mostly women (86%) [12, 18, 19]. However, because of the persistent belief that TCs are clinically irrelevant, STCs are systematically overlooked [2, 4–6, 9, 10, 14, 16, 18, 23].

Generally, the diagnosis of STCs is established when (1) all other causes of low back pain or sciatica are ruled out, (2) there are no other alterations on MRI that might explain the

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low back pain and/or sciatica, and (3) there are TCs that are on the same side as the radiculopathy [15].

However, the diagnosis is less clear if the patient with back pain or radiculopathy harbors smaller TCs and/or if significant concomitant degenerative or inflammatory alterations are seen on MRI.

Yet, STCs are typically associated with perineal or pelvic pain and paresthesia as well as neurogenic bladder, bowel, and/or sphincter symptoms [1, 3–5, 15, 21]. However, when taking patient history, the focus is typically on the back pain and the ischialgia, with these other complaints not being included.

NCS/EMG of the sacral nerve root myotomes can help confirm a diagnosis of STCs [6, 7]. However, a neurophysiologist skilled in perineal EMG is not always available.

Therefore, the aim of this study is to investigate whether comprehensive history taking reveals significantly different responses between patients with an established diagnosis of STCs and those without TCs who are suffering from long-standing back pain and/or sciatica due to recurring disc herniations, lumbar degenerative disorders, or inflammatory disease. Increased knowledge of the symptoms might offer the patient a more accurate diagnosis.

Materials and methods

The entire study was conducted with an explorative retrospective series of cases and is based on data collected between July 2015 and October 2016.

The study group consisted of 33 patients (30 women and 3 men) diagnosed with STCs and with a mean age of 49.8 ± 9.5 years. As there is no consensus on the size at which TCs become symptomatic, we included only those patients with TCs at least 15 mm in diameter. The diagnosis of STCs was established if at least one cyst was located on the same side as the patient's pain and after all other causes of low back pain were ruled out. Subsequently, the diagnosis was confirmed in all patients using NCS/EMG of the lumbar and sacral nerve root myotomes.

The electrodiagnostic tests consisted of conduction studies performed on the sensory sural nerves (which contain fibers from nerve roots S1 and S2) and tests of the S1 Hoffmann reflex (the electrophysiological equivalent of the Achilles tendon reflex). The needle EMG included the S1 (gastrocnemius muscle medial head), S2 (tibial nerve-innervated intrinsic foot muscles), and the S3–S4 myotomes (external anal sphincter). Additionally, the S3–S4 ano-anal reflex (the electrophysiological equivalent of the ano-cutaneous reflex) was analyzed. This reflex requires an intact reflex arc that consists of both afferent and efferent limbs, as it is meant to prevent fecal incontinence. When using needle EMG on the anal sphincter, only the efferent (motor) limbs are evaluated, whereas when analyzing the ano-anal reflex, the afferent (sensory) limbs are also assessed. The EMG was performed by an expert electrophysiologist who was skilled in perineal EMG.

The control group was matched for sex and age and consisted of 42 consecutively selected patients (38 women and 4 men) diagnosed with low back pain due to recurrent disc herniation, disc and/or facet degeneration, or spondyloarthropathy. Their mean age was 49.4 ± 12.4 years. The MRI of the lumbar and sacral spine of the control patients revealed no TCs. Therefore, NCS/EMG of the perineum was not performed.

The history of each patient was taken by the same expert physiatrist. The physiatrist inquired about specific symptoms associated with STCs using a questionnaire based on a modified version of the International Tarlov Cyst Questionnaire (F. Feigenbaum and D. West, unpublished). The questionnaire includes questions regarding perineal symptoms; bladder, bowel, and sphincter symptoms; lower limb pain and paresthesia; headache; aggravating and alleviating factors; and social and professional functioning.

The obtained data were entered into an Excel spreadsheet. Fisher's exact test was used to compare the questionnaire scores between the patient groups. All of the tests were two-sided. Analyses were performed using SAS software (version 9.4).

Results

Statistically significant differences in symptoms between the two groups were found and are listed in Table 1.

Compared with the control patients, the STC patients showed significantly more frequent pain and paresthesia in the feet and the legs and subjective weakness in the legs.

STC patients also reported significantly more perineal pain and paresthesia, dyspareunia, and coccygodynia compared with the control group.

Moreover, bowel dysfunction, anal and urinary sphincter problems were reported significantly more often by the STC patients than by control patients.

Additionally, the STC group reported headache significantly more frequently.

Additionally, physical exertion and sitting aggravated the pain and lying down alleviated the pain more frequently in STC patients than in the control patients.

STCs have significant impacts on social and professional functioning. Compared to the control patients, STC patients reduced their social activities significantly more often and they were forced to stop working more frequently.

Discussion

As approximately 5% of the population harbor TCs, the combination of disc protrusion or degenerative alterations or

Table 1 Comparison of history findings for the STC group ($N = 33$) and the control group ($N = 42$)

	Symptoms	Patients with STCs (%)	Patients with specific low back pain (%)	p value
Symptoms in the lower limbs	Pain in one foot	51	21	0.001***
	Pain in both feet	24	10	0.001***
	Paresthesia in one foot	50	39	0.002**
	Paresthesia in both feet	31	5	0.002**
	Pain in one leg	45	33	0.001***
	Pain in both legs	25	5	0.001***
	Slower walking pace	76	54	0.06 NS
	Subjective weakness in the legs	72	28	< 0.001**
	Restless legs	61	49	0.4 NS
Pelvic symptoms	Perineal pain	24	7	0.05*
	Perineal paresthesia	24	5	0.02*
	Dyspareunia (in the sexually active patients)	37 (10/27)	5 (2/37)	0.002**
	Coccygodynia	49	17	0.005**
	Saddle pain when cycling	58	31	0.04*
	Pain in the buttocks	64	64	0.8 NS
	Pain in the groin	21	27	1.0 NS
Bowel symptoms and anal sphincter problems	Constipation	63	33	0.008**
	Diarrhea	63	40	0.05*
	Intestinal cramps	70	45	0.06 NS
	Anal pain	57	10	< 0.001***
	Mild fecal incontinence	55	7	< 0.001***
Bladder symptoms and urinary sphincter problems	Hesitation	38	5	< 0.001***
	Retention	55	17	0.003**
	Valsalva voiding	45	33	0.5 NS
	Urinary incontinence	52	29	0.1 NS
	Frequency	69	38	0.01*
	Urgency	57	34	0.1 NS
	Lower tract urinary infections	35	31	0.3 NS
Other symptoms	Headache	70	45	0.05*
Pain-aggravating factors	Long walks	75	34	< 0.001***
	Straining	84	61	0.04*
	Sitting	100	74	0.002**
	Standing	91	73	0.07 NS
	Valsalva maneuvers	37	29	0.6 NS
	Nocturnal pain	72	76	0.8 NS
	Previous trauma	21	9	0.3 NS
	Pregnancy	23	23	1.0 NS
Pain-alleviating factors	Lying down	90	63	0.02*
	Changing body position	71	64	0.6 NS
Social and professional functioning	Forced to reduce social activities	78	18	< 0.001***
	Forced to change to a physically less demanding work	33	13	0.06 NS
	Forced to stop working	54	18	0.003**

Significantly different at $*p \leq 0.05$ $**p \leq 0.01$ $***p \leq 0.001$

STCs symptomatic Tarlov cysts

inflammatory signs with TCs on the MRI of patients with chronic low back pain is common. From the TCs seen on MRI, approximately one out of four produce symptoms, thus affecting as much as 1% of the population [12, 18, 19]. It is not always clear which TCs are symptomatic and which are not. Because TCs have been assumed to be clinically irrelevant, it is typically concluded that concomitant alterations are responsible for the pain. However, this conclusion is not always accurate, as not all degenerative alterations on the lumbar spine are symptomatic. In an MRI study performed by Jensen et al. [8], disc bulging and disc protrusions were found in 52% of 98 asymptomatic volunteers between 20 and 80 years of age.

Moreover, TCs are often multiple and small and frequently appear bilaterally; furthermore, it is not always the largest cyst that causes the worst symptoms [7, 21]. Therefore, there may be a lack of correlation between radiologic findings (the size and side of the cyst) and symptomatology [17]. Consequently, STC patients typically have a long history of back pain and ischialgia and may have undergone several technical investigations and interventions [5].

As the clinical presentation of STCs is generally unknown to most physicians, we aimed to investigate whether history taking can reveal significantly different responses between patients with STCs and patients suffering from specific chronic low back pain.

Symptoms in the lower limbs

In the STC patients in our study, pain and subjective weakness in the legs and pain and paresthesia in the feet were reported significantly more often than in the control group.

STCs affect mainly the S1, S2, and S3 nerve roots. The S1 and S2 nerve roots supply the dermatomes and myotomes of the legs. The S2 nerve roots supply the intrinsic foot muscles and the sensory innervation of the soles of the feet. In a case report, Acosta et al. [1] described a diminished sensory perception to pinprick on the soles of the feet and the S1–S2 dermatomes of the legs in STC patients. Murphy reported S1–S2 neuropathy in 87% of 2013 STC patients [15].

Additionally, in this study, bilateral symptoms occurred significantly more frequently in the STC patients than in the patients with specific low back pain. This finding might be due to the increased hydrostatic pressure, affecting several nerve roots on both sides at the same time. Additionally, not only large TCs can cause symptoms, as one or more smaller cysts can also be problematic [6, 10, 20, 21].

Pelvic symptoms

Perineal pain and paresthesia, dyspareunia, coccygodynia, and saddle pain when cycling were significantly more common in the STC group than in the low-back-pain group. The perineum is innervated by nerve roots S3, S4, and S5.

Other studies have also reported perineal symptoms in STC patients. Van de Kelft and Van Vyve reported that among patients complaining of unexplained perineal pain, 75% harbored TCs [23]. Murphy et al. [15] reported that 209 of 213 STC patients with TCs had perineal and/or pelvic pain.

Bowel symptoms and anal sphincter problems

Constipation and diarrhea were more frequently reported in the STC group than in the control group.

The S2, S3, and S4 nerve roots provide the autonomic innervation of the colon transversus, the descending colon, and the sigmoid. Irritation of the sacral nerves may lead to irritable bowel presenting with diarrhea, whereas insufficient innervation of the smooth muscles of the colon causes constipation. Constipation in STC patients has been reported by other authors [1, 4].

Additionally, in our study, anal sphincter pain (cramps, stabbing, or painful pressure) and mild fecal incontinence were significantly more often reported by STC patients than by control patients. Nerve roots S2, S3, and S4 provide the motor innervation of the external anal sphincter. Murphy found mild fecal incontinence and sphincter tone disturbances in 29% of 213 STC patients [15].

Bladder symptoms and urinary sphincter problems

Symptoms of bladder dysfunction such as hesitation, retention, and urinary frequency were reported significantly more frequently by the STC patients than by the control group.

The detrusor of the bladder is innervated by the sacral nerve roots. Compression of the sensory and motor nerve root fibers inside the nerve root cysts can result in an overactive detrusor and/or insufficient muscle strength of the detrusor, which compromises bladder voiding. An overactive detrusor muscle can cause urinary frequency, whereas a weak detrusor muscle can cause urinary hesitation and retention [1, 4, 11].

Additionally, nerve roots S2, S3, and S4 supply the motor innervation of the urinary sphincter. Therefore, stress incontinence may occur in STC patients. Murphy et al. [15] found urinary sphincter disturbances in 43.2% of patients with TCs. However, in our study the difference between study groups was not significant.

Headache

The patient histories showed that headache was prevalent in 70% of the STC patients; this prevalence was significantly higher than that of the control group (45%). Few authors have reported headache in STC patients, likely because the association with STCs has been unclear. These headaches might potentially be explained by increased intracranial pressure associated with the pathologically increased intraspinal pressure in STC patients.

Pain-aggravating and pain-alleviating factors

All the patients in the STC group had difficulties when sitting, and almost all had difficulties when standing. Indeed, the pain from STCs can be exacerbated when walking, sitting, or standing and when exerting physical effort, all of which increase spinal pressure [3, 16].

This study also demonstrated that STC patients experienced pain reduction when lying down significantly more often than did the specific low-back-pain group. This result might reflect a decrease in hydrostatic pressure when lying down, which reduces the pressure inside the cysts [4, 21, 23].

When performing a Valsalva maneuver, the two patient groups reported pain aggravation equally. A Valsalva maneuver increases the pressure inside the cerebrospinal canal and in the nucleus pulposus of the lumbar intervertebral discs, both of which may provoke pain.

It has been reported that the first symptoms in STC patients occur or are aggravated after a trauma [13]. However, our study revealed no significant difference regarding previous trauma between the STCs and the low-back-pain groups.

Social and professional functioning

The most striking finding of the present study was that the STC patients reported the need to stop working and to limit social activity significantly more often than did the control group. Jobs that required sitting and straining were the most affected.

The STC patients could not participate in sitting-related social activities such as going to a movie or having dinner with friends. Being unable to work and/or participate in social activities is largely responsible for limiting their quality of life [4, 5, 15].

In this study, the decision for intervention was based on whether the patient with STCs had debilitating pain, developed fecal or urinary incontinence, or had progressive motor dysfunction.

There are limitations to our study. One limitation is that the study was a retrospective study. In addition, the physician taking the history and the physician performing NCS/EMG were not blinded to the presence of TCs on MRI. Furthermore, EMG of the sacral nerve root myotomes was not performed in the control patients and thus could not be compared with the STC patients. Further blinded prospective studies of larger groups of patients are required. Including clinical examination with history taking in the diagnostic workup would be useful for developing easy-to-use clinical guidelines to establish the diagnosis of STCs.

Conclusion

This exploratory, retrospective analysis of a series of cases demonstrates that specific differences in clinical presentation between STCs and other back disorders can be detected by taking a comprehensive history that includes specific questions about STC symptoms.

These findings are relevant to establish a surgical indication in patients with TCs. A history revealing several of these symptoms, especially those that are due to sacral nerve involvement, may guide the diagnosis towards STCs. In STC patients, a surgical intervention for coexisting degenerative disorders would likely be less successful than it would be in patients who have none of these symptoms. Such patients might benefit more from a surgical intervention for their TCs.

If TCs are seen on the MRI of a patient with low back pain and/or ischialgia, a comprehensive history taking should be performed to distinguish between asymptomatic and symptomatic TCs.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval For this type of study, formal consent is not required.

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