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Technical Notes & Surgical Techniques

Wrapping a man-made dura around reconstructed nerve sleeve avoid residue or recurrence of sacral Tolav cysts☆

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ABSTRACT

Objective: To evaluate the therapy effect on patients with sacral extradural meningeal cysts with nerve root fibers by wrapping a man-made dura around reconstructed nerve sleeve.

Patients and methods: In order to avoid the cysts residue and recurrence postoperative, wrapping a man-made dura around reconstructed nerve sleeve, a new operating method, was performed in 24 patients with 42 cysts. After the nerve sleeve being reconstructed, wrapping and suturing a man-made dura around the nerve sleeve was performed. The Improved Japanese Orthopedic Association (IJOA) scoring system was used to evaluate preoperative and postoperative neurological function.

Results: Among 24 patients, 14 were females, and 10 males. Three-month after open surgery, no cyst residue or recurrence was grasped in any patient. The mean IJOA score was 18.8 ± 1.32 preoperatively and 19.6 ± 0.65 postoperatively, which was a significant difference between the preoperative and postoperative scores ($t = -3.82$, $P = 0.001$). These results indicate a significant improvement in neurological function after surgery.

Conclusion: Based on reconstruction the nerve sleeve, wrapping with man-made dura and forming double nerve sleeve was safety and effective operating method for treatment the sacral extradural meningeal cysts with nerve root fibers.

1. Introduction

Sacral extradural spinal meningeal cysts (SESMCs) are meningeal cysts located extradural in the sacral canal. These cysts are divided into two types: those with spinal nerve root fibers (SNRFs) and those without. SESMCs with SNRFs, also known as sacral Tarlov cysts, are characterized by collection of cerebrospinal fluid (CSF) between the endoneurium and perineurium of the nerve root sheath [1]. The size of Tarlov cyst varies from 5 mm to > 10 cm. Sacral Tarlov cysts are quite often coincidental findings when lumbosacral magnetic resonance imaging (MRI) is performed for other reasons [2]. Sacral Tarlov cysts are composed of vascularized connective tissue lined with flattened arachnoid tissue. The incidence of Tarlov cysts was estimated at approximately 5% in adults with approximately 20%–25% causing symptoms [3–5]. However, Burke JF et al. [6] considered that Tarlov cysts with an estimated prevalence between 4.6% and 9% of the general adult population. Progressive enlargement of sacral Tarlov cyst might occur. Associated symptoms include sciatic or sacral radiculopathy, sacrococcygeal and perineal pain, lower extremity and saddle distribution sensory or motor deficits, and urinary or anal sphincter dysfunction with occasional sexual impotence. Symptoms are often exacerbated by sitting, changes in position and posture from supine to standing, or Valsalva type maneuvers such as sneezing or straining to defecate [4]. Symptomatic Tarlov cyst requires surgical treatment. Open surgery and percutaneous intervention or combined two kinds methods are measures for treatment on Torlav's cysts.

Computed tomography or X-ray guided cyst drainage and fibrin glue injection are a minimally invasive procedure that aims at closure of the

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aperture between the cyst and the subarachnoid space, and as a result, prevention from refilling of the cyst [2,3,7]. However, fibrin glue application may be associated with aseptic meningitis. In advanced, the nerve root fibers can't showed on computed tomography or X-ray scans, the direct puncture procedure is blindfold and incline to injury the nerve root fibers inside the cysts. The direct puncture injury usual results in dysfunction of sensation and motion. As the injected glue became solid, the solid glue would adhere around nerve root fibers and result in iatrogenic tethered cord, and like a waved weight.

So, most surgeons advise open surgical techniques should be performed for sacral Tarlov cysts [1,2,4,6,8]. Neurosurgical techniques for symptomatic Tarlov cysts included simple decompressive laminectomy, cyst and/or nerve root excision and partial cyst removal and cyst wall closure using ligature or clipping, as well as imbrication and filling the epidural space using muscle, fat, fibrin glue, and free muscle flap. In our previous study, reconstruction of nerve root sheaths was advised to be performed to restore the normal physiological environment of the spinal nerve root fibers [8]. Oversewing of redundant cyst walls aimed to strengthen the congenital weakness, and restored the normal outer diameter of the involved nerve root sheath. However, even with appropriate operative techniques, the cyst residue and recurrence showed occasionally in postoperative MRI scans by following-up. How to prevent the sacral Tarlov cysts from residue or recurrence? Potts MB et al. [4] supported that a strategy of cyst fenestration and imbrication followed by packing of the epidural space with free autologous muscle or fat grafts. In advanced, Potts MB et al. [4] reformed with using proximally based vascularized muscle flaps mobilizing and rotating deep to the giant cavity. However, in our opinion, when the mobilized and filled muscle or fat became harden and connected tissue, which would incline to adhere the surrounding nerve root fibers sacral canal.

In order to improve this awkward condition, a new operative method was performed in this clinical study. We now report the results of our refined technique in a series of 24 consecutive patients with 42 symptomatic sacral Tarlov cysts. Based on reconstruction the nerve sleeve, wrapping with man-made dura and forming double nerve sleeve was safety and effective operating method for treatment the sacral Tarlov cysts.

2. Materials and methods

2.1. Study design

This was a prospective cohort study using clinical records. Approval for the study was obtained from the Research Ethics Board of Peking University Third Hospital. All subjects provided written informed consent for surgical operation and medical photography as well as inclusion in the prospective cohort clinical study.

2.2. Patients

From Dec. 2015 to Dec. 2017, a consecutive series of 24 patients who were diagnosed with SESMCs with SNRFs underwent nerve root sheath reconstruction. Basic demographic, clinical and radiological data were evaluated. Preoperative neuroimaging including MRI was performed in all patients. The period of symptoms before presentation was defined as the time from the onset of symptoms to the time of presentation, and was recorded in months.

2.3. Inclusion and exclusion criteria

Patients were included in the study if they met the following criteria: (i) MRI scans consistent with SESMCs with nerve root fibers, (ii) neurological symptoms attributable to SESMCs, and (iii) SESMCs with SNRFs observed on preoperative MRI scans and under the operating microscope. Patients were excluded if their symptoms could not be differentiated from lumbar spinal stenosis or lumbar intervertebral disc herniation. The clinical data were collected prospectively by clinicians who were blind to the SNRF status of the patients. The data were then analyzed using appropriate statistical methods.

2.4. Clinical presentation

The initial presenting symptoms, location of presenting symptoms, and period of symptoms before initial presentation were recorded. The presenting symptoms were categorized into (i) bowel/bladder and sexual dysfunction, (ii) lower extremity weakness, (iii) lower extremity numbness, (iv) pain, (v) bearing-down feeling, and (vi) more than one symptom. The locations of the presenting symptoms were categorized as (i) sacrococcygeal, (ii) perineum and external genitalia, (iii) lower waist, (iv) buttocks, (v) legs, or (vi) more than one region.

Abnormalities on neurological examination were classified as sensory dysfunction, lower extremity weakness, and bowel/bladder dysfunction. The IJOA scoring system was used to evaluate preoperative and postoperative neurological function. The IJOA is based on the JOA scoring system, with the addition of a score for bowel function as normal, slightly dysfunctional, severely dysfunctional, or incontinent [1,8].

2.5. Surgical techniques

Our operative technique follows the standard procedures for SESMC surgery. An incision is made according the location and size of cysts ranged from L5 to S3, and the sacral laminae are completely exposed according to the location of the SESMCs. Laminectomy is performed with a rongeur, while carefully preserving the integrity of the underlying cysts. The surgical microscope is then brought into the field. The terminal thecal sac is identified and dissected free from the overlying cysts. Each cyst is dissected from the surrounding structures to reveal its origin and relationships with the SNRFs (Fig. 1) by Dr. Sun JianJun.

After open the cyst intraoperation, the nerve root fibers were confirmed inside. If there were multiple cysts in the field of one patient, every cyst should be opened to confirm the nerve root fiber inside. The fistulae of cyst and braches of nerve root fibers inside were identified before reconstruction the nerve sleeve. The extra-wall was resected after dissociated the surrounding of cyst. The nerve sleeve was reconstructed according to the nerve root fibers and braches inside the cyst. Redundant cyst wall is shrunk using bipolar cautery. After the reconstruction of nerve sleeve, wrapping and suturing a man-made dura around the nerve sleeve was performed in following (Fig. 2). Intraoperative neurophysiological monitoring is used to differentiate SNRFs from other tissues, and electrical stimulation is used to verify that no motor nerve fibers are involved.

2.6. Characteristics of SESMCs

Under the surgical microscope, the number, maximum diameter, and types of cyst were determined. If there were multiple cysts in the sacral canal, the maximum diameter, type of cyst and surgical technique were recorded for the largest cyst. The number of nerve root fibers was counted after opened the cyst and lifting the wall of cyst.

2.7. Postoperative management and evaluation

Postoperatively, all patients were kept prone for several days. Wound healing was classified as healing well, delayed healing, or requiring debridement/suturing. MRI was performed two weeks after surgery. Postoperative radiological evaluation of the sacral canal was performed by a neuroradiologist blind to the patient's intraoperative diagnosis, and was classified as complete cyst resolution, residual cyst, or disappearance of cysts but with effusion into the canal cavity. Postoperative IJOA scores were evaluated three months after surgery.

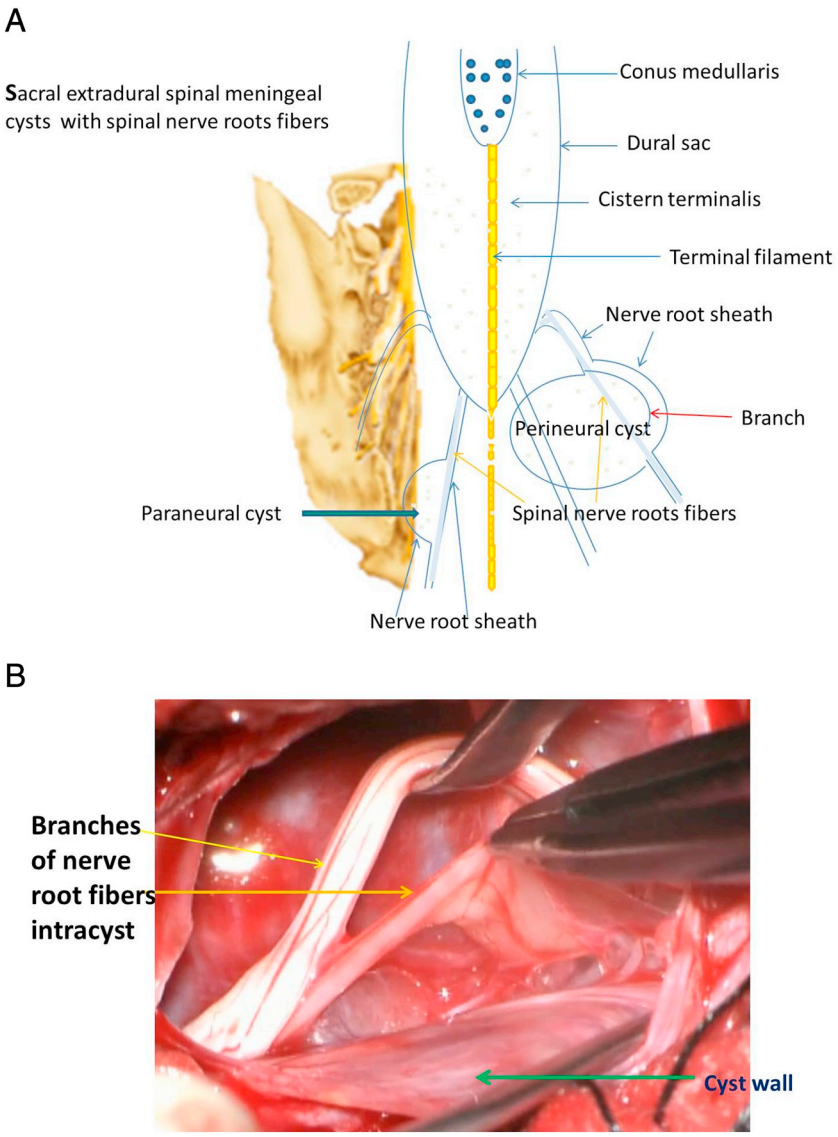


Fig. 1. Two kind of relation between sacral cyst and spinal nerve root fibers was shown in Fig. A. After open the cyst intraoperation, two nerve root fibers were confirmed inside (B).

2.8. Follow-up

All patients were followed up until March 20, 2018 by outpatient clinic visits or by telephone questionnaire. The prognosis of each patient was categorized as good or poor.

2.9. Statistical analysis

Data analysis was performed using SPSS 17.0 (SPSS, Chicago, IL, USA). Data were compared using the two-sample *t*-test for parametric data and the Mann-Whitney test for non-parametric data. Data are expressed as the mean ± standard error. *P*-values of < 0.05 were considered statistically significant.

3. Results

3.1. Demographics

The 24 patients included in this study were 10 males (41.7%) and 14 females (58.3%). Patients ranged in age from 13 to 70 years, with a mean age of 40.2 ± 14.26 years. The mean length of hospital stay was 13.6 ± 2.86 d.

3.2. Presentation

The most common presentation involved symptoms in multiple locations (45.9%, 11/24), followed by symptoms located at the lower waist (25%, 6/24), legs (12.5%, 3/24), sacrococcygeal region (8.3%, 2/24) and buttocks (8.3%, 2/24). The most common presenting symptom was pain (79.1%, 19/24), followed by more than one symptom (12.5%, 3/24), numbness (4.2%, 1/24) and a bearing-down feeling (4.2%, 1/24). The mean period of symptoms before initial presentation was 33.6 ± 46.3 months.

3.3. Nerve function

Preoperative symptoms included sensory dysfunction in 10 patients, legs weakness in 8 patients and bowel/bladder dysfunction in 6 patients. The mean preoperative IJOA score was 18.8 ± 1.32. Preoperative bowel/bladder function was recorded as normal in 18 patients, slight dysfunction in 4 patients, and severe dysfunction in 2 patients.

The mean three-month postoperative IJOA score was 19.6 ± 0.65, which was significantly different to the preoperative IJOA score ($t = -3.82$, $P = 0.001$), indicating a significant improvement in neurological function after surgery. The most significant area of improvement in neurological function was sensation ($z = -2.93$, $P = 0.003$), followed by bowel/bladder function ($z = -2.67$, $P = 0.01$). Of the 6 patients with preoperative bowel/bladder dysfunction, two still had abnormal bowel/bladder function postoperatively (one had slight dysfunction and one had severe dysfunction).

3.4. Characteristics of SESMCs

Among 42 cysts, the maximum cyst diameter ranged from 1.5 to 9 cm, with a mean maximum diameter of 3.3 ± 1.36 cm. 8 patients had a single cyst, 7 patients had two cysts, 4 patients had three cysts, and one patient had eight cysts (Fig. 3). The mean number of cysts was 2.8 ± 1.69. Only one nerve root fiber was identified in 14 cysts (one patient with one nerve root fiber, however with three branches, Fig. 4), two root fibers in 23 cysts, three fibers in 5 cysts (Fig. 5).

3.5. Postoperative course

The mean duration of prone positioning after surgery was 5.6 ± 2.36 d. Wound healing was classified as well healed in 24 patients. Three-month after open surgery, no cyst residue or recurrence was grasped in any patient on MRI scans.

3.6. Follow-up and prognosis

The mean follow-up period was 15.0 ± 11.76 months (range 3–27 months). Of the 10 patients with preoperative sensory dysfunction, 3 still had sensory dysfunction postoperatively. The sensory dysfunction was aggravated in one patient, and was unchanged in the other two patients. Seven patients experienced improved leg muscle strength, and one had remaining leg weakness. The prognosis was classified as good in 23 patients and poor in one patient. The patient with poor prognosis had severe bowel/bladder dysfunction, aggravation of sensory dysfunction, and leg weakness. Postoperative radiographic evaluation of the sacral canal was classified as complete cyst resolution in 23 patients by long-time follow-up. A De novo sacral cyst was found in left side six-month after right sacral Tarlov cyst had been operated with double sheath reconstructed method in a 29-year-old male patient.

4. Discussions

4.1. Indication for surgery

The pathophysiological mechanism underlying the formation of sacral Tarlov cysts was congenital weakness of the nerve root sheath. High hydrostatic pressure generated by upright walking and a ball-valve effect resulted in cyst enlargement. The walls of the cysts were formed by enlargement of the perineural nerve root sheaths, resulted in alteration of the environment around the spinal nerve root fibers. This alteration caused dysfunction of the involved spinal nerve root fibers, similar to axonal transport dysfunction [6]. This malfunction differed from malfunction associated with compression against adjacent bone or other nerve roots. It would be useful to determine methods of restoring the normal environment of spinal nerve root fibers by surgical treatment of sacral Tarlov cysts [8].

The following indications for surgery in a clearly symptomatic

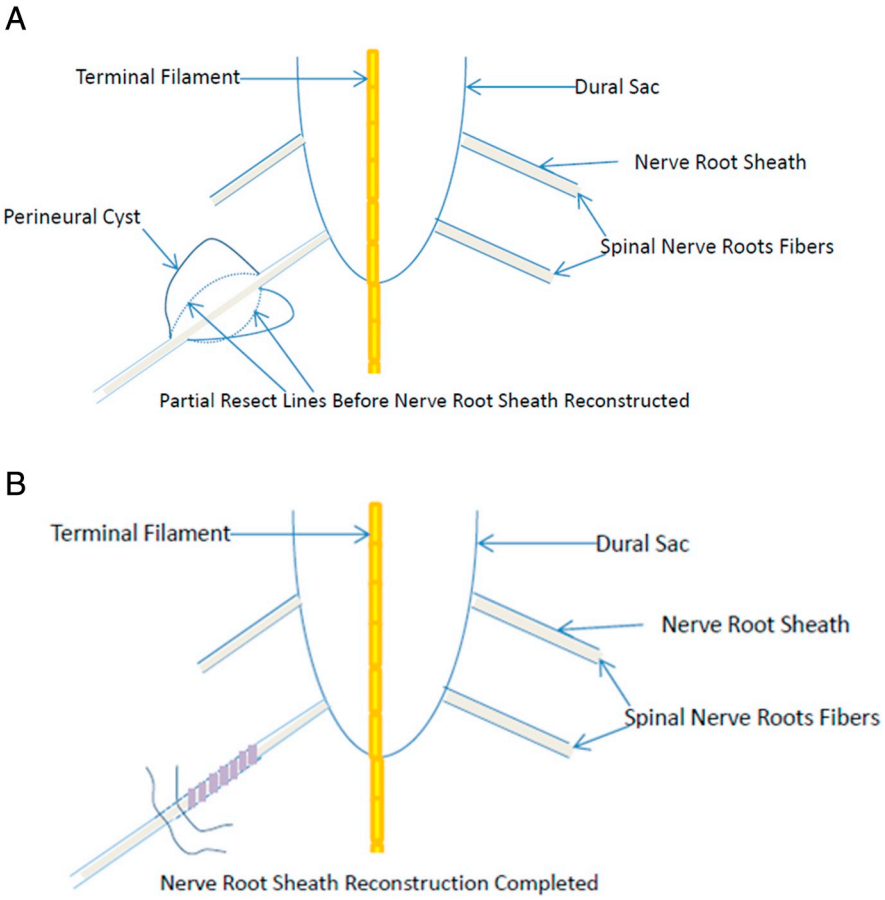


Fig. 2. The extra-wall was resected after dissociated the surrounding of cyst (A). The nerve sleeve was reconstructed according to the nerve root fibers and braches inside the cyst (B, C). After the reconstruction of nerve sleeve, wrapping and suturing a man-made dura around the nerve sleeve was performed in following (C, D).

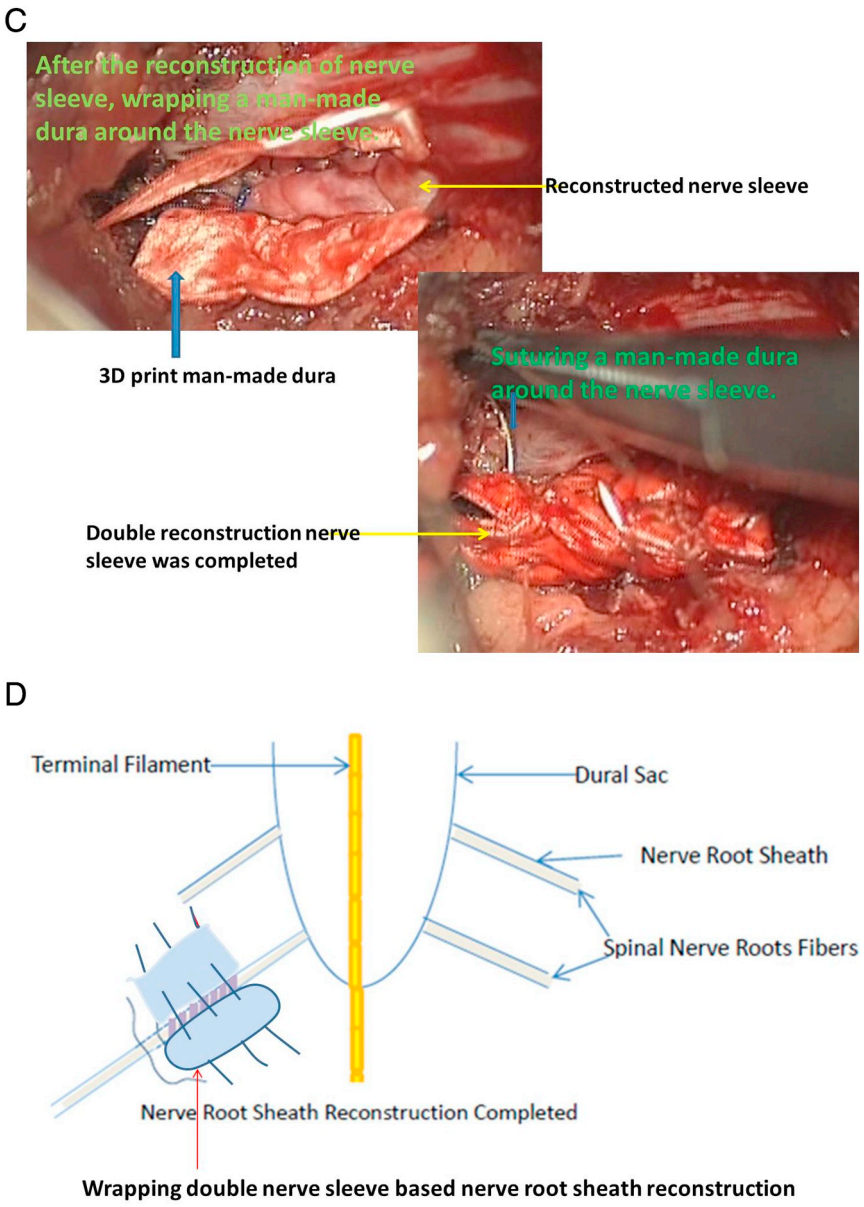


Fig. 2. (*continued*)

patient (ensuring that there were no other potential causes of the symptoms) were suggested: (i) 1 or multiple cyst(s) demonstrated on MRI, (ii) the cysts caused unendurable pain seriously affecting life and work, (iii) accompanied bladder or bowel dysfunction, (iv) symptoms consistent with cyst location, (v) symptoms developed within the last 6 months, and (vi) single, unilateral cysts larger than 2 cm [2,6].

Contraindications for surgery were small (< 1 cm) presumably incidental cysts without bone erosion, and lack of anatomic-clinical correlation (i.e., lumbar back pain only, wrong side, or wrong root level) [4].

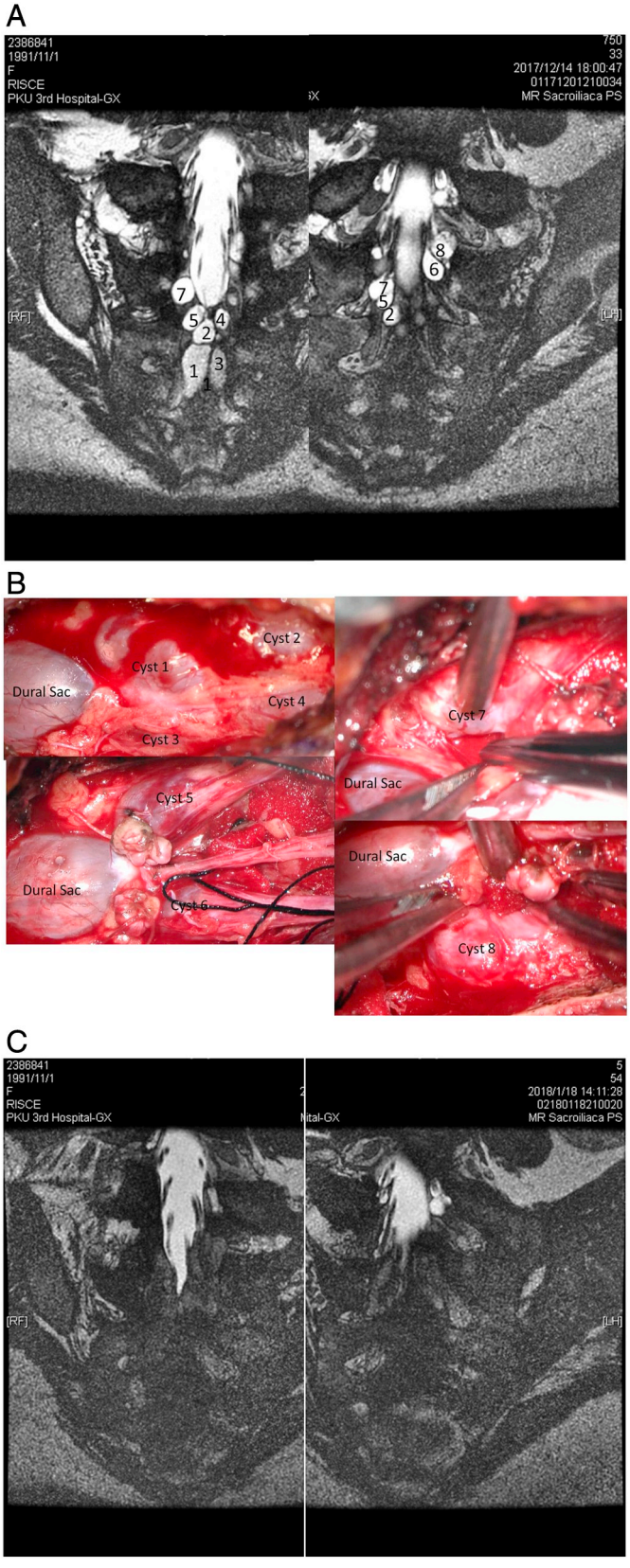
4.2. Various surgical treatments

Various microsurgical techniques had been used to treat sacral Tarlov cysts including lumbar-peritoneal and cyst-subarachnoid drainage and shunting [9,10]; bipolar cautery to shrink cysts [11]; decompressive laminectomy [12]; and laminectomy with total cyst resection [13], partial cyst wall resection, or duroplasty/plication of cyst walls [14]. The lumboperitoneal shunt procedure had produced uncertain results and carried the accompanying risk of infection. Symptoms often recurred after percutaneous cyst drainage due to recollection of the CSF. Jiang W et al. [7] and Patel MR et al. [15] advocated CT-guided percutaneous injection of fibrin gel after cyst drainage was used to treat symptomatic sacral perineural cysts with 86.8% favorable outcomes. However, Voyadzis et al. [16] did not recommend percutaneous drainage because of the potential for unexpected nerve root injury. In our previous experience, fibrin glue infusion might result in pulling of the involved nerve root fibers by the hardened glue. The symptoms of nerve stimulation were not relieved, and may even be aggravated by the fibrin glue [8]. If cysts were opened and confirmed without involved nerve roots fibers, using fibrin glue to seal the fistula may be considered. Xu et al. [17] reported favorable results after microsurgical cyst fenestration and imbrication.

Open surgery for sacral Tarlov cysts should be focused on fistulae and cyst walls. Surgical methods included two options: a direct microsurgical approach (cyst resection with or without muscle patch, cyst fenestration and imbrication, cyst resection with neck ligation and cyst resection combined with nerve root sheath reconstruction.

Open surgery focus on fistulae.

Asamoto et al. [18] recently reported a technique of eliminating the



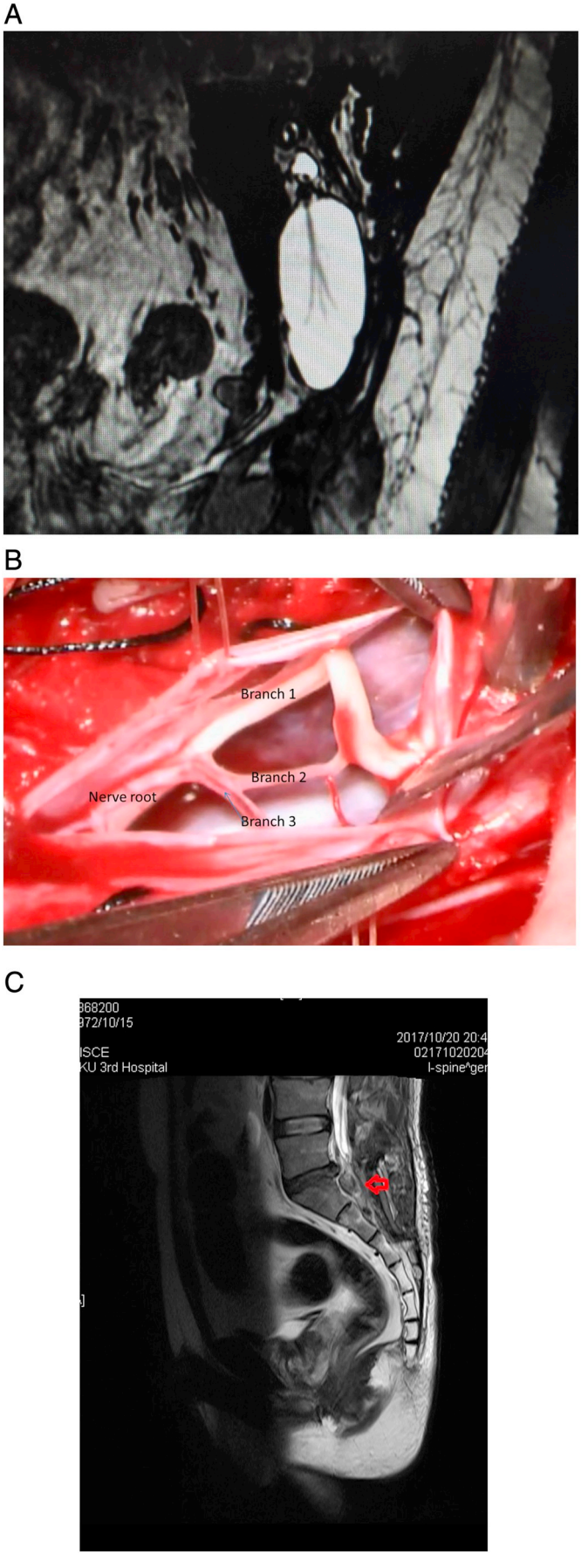


Fig. 3. A 26-year-old previously healthy female presented with a 2-month history of pain in the perineum and external genitalia and left leg. The pain was aggravated by long-time walking. She also suffered slightly bowel dysfunction. The preoperative IJOA score was 18. MRI showed eight cysts in the sacral canal (A). Eight cysts were confirmed intraoperation (B). Two weeks after surgical intervention, eight cysts were absent in the sacral canal on T2-weighted images (C). The patient's pain resolved completely.

(caption on next page)

Fig. 4. A 45-year-old previously healthy male presented with a 3-month history of swelling pain in the right thigh and sacrococcygeal region. The pain was relieved by bed rest, and was aggravated by walking and standing. He also suffered abnormal awareness of defecation. The preoperative IJOA score was 18. MRI showed one large cyst in the sacral canal. The spinal nerve root with three branches was shown inside the cyst on T2-weighted images (A). The spinal nerve root with three branches was confirmed intraoperation (B). Two weeks after surgical intervention, no cysts were observed in the sacral canal on T2-weighted images, the double reconstructed nerve sleeve around the spinal nerve root showed by red arrow (C). The patient's pain resolved completely. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

presumed ball valve mechanism of partially or intermittently communicating meningeal cysts by effectively opening the CSF fistula to facilitate two-way CSF flow and emptying of the cyst fluid. We also advocated this operating method for one-way valve fistulae of Tarlov's cyst. In our previous report, the unidirectional valve was changed and allowed bidirectional flow; the narrow obliterated fistula is enlarged to the size of the outer diameter of the reconstructed nerve root sheath [8].

Del Castillo-Calca'neo JD et al. [19] advocated anchoring and ligating the neck of sacral Tarlov cyst. A tubular retractor was placed on the right side at the L5-S1 junction, through which a small laminotomy was performed to reach the cyst; the cyst was then exposed and dissected to avoid radicular injury. The cyst cavity was opened and drained, and the neck was ligated to avoid relapse of the cyst. A single stitch was placed at the edge of the neck for anchoring, and then the Chitwood Knot Pusher was used to tie a knot around it.

Zheng XS et al. [2] advocated that stitching the fistula with a small piece of muscle. A nonabsorbable Monofil 6/0 suture is stitched through a small piece of muscle 5 mm in diameter, then through the edge of the fistula. After the suture is tied, the small piece of muscle is just blocking the fistula. Then some fibrin glue was injected to the suture to reinforce the sealing.

Smith ZA et al. [20] advocated that a surgical clip was used to close the connection between the thecal sac and the cyst. Closing this communication was critical to preventing both postoperative cerebrospinal fluid leakage and long-term recurrence.

However, in our opinion, the nerve root fibers inside the cyst should be protected in first. Nerves within the cyst-bearing dorsal nerve roots, structures notoriously sensitive to compression, may be distorted, compressed, and injured by the bulging cyst, and adjacent nerves are often also compressed. Meanwhile, the worsening of both radicular and local symptoms with time could be attributed to the gradual enlargement [3]. So, the nerve root fibers inside the Tarlov's cysts should also be protected intraoperation. Dredging the fistulae was better than blocking.

4.3. Open surgery focus on cyst walls

Removal of the partial cyst wall was another advanced procedure. This procedure required cyst fenestration, CSF drainage, and closure of the cyst wall with suture or clipping. Potts MB et al. [4] emphasized that plugging and ligating the cyst subarachnoid space communication, reinforcing the collapsed cyst wall with a dural substitute for surgical treatment sacral Tarlov cysts. In advanced report, he mobilized and rotated vascularized muscle as proximally based pedicle flaps deep to the lumbosacral fascia.

In our opinion, the redundant cyst wall should be removed and then to reconstruct the nerve root sheath. Reconstruction of nerve root sheaths was performed to restore the normal physiological environment of the spinal nerve root fibers. Oversewing of redundant cyst walls aimed to strengthen the congenital weakness, and restored the normal outer diameter of the involved nerve root sheath.

The excess meningeal tissue was imbricated to reconstruct the normal nerve root sheath and thecal sac. However, the nerve root sheath and adjacent dura were attenuated and transparent, exhibiting the thickness and consistency of atrophic arachnoid tissue, and in many cases the perineural cyst wall was too thin and friable to hold micro

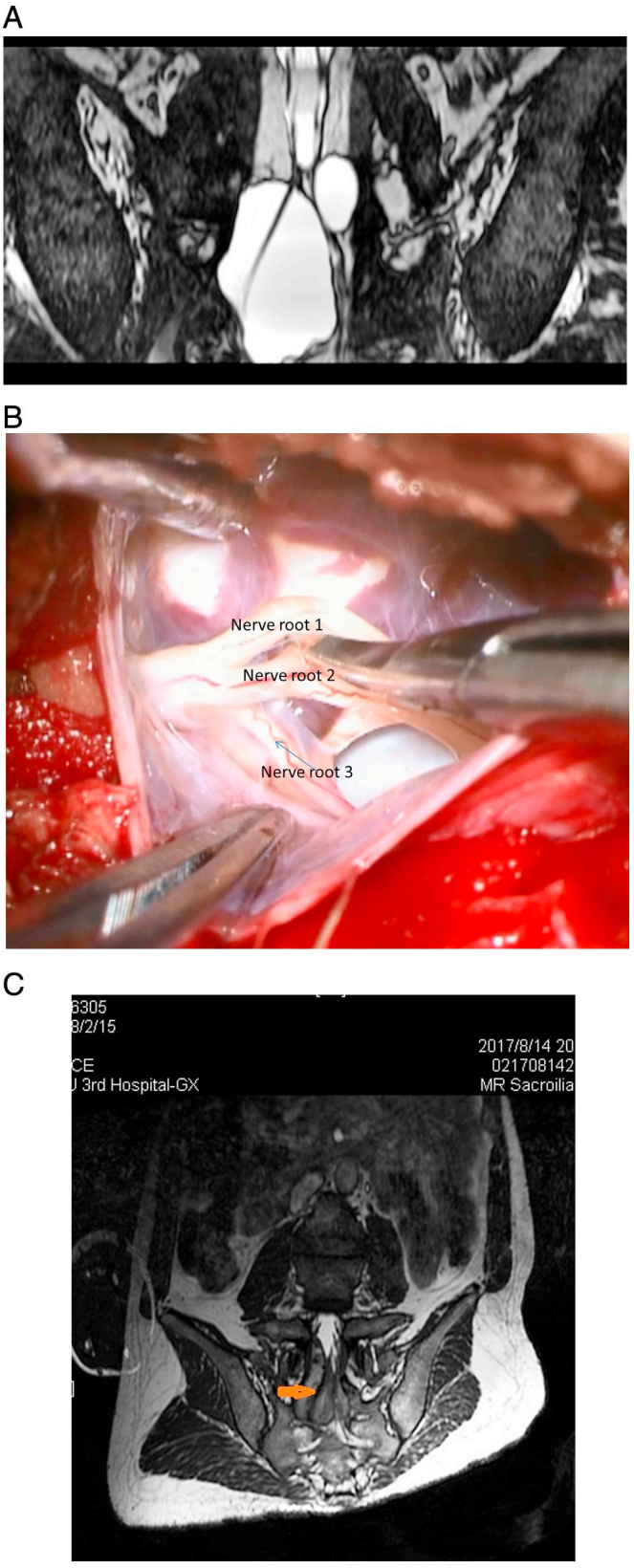


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Fig. 5. A 29-year-old previously healthy male presented with a 6-month history of radiating pain in the right leg. The pain was relieved by bed rest, and was aggravated by siting and standing. The preoperative IJOA score was 19. MRI showed one large cyst in the sacral canal. Three spinal nerve root fibers were shown inside the cyst on T2-weighted images (A). Three spinal nerve root fibers were confirmed intraoperation (B). Two weeks after surgical intervention, no cysts were observed in the sacral canal on T2-weighted images, the double reconstructed nerve sleeve around three spinal nerve roots showed by yellow arrow (C). The patient's pain resolved completely. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

sutures in order to allow watertight closure and repair [4].

It was another trouble that CSF fluids leaked out through reconstruction attenuated sheath in the sacral space. Potts MB et al. [4] designed that a few muscle flaps were rotated into the sacral cavity created by the Tarlov cyst. In some cases, a free subcutaneous fat graft harvested through a separate lateral buttock incision was added to provide more bulk around nerve roots and into dilated lateral foramina extensions of the cystic cavity into which the muscle flap did not reach or for which it was too bulky. After the fascial closure was completed, fibrin glue was added to seal the muscle-filled cavity if there was sufficient space [4].

However, when above mentioned muscle and fat graft together with fibrin glue became connective tissues and scar, the surrounding sacral nerve root fibers would be adhered, local compressed and occupied. That resulted in severe symptoms. It was emphasized that postoperative CSF leakage and cyst recurrence should be avoided by careful microscopic surgical techniques. However, in fact, the reconstructed cyst wall was very thin even after oversewing. So, the residue small cyst and recurrence of sacral cyst can't be avoided. In this report, in order to avoid the cyst residue and recurrent, a man-made dura was wrapped around the reconstructed nerve root sleeve. Attenuated and transparent nerve root sheath easy resulted in cyst residue and recurrent. So it was considered that using tough 3D-print man-made dural substitute to strengthen the attenuated and transparent nerve root sheath in this clinical study. Double sheath was reconstructed around the involved nerve root fibers. It can prevent the cyst from expanding and recurrence. Even if a little fluid leaked out through thin reconstructed sheath, the fluid would be absorbed by surrounding muscle and fat tissues. This hypothesis was confirmed by our clinical results. Three-month after open surgery, no cyst residue or recurrence was grasped in 24 patients on MRI scans. However, a De novo sacral cyst was found in left side six-month after right sacral Tarlov cyst had been operated with double sheath reconstructed method in a 29-year-old male patient. We can't explain the exact reason for this condition.

In summary, based on reconstruction the nerve sleeve, wrapping and forming another nerve sleeve with man-made dura was safety and effective operating method for treatment the sacral extradural meningeal cysts with nerve root fibers.

Appendix A. Supplementary Data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.inat.2018.10.020.

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