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Urogenital symptoms in women with Tarlov cysts

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Abstract

Aim—To describe the clinical findings and urogenital symptoms associated with sacral perineural cysts (Tarlov cysts).

Methods—A retrospective chart review including 65 female patients with Tarlov cysts was completed. Clinical findings were collected from a database of subjects seen in our institution's urogynecology and neurosurgery clinics between 2004 and 2015. A statistical analysis was performed to test for any correlation between cyst size or location, and patient symptoms or exam findings.

Results—Tarlov cysts were most commonly located from S2 to S3 (73%), and ranged in size from 1-2cm (55%). Frequently reported symptoms included lower back pain (83%, 95% confidence interval [CI] 0.71-0.91), lower extremity radiculopathy (75%, CI 0.63-0.85), positional pain (62%, CI 0.50-0.73), urinary urgency (54%, CI 0.41-0.66), and urinary frequency (48%, CI 0.35-0.61). Common urodynamic findings included: an early sensation of filling (70%), involuntary detrusor contractions (33%), urethral instability (33%), and stress urinary incontinence (33%). A statistical analysis comparing cyst size and location to clinical findings was significant for a correlation between an S2 location and CNS symptoms (p=0.02), larger cyst size and urinary dysfunction (p=0.05), and smaller cyst size and an early sensation of filling (p=0.05).

Conclusion—Patients with symptomatic sacral Tarlov cysts frequently report pain and neuropathy related to the lower back, pelvis, and urogenital system. As compared to the general population urinary urgency and urodynamic findings associated with urgency, were more frequent in our patient sample. These findings suggest that Tarlov cysts may have a clinically significant impact on urogenital function.

Keywords

urodynamics; urinary incontinence; neurology; sexual dysfunction; pelvic pain

INTRODUCTION

Tarlov cysts, or perineural cysts, are cerebrospinal fluid (CSF) filled outpouchings of spinal nerve root sheaths that most commonly arise along the sacral spine. The incidence of Tarlov

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cysts ranges from 1-9%, and they are more prevalent in women.^{1–4} While they were historically described as incidental findings, several case reports have described symptoms associated with Tarlov cysts including lower back pain, pelvic pain, lower urinary tract dysfunction, and sexual dysfunction.^{5–7}

Whether these cysts are actually the source of symptoms has been a topic of debate. While prior publications estimated that less than 1% of Tarlov cysts produce symptoms², a more recent prospective trial proved that over 80% of patients undergoing cyst aspiration reported satisfactory relief of local back pain, perineal pain, and urogenital dysfunction.⁸ This new data suggests that for certain patients Tarlov cysts do have a significant clinical impact.

We hypothesized that there is an association between large sacral Tarlov cysts, pelvic floor symptoms, and urodynamic findings. In order to better understand this association, we created a database of patients evaluated in our institution's urogynecology and neurosurgery clinics, and tracked their clinical course over time. The goal of this study is to describe the initial presenting symptoms and physical exam findings within that group of patients in order to more clearly define the potential impact of Tarlov Cysts on sacral nerve roots.

MATERIALS & METHODS

After obtaining Institutional Review Board approval from the University of California Davis, we created a database of female patients with a Tarlov cyst confirmed by CT or MRI. All patients evaluated in our institution's urogynecology and/or neurosurgery clinics between July 2004 and October 2015 were included. Patients were excluded from this case series if they had a history of prior incontinence surgery or neurologic malformation.

Demographic information, indication for clinic visit (back pain radicular pain, pelvic pain, sexual dysfunction, urinary dysfunction, or bowel dysfunction), chief complaint, symptoms documented in the history, physical exam findings, and radiologic findings were abstracted from the medical records. Specifically, we reviewed and recorded findings from any visits to the departments of obstetrics and gynecology, urogynecology, neurology, neurosurgery, orthopedic surgery and radiology. Tarlov cyst size and location were abstracted from the radiology report when present; when not documented, a study investigator reviewed MRI images. The widest diameter of each Tarlov cyst in the sagittal plane was documented as cyst size, as is consistent with our institution's standard radiology practice. Patients with multiple cysts in different locations, or a large cyst spanning two locations were excluded from our statistical univariate analysis.

We used descriptive statistics to calculate the mean value for each variable (cyst characteristics, patient reported symptoms, and urodynamic findings) with 95% confidence intervals. In a subset of patients with one Tarlov cyst at a single nerve root location, we additionally performed Fisher's exact and Satterthwaite t tests to test for associations between cyst location vs. clinical findings, and cyst size vs. clinical findings respectively. Clinical findings included patient reported symptoms, urodynamic findings, and lower extremity numbness or weakness detected upon physical exam. Symptoms reported by patients at any clinic visit (prior to surgery if performed) were grouped into five categories

(lumbosacral, urinary, bowel, central nervous system, and sexual symptoms). A p-value of 0.05 or less was considered significant. SAS® software version 9.4 (SAS Institute, Cary, NC) was used in all analyses.

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RESULTS

A total of 65 patients met our study inclusion criteria. Participants had a mean age of 53 (range 25 to 80). Table 1 contains demographic information. Sixty-five percent of patients presented to urogynecology and 92% to neurosurgery clinic. Fifty-seven percent underwent evaluation in both clinics.

All patients had at least one Tarlov cyst in the sacral location, and cysts were most commonly located at S2 (42%). The majority of patients had a single Tarlov cyst (68%). Mean cyst size was 17mm (range 5 to 50mm). Ninety-one percent of cysts were larger than 10mm (table 2).

Table 3 and 4 summarize patient findings upon initial clinical evaluation. The most frequently reported symptoms included lower back pain (83%), lower extremity pain (75%), pain that arises when moving to a sitting or standing position (62%), urinary urgency (54%) and urinary frequency (48%). Individual symptoms were separated into broader categories as listed in table 3. Lumbosacral symptoms were reported by 95% of patients, and urinary dysfunction was reported by 78% of patients. Among the 40 patients who underwent urodynamic testing, the most frequent finding was that of an early sensation of bladder filling (70%).

Forty-five patients in our study group had a single cyst impacting either the S1, S2, or S3 nerve root. These patients were included in our univariate analysis (table 5). Cyst location was significantly associated with central nervous system (CNS) symptoms (p=0.02). Symptoms categorized as CNS are listed in Table 3. Forty-eight percent of women with cysts at S2, 9% with cysts at S3, and zero patients with cysts from L-S1 reported CNS symptoms. Cyst location was not associated with any other patient symptoms, urodynamic findings, or physical exam findings.

Regarding cyst size, there were no significant associations between the size of a Tarlov cyst and bowel dysfunction (p=0.83), sexual symptoms (p=0.92), CNS symptoms (p=0.33), lumbosacral symptoms (p=0.09), or lower extremity weakness or numbness (p=0.52). There was a significant association, however, between urinary symptoms and cyst size (p=0.05). Tarlov cysts were an average of 4.3mm larger in women reporting urinary symptoms. These data are summarized in table 6.

Twenty-eight of the 45 patients included in our univariate analysis underwent urodynamic testing (table 6). Among this subset of patients, early sensation of filling was associated with cyst size (p=0.05). Those with an early sensation of filling had a Tarlov cyst measuring 5.7mm smaller on average than patients without an early sensation of filling. There were no

significant associations between cyst size and urethral instability (p=0.59), or involuntary detrusor contractions (p=0.97).

DISCUSSION

The goal of this case series is to provide further insight into the clinical presentation of patients with sacral Tarlov cysts. Despite a common report of urogenital symptoms in patients with sacral Tarlov cysts, there is very little recognition of these cysts within the gynecologic literature. A Pubmed search (English language; 1948-2016; search terms: "Tarlov cyst" and "sacral") revealed numerous case reports of symptomatic Tarlov cysts;^{5–7} only three of these were published in journals related to gynecology including one case of a Tarlov cyst mistaken for an adnexal mass and two case series of persistent genital arousal disorder associated with sacral Tarlov cysts.^{6, 9, 10} This case series is the largest to date focusing solely on female patients and the first to include a detailed description of urodynamic findings in a subset of those patients.

In agreement with previously published data, we found that the majority of cysts were located at S2 and ranged in size from 1-2cm (table 2).⁵ The most common symptoms were similar to those reported in prior case reports, and included lower back pain, lower extremity radiculopathy, positional pain, and urinary urgency and frequency (table 3).^{2,5,11}

The mechanism by which Tarlov cysts produce symptoms remains a topic of debate. The most popular theory is that of the "ball valve" mechanism.² Supporters of this theory believe that Tarlov cysts form when a defect in the perineural sheath acts as a one-way valve, allowing CSF to become trapped within the cyst. Eventually, enough CSF accumulates to create a high-pressure environment that can compress adjacent nerve roots (figure 1).

Treatment options for Tarlov cysts range from conservative approaches with analgesics and physical therapy to invasive techniques such as CT guided cyst aspiration⁸, or sacral laminectomy with cyst fenestration, resection or imbrication.^{12,13} Postoperative outcomes vary widely, ranging from 38-100% symptomatic relief.¹¹ Serious complications such as meningitis and CSF leaks have been reported. Prior to proceeding with invasive treatment, it is therefore imperative for a physician to correlate a patient's symptoms to the size and location of their spinal cyst.⁷

In order to determine whether cyst size or location correlated with the clinical findings in our study, we performed a statistical analysis on a subset of patients whose cyst was isolated to a single nerve root (N=45). From this analysis, we found a strong association between CNS symptoms and cysts at S2 (table 5). This finding is surprising given that CNS symptoms are thought to arise due to leakage of cerebral spinal fluid into a cyst, which occurs regardless of cyst location. Because S2 was the most common location for a Tarlov cyst in our study, and the majority of patients were evaluated by neurosurgeons who are more likely to elicit CNS symptoms, this association may be the result of selection bias. Prospective studies investigating pre and post-op symptoms are needed in order to more clearly correlate patient symptoms with cyst location.

In regards to cyst size, we found that larger cysts were strongly associated with urinary dysfunction (table 6). Urinary dysfunction included patient reported symptoms of urgency, frequency, incontinence, hesitancy, and incomplete void. These findings lend statistical support to prior postulations that large cysts (>1cm) are more clinically significant⁷ and more likely to impact urogenital function.

Interestingly, while the prevalence of urinary urge incontinence in the general population is estimated to be 1-7%,¹⁴ in our case series 31% of patients reported urge incontinence. In addition, urinary urgency (with or without incontinence) was the fourth most frequently reported symptom followed by urinary frequency (table 3). This is in contrast to patient reported stress incontinence (25%), which was similar to the general population prevalence of 10-39%.¹⁴

To further investigate any relationship between Tarlov cysts and lower urinary tract dysfunction, we recorded the urodynamic findings of all patients who underwent this testing (table 4). Notably, the prevalence of both detrusor overactivity and an early sensation of filling were higher than anticipated in our case series (33% and 70% respectively). When looking at cyst size specifically, we found that patients with an early sensation of filling had Tarlov cysts 5.7mm smaller on average than those with a Tarlov cyst and no early sensation of filling. One explanation for this finding may be that larger Tarlov cysts are more likely to compress the afferent nerve roots involved in sensation of bladder filling. This would result in decreased sensation. In contrast, a smaller cyst would be less likely to compress nerve roots, resulting in a higher likelihood of enhanced sensation of filling during urodynamic testing. The potential for a Tarlov cyst to either enhance or dampen sensation is currently unknown. More research is needed in order to better clarify this finding. Finally, in regards to cyst location, we found no association between nerve root location and urodynamic findings (table 5).

We found that a larger than anticipated percent of patients in our cohort had urethral instability on urodynamic testing. In their study focusing on the beneficial effects of sacral nerve stimulation on urethral instability, Groenendijk et al¹⁵ point out that the prevalence of urethral instability ranges from 12-16%. Our finding of 33% prevalence is substantially greater. We did not find any significant relationship between cyst size or location and urethral instability in our statistical analysis. It is important to point out that our sample size of patients meeting criteria for a statistical urodynamic analysis was small (N=28). Therefore, larger studies are needed in order to draw further conclusions regarding any significant relationship between urodynamic findings and Tarlov cysts.

This study has several limitations. First, this is a case series and our data is not representative of a random sample. While we identified several potentially important associations between Tarlov cysts and urinary symptoms, we cannot draw conclusions about causality. Additionally, given that all patients in this study were evaluated by either a neurosurgeon or urogynecologist, a certain degree of bias may exist due to the likelihood of patients to report sensitive issues such as urinary dysfunction to a sub-specialist.¹⁴ Clinicians may also selectively record patient symptoms depending on their own sub-specialty based bias. We attempted to correct for this by grouping symptoms into broader categories. By pooling

similar symptoms, we were also able to increase the sample size of each clinical finding. This allowed us to complete a statistical analysis. However it is important to note that the outcomes of our statistical analysis may be influenced by our method of symptom categorization. Finally, confounding variables including stage II or greater prolapse (8%), or a history of minor spinal trauma (25%) may have influenced our outcomes.

Despite these limitations, this case series adds a substantial set of observational data to the existing evidence regarding the clinical impact of Tarlov cysts. In patients with Tarlov cysts, and especially cysts greater than1cm in size, our findings suggest that a correlation may exist between the presence of a sacral spinal cyst and urinary dysfunction. Comparative studies are needed however, in order to more clearly define this relationship.

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FIGURE 1.

Sagittal T2 weighted MRI of a Tarlov cyst (arrow) spanning the S2-S3 nerve root (A), and corresponding axial image showing the proximity of the nerve root (arrowhead) to the Tarlov cyst (B).

Demographic Characteristics for Patients with Symptomatic Sacral Tarlov Cysts (N=65)

Characteristic	
Age (y)	53±12
Parity	2±1.2
BMI, kg/m ²	27±8.2
Race/Ethnicity	
White, non-hispanic	54 (85)
White, Hispanic	4 (6)
Black, non-hispanic	3 (5)
Asian or Pacific Islander	2 (3)
Other	2 (3)
Native American	0
Black, Hispanic	0
Medical or Surgical History	
Smoking History	26 (40)
History of trauma affecting spine	16 (25)
Connective tissue disorder	4 (6)
POPQ [*] exam completed	44 (68)
Late Stage II or greater †	5 (8)

Data are mean \pm standard deviation or n (%)

Pelvic Organ Prolapse Quantification System (POP-Q) measurements were obtained as defined by the International Continence Societywww.ics.org/terminology

 \dot{f} Late stage II is defined as stage II prolapse with the leading edge > 0cm from the hymen as defined by the POPQ system

Tarlov Cyst Characteristics

Characteristics	No. Patients (%) N=65
Nerve root location*	
L1-5	3 (4)
L5-S1	2 (3)
S1	6 (8)
S1-2	8 (10)
S1-3	1 (1)
S2	33 (42)
S2-3	10 (13)
S3	14 (18)
S3-4	1 (1)
Number of cysts	
1	44 (68)
2	9 (14)
>2	12 (18)
Size of largest cyst	
<1cm	6 (9)
1-2cm	36 (55)
>2cm	23 (35)

* Several patients had multiple cysts in different locations. All patients had at least one sacral cyst. Therefore the total number of patients under nerve root location is greater than 65.

Symptoms reported by patients with sacral Tarlov cysts*

SYMPTOM	No. Patients (N= 65)	%	CI
Lumbosacral Symptoms	62	95	
Low Back Pain	54	83	(0.71-0.91)
LE Radicular Pain *	49	75	(0.63-0.85)
Positional Pain ^{\dagger}	40	62	(0.50-0.73)
Sacral & Buttock Pain	24	37	(0.25-0.50)
LE numbness *	25	38	(0.26-0.50)
Perineal Pain or numbness [‡]	20	31	(0.20-0.43)
LE Weakness *	18	28	(0.17-0.40)
Pelvic and groin pain	12	18	(0.10-0.30)
Urinary Dysfunction	51	78	
Urinary urgency	35	54	(0.41-0.66)
Urinary frequency	31	48	(0.35-0.61)
Urge incontinence	20	31	(0.20-0.43)
Incomplete Void	19	29	(0.19-0.42)
Nocturia	18	28	(0.16-0.39)
Stress incontinence	16	25	(0.16-0.39)
Other Voiding problems $^{\$}$	17	26	(0.16-0.39)
Urinary hesitancy	12	18	(0.10-0.30)
Bowel Dysfunction	36	55	
Other fecal symptoms [#]	22	34	(0.23-0.47)
Constipation	20	31	(0.19-0.42)
CNS Symptoms ¶	26	40	
Headache	23	35	(0.24-0.48)
Dizziness & balance problems	6	9	(0.04-0.19)
Neck Pain	2	3	(0.00-0.11)
Sexual dysfunction	16	25	(0.15-0.37)

* LE= lower extremity, TC= Tarlov cyst

 ${}^{\dot{T}}\!Positional pain is defined as worsening pain when moving from sitting to standing or vice versa$

 \ddagger "."Perineal" includes vaginal and rectal pain and numbress.

 $^{\$}$ Other voiding problems include weak stream, dysuria, insensible loss, and incontinence not otherwise specified (NOS)

 $\overset{/\!\!/}{\operatorname{Fecal}}$ symptoms include fecal urgency and frequency, diarrhea, and incontinence of flatus

[¶]CNS= central nervous system

Urodynamic Findings in Patients with Tarlov Cysts

Urodynamic Finding	No Patients (%) N=40
Cystometry	
Early first sensation of filling *	28 (70)
Involuntary detrusor contraction	13 (33)
Evidence of Urethral Instability	13 (33)
Evidence of SUI $^{\not \tau}$	13 (33)
Intrinsic sphincter deficiency \ddagger	0

* Early first sensation was defined by ICS criteria as sensation at < 170cc.

 \dot{f} Evidence of stress urinary incontinence (SUI) defined by leakage with cough or valsalva.

 \ddagger Intrinsic sphincter deficiency defined by maximum urethral closure pressure (MUCP) less than 20cc.

Clinical Findings Compared to Tarlov Cyst Location

	No. PATIENTS (%)			
SYMPTOM	L-S1, N=5	S2, N=29	S3, N=11	P value
Lumbosacral Symptoms	5 (100)	27 (93)	10 (93)	1.0
Urinary Dysfunction	4 (80)	22 (76)	9 (82)	1.0
Bowel Dysfunction	3 (60)	17 (59)	8 (73)	0.81
CNS Symptoms*	0	14 (48)	1 (9)	0.02
Sexual Dysfunction	0	7 (24)	3 (27)	0.66
URODYNAMIC FINDING	L-S1, N=2	S2, N [‡] =19	S3, N= 7	
Early Sensation of Filling †	1 (50)	11 (58)	6 (86)	0.44
Involuntary Detrusor Contractions	0	8 (44)	3 (43)	0.69
Urethral Instability	1 (50)	5 (29)	3 (43)	0.83
PHYSICAL EXAM FINDING	L-S1, N=5	S2, N= 29	S3, N= 11	
Lower Extremity Numbness or Weakness	3 (60)	11 (38)	3 (27)	0.52

CNS= central nervous system

 \dot{f} Early sensation of filling was defined by ICS criteria as sensation at < 170cc.

 \ddagger The total number of patients that underwent specific urodynamic tests varied for patients with a cyst at S2. A total of 19 patients were evaluated for early sensation of filling. A total of 18 patients were evaluated for involuntary detrusor contractions. A total of 17 patients were evaluated for urethral instability.

Clinical Findings Compared to Tarlov Cyst Size

	AVERAGE TARLOV CYST SIZE (mm)*		
CLINICAL SYMPTOM (N=45)	SYMPTOM PRESENT, (N)	SYMPTOM ABSENT, (N)	P Value
Lumbosacral Symptoms	18 ± 8, (42)	12 ± 4 , (3)	0.09
Urinary Dysfunction	18 ± 8, (35)	14 ± 5, (10)	0.05
Bowel Dysfunction	$17 \pm 7, (28)$	17 ± 8, (17)	0.83
CNS Symptoms ^{\dagger}	16 ± 6, (15)	18 ± 8, (30)	0.33
Sexual Dysfunction	$17 \pm 7, (10)$	17 ± 8, (35)	0.92
URODYNAMIC FINDING (N=28)	FINDING PRESENT, (N)	FINDING ABSENT, (N)	
Early Sensation of Filling [‡]	15 ± 6, (18)	21 ± 7, (10)	0.05
Involuntary Detrusor Contractions	17 ± 8, (11)	17 ± 7, (16)	0.97
Urethral Instability	19 ± 10, (17)	17 ± 5, (9)	0.59
PHYSICAL EXAM FINDING (N=45)	FINDING PRESENT, (N)	FINDING ABSENT, (N)	
Lower Extremity Numbness or Weakness	18 ± 9, (17)	17 ± 6, (28)	0.52

* Average (mean) Tarlov cyst size measured in mm \pm standard deviation (SD)

 † CNS= central nervous system

^{*t*} Early sensation of filling was defined by ICS criteria as sensation at < 170cc.