Diagnosis and Treatment of Symptomatic Multiple Sacral Perineural Cysts-Technical Note

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Abstract:

Introduction: Sacral perineural cysts are rarely symptomatic; however, they may occasionally cause various symptoms. As the patient exhibits multiple cysts, it often becomes difficult to determine if these cysts are symptomatic.

Technical Note: Six patients with multiple sacral cysts, identified using magnetic resonance imaging (MRI), were further examined using myelography and computed tomography (CT) immediately and 6-18 h after myelography. Symptomatic cysts were exclusively diagnosed as not enhanced immediately (filling defect sign) but displayed enhancement later (delayed filling sign/retention sign) compared to the subarachnoid space. A minimal laminectomy was performed on the target cyst. The dura and epineurium with the arachnoid of the cyst were then longitudinally incised along the nerve root, and the adhesion at the junction between the cyst and the dura mater was released. The incised epineurium and dura mater were sutured using 6-0 nylon and covered with multiple layers of polyglycolic acid seat and fibrin glue. A suction drain was placed for 1 or 2 days, and the patients were mobilized on postoperative day 1. Symptoms improved in all patients; however, the improvement ratio varied. At an average follow-up of 39 months, no recurrence was observed on the MRI.

Conclusions: This case series reports the diagnostic and surgical methods for multiple sacral perineural cysts and their outcomes. Delayed CT myelography is helpful in diagnosing symptomatic cysts. Moreover, all cysts with filling defect signs or delayed filling/retention signs demonstrated neural adhesions in the neck. Microsurgical fenestration and the release of adhesions are effective for the improvement of symptoms without recurrence.

Keywords:

Sacral perineural cyst, Tarlov cyst, Myelography, CT myelography, Clinical outcome, Fenestration, Minimally Invasive Surgery (MIS)

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Introduction

A perineural cyst is a cystic lesion originating at the posterior nerve root sheath of the dorsal root ganglion (DRG)[1,2]. It was first reported by Tarlov in 1938[3]. Although the perineural cyst occurs at various spinal levels, it is most common in the sacral region[4]. A sacral perineural cyst is relatively common, with a reported prevalence of 1.5%-13.2%[5,6].

Most cases with perineural cysts are asymptomatic[7] and diagnosed accidentally via magnetic resonance imaging (MRI). However, these cysts occasionally cause symptoms, such as low back pain (LBP), leg pain, numbness, perineal sensory disturbances, and bladder-bowel dysfunction (BBD)[7-13]. Along with the complexity of symptoms, many patients exhibit multiple cysts, making it difficult to determine which cyst causes symptoms. Previous reports have suggested that tumor size is related to symptoms[14,15]; however, achieving a consensus on its diagnosis remains elusive. Furthermore, various treatments are available, and the choice of treatment remains controversial. The present study introduces the diagnostic and surgical methods and clinical outcomes of symptomatic sacral perineural cysts.

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Table 1. Summary of Six Patients.

Case no.	Age	Sex	No. of cysts	Location	Operated cyst	Cyst size (mm)	CT interval (hours)	Filling defect sign	Delayed filling sign	Retention sign	Scalloping	Symptoms and neurological findings	Pre-op NRS	Final NRS	Follow-up duration (months)
1	55	F	6	S1-3	Rt. S1	30×20×20	7	-	+	+	+	Rt. leg pain (posterior calf-lateral foot), Rt. leg hypesthesia (S1 region)	9	0	63
2	32	F	2	S1	Lt. S1	15×15×10	18	-	+	+	+	LBP Lt. leg pain (buttock-posterior thigh)	LBP 3, Leg pain 8	LBP 1, Leg pain 0	60
3	49	F	3	S1-3	Rt. S2	13×10×8	3	+	+	+	+	Perineal sensory disturbance/Dysuria	7	3	36
4	50	F	3	S2-3	Rt. S2	14×16×18	6	+	+	+	+	LBP	LBP 5, Leg pain 5, Leg numbness 9	LBP 2, Leg pain 2, Leg numbness 0	24
					Lt. S3	16×15×25		+	+	+		Bil. leg pain (Rt. posterolateral thigh-calf, Lt. posterior thigh) and numbness (Bil. plantar) Rt. leg hypesthesia (S1 region)			
5	68	M	3	S2-3	Lt. S2	29×17×15	7	+	+	-	+	Perineal pain Lt. leg pain (buttock) and Bil. Leg numbness (Lt. plantar, Rt. toe) Lt. leg hypesthesia (S1-S3 region)	Perineal pain 5, Bilateral leg pain 5, Bilateral leg numbness 5	Perineal pain 0, Bilateral leg pain 0, Bilateral leg numbness 3	24
6	71	F	3	S1-3	Rt. S2	22×22×23	6	+	+	+	+	LBP, Perineal numbness/dysuria	LBP 5, Leg pain 5, Perineal numbness 8	LBP 2, Leg pain 2, Perineal numbness 4	24
					Lt. S3	10×10×12		+	+	+		Bil. leg pain and numbness (buttock-posterior thigh)			

Technical Note

Diagnosis

All the patients were diagnosed using MRI, myelography, and computed tomography (CT) myelography. The CT scans were obtained twice-immediately and 6-18 h after myelography. Cysts that were not enhanced initially (filling defect sign[16]) and/or demonstrating enhancement later (delayed filling sign) without washout (retention sign) in comparison to the subarachnoid space were considered to be cysts that can cause the symptoms. Then, the final diagnosis was made by careful comparison between the neurological findings or symptoms and the location or size of the cyst.

Surgical technique

Surgery was performed only for the cyst with filling defect sign, delayed filling sign, and/or retention sign. Under general anesthesia in the prone position, minimal laminectomy at the target cyst was performed. After the cyst was exposed, the epineurium and arachnoid were longitudinally incised. For extending the incision, a minimal incision was made in the dura mater and epineurium at the junction to confirm the exit of the nerve root. In all cases, severe adhesions were observed at the junction between the cyst and the dura mater. The adhesion was released, and fluent cerebrospinal fluid flow was confirmed. The incised epineurium and dura mater were sutured with 6-0 nylon and covered with multiple layers of polyglycolic acid (PGA) seat and fibrin glue. A suction drain was placed for 1 or 2 days, and the patients were mobilized on postoperative day 1. A sample video showing the surgical technique is provided as supplementary material (Supplemental example video1).

Clinical results

We retrospectively reviewed six consecutive patients (one male and five females; mean age of 54.2 years) who underwent surgery for multiple sacral perineural cysts at our institution (Table 1). The study protocol was approved by our Institutional Review Board (No. 3170). Various symptoms, including LBP, leg or perineal pain, numbness, and urinary discomfort, were observed. Their sizes ranged from 10 to 30 mm. Postoperatively, the visual analog scale for symptoms improved in all cases; however, the improvement ratio varied. Temporary worsening of BBD was observed in Case 6; however, it improved gradually. No cyst recurrence was observed in any patient during the latest follow-up; the mean follow-up period was 38.5 months.

Illustrative case

A 55-year-old female exhibited right leg pain, which was persisting for 6 months (Case 1). Physical examination revealed hypesthesia in the right S1 area and hyporeflexion of the right Achilles tendon; however, motor weakness and BBD were not observed. MRI revealed multiple cystic lesions at the lumbar and sacral levels (Fig. 1A-C). Myelogra-

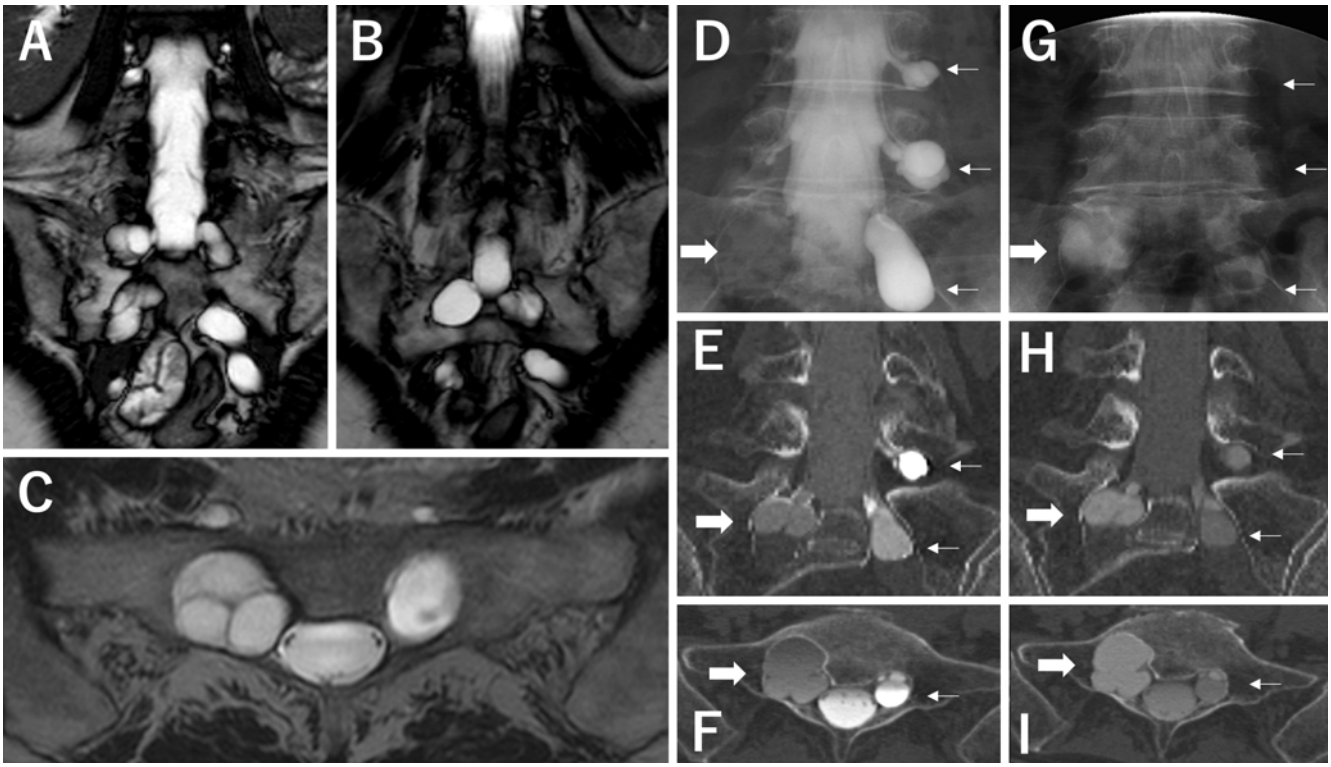

Figure 1. Preoperative T2 weighted coronal (A, B) and axial (C) magnetic resonance imaging displaying multiple cystic lesions at the lumbar and sacral levels. Myelography (D) and computed tomography (E, F) immediately after injection and 6 h later (G, H, I) demonstrated that a cyst in the left L4, L5, and S1 regions (small arrow) is enhanced immediately and washed out 6 h later, whereas a cyst in the right S1 region (large arrow) is not enhanced immediately (filling defect sign) but enhanced 6 h later (delayed filling sign/ retention sign).

phy and CT demonstrated that the cysts at left L4, L5, and S1 levels were enhanced immediately but were washed out 7 h later. However, the cyst at the right S1 cyst was not enhanced immediately but was enhanced 7 h later (Fig. 1D-I). Based on these findings, the patient was diagnosed with right S1 radiculopathy secondary to a right S1 cyst. An S1 laminectomy revealed a cyst at the S1 nerve root (Fig. 2A, B). After incising the epineurium, we noted scar tissue overlying the S1 nerve fibers at the proximal end of the cyst (Fig. 2C, D). Nerve fibers were not observed inside the cyst, indicating integration of nerve fibers into the cyst wall. After releasing the adhesion (Fig. 2E, F), the epineurium and dura mater were sutured with 6-0 nylon and covered with PGA seats and fibrin glue. The right leg pain was relieved immediately after surgery. Compared to preoperative MRI (Fig. 2 G), a follow-up MRI after 1 year revealed a reduction in size without fluid collection (Fig. 2H). No complications or recurrences were observed after 5 years (Fig. 2I).

Discussion

Tarlov et al. reported that perineural cysts occur along the nerve root, or at or distal to the junction of the posterior root and DRG[2,3]. They also reported that the cyst wall is composed of neural tissue [3,17] (Fig. 3). In our study, nerve fibers migrated into the cyst wall, and no nerve fibers were observed in the cyst. The mechanism of cyst formation is unclear; however, Tarlov proposed that hemorrhage due to mechanical injury results in venous drainage between the endoneurium and perineurium, leading to the formation of perineural cysts[2]. Paulsen et al. suggested that stenosis of the ostium of the nerve root sheath causes a ball-valve phenomenon that does not restrict inflow but restricts outflow[18]. In all six cases, scar tissue surrounding the nerve root was detected at the proximal pedicle of the cyst. Our findings also indicate that scar tissue formation, possibly due to factors such as injury, inflammation, and hemorrhage, creates a check-valve mechanism, which may lead to an increase in pressure inside the cyst (Fig. 4).

Sacral cysts are relatively common but rarely symptomatic (reportedly, 1%-4.6%)[6,18]. Therefore, diagnosing the cyst causing the symptoms is essential; however, achieving a consensus on the diagnosis, especially in cases with multiple cysts, remains challenging. Several reports have highlighted the use of myelography for diagnosis[16,19], which is effective in assessing the communication between the dural sac and the cyst. We considered immediate cyst filling with contrast medium postinjection as asymptomatic because the intracystic pressure was maintained equal to the intradural pressure. In contrast, the cysts exhibiting the "filling defect sign,"[16] "delayed filling sign," and/or "retention sign" are considered to possess a check-valve mechanism at the junction between the dural sac and cyst. This mechanism may cause symptoms due to the compression of other surrounding nerve

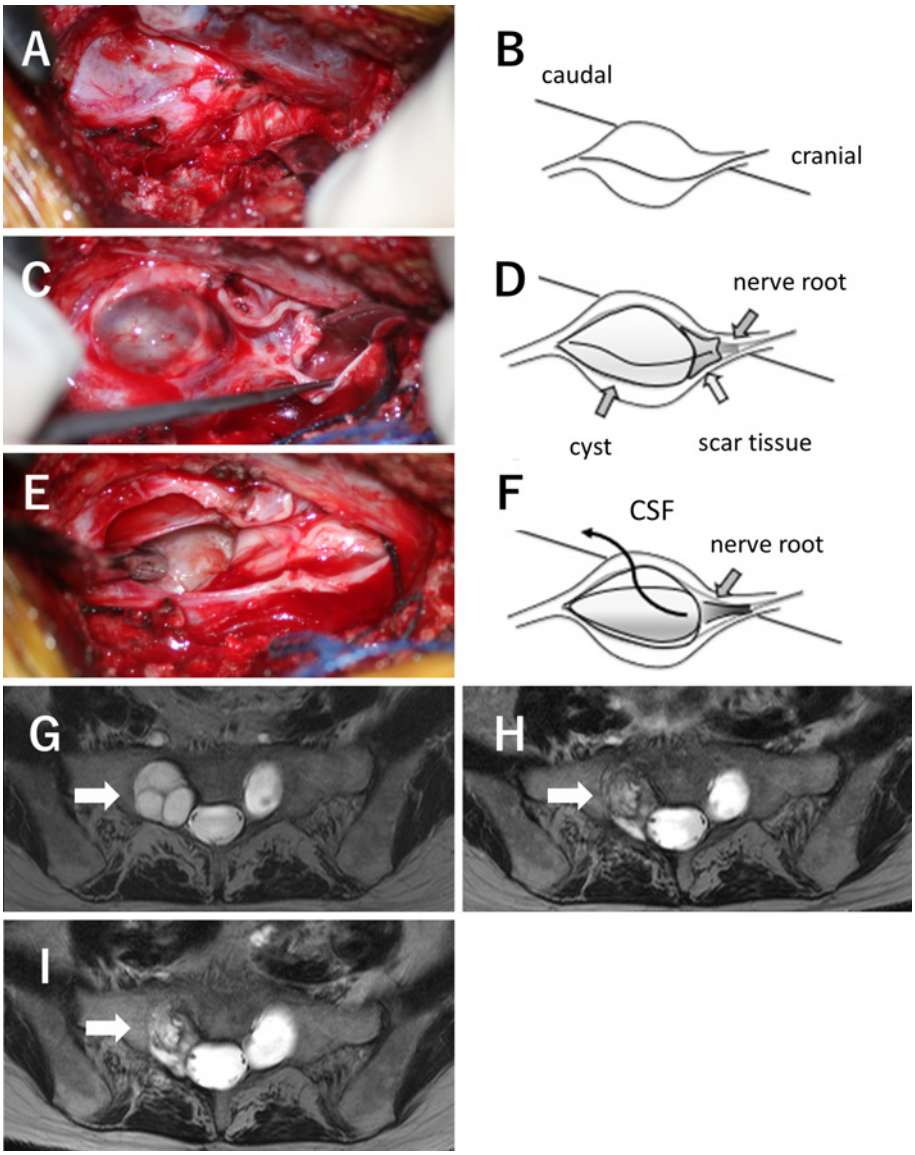

Figure 2. Intraoperative photographs (A, C, and E) and their schematic representations (B, D, and F). S1 laminectomy revealed a cystic lesion at the distal S1 nerve root (A, B). After an incision from the dura mater to the epineurium, scar tissue overlying the S1 nerve fibers is identified at the junction between the dural tube and cyst (C, D). Fluent cerebrospinal fluid flow is confirmed after incision of the cyst wall and release of scar tissue (E, F). Nerve fibers are confirmed under the scar tissue but spread over the cyst wall distal to the scar tissue. A multilocular cyst in the right S1 region on preoperative magnetic resonance imaging (G, arrow) displays shrinkage without fluid collection after 1 year (H, arrow) and 5 years (I, arrow).

roots or nerve fibers running along the cyst wall due to increased intracystic pressure. Based on this idea, we only surgically treated cysts with the "filling defect sign" or "delayed filling sign" in the CT myelography at early and delayed periods.

Several treatments are available for sacral cysts. Percutaneous aspiration can achieve temporal effects but cannot eradicate sacral perineural cysts[14,18,20]. Another minimally invasive option is injecting fibrin glue into cysts. Patel et al. first described this procedure and reported successful outcomes in all the cases[21]. After Patel's report, several studies reported favorable outcomes for fibrin glue therapy, with a success rate of 65%-86%[22-25]. In addition to complications such as aseptic meningitis and transient sciatica[26], severe complications such as adhesive arachnoiditis were reported[27]. Although the injection of fibrin glue is a minimally invasive technique, it should be applied with caution.

Open surgery is an alternative for sacral cysts, and several methods have been reported. The surgical options were classified into three groups: cyst resection[14], neck occlusion[28,29], and fenestration[19,30,31]. Both procedures yielded favorable outcomes[28]. However, histological[14] and intraoperative findings

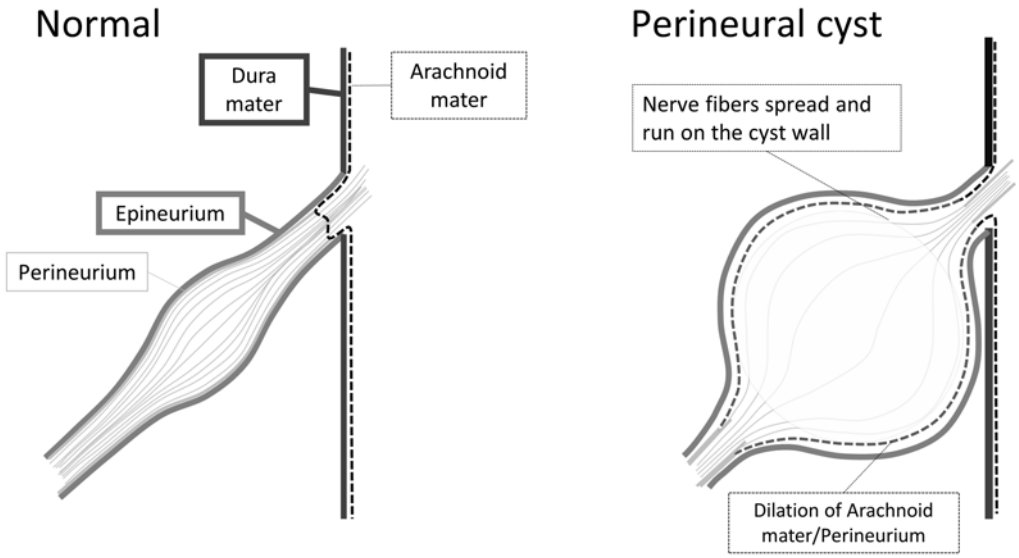

Figure 3. Schematic representation of the normal nerve root and sacral cyst. In the normal nerve root, the arachnoid mater is loopback proximal to the dorsal root ganglion (DRG) and continues to the perineurium. In contrast, sacral cysts develop between the endoneurium and perineurium, at or distal to the junction of the posterior root and DRG [2]. The nerve fibers spread within the cyst and run along the cyst wall.

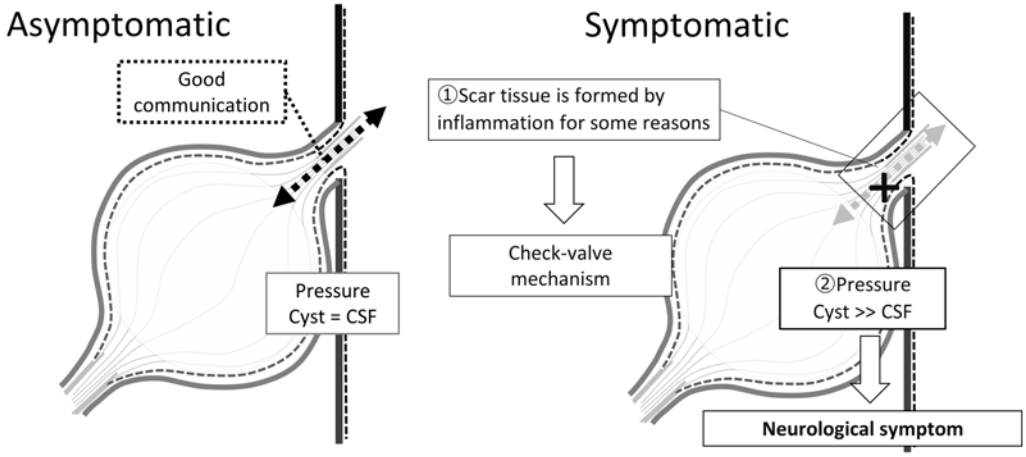

Figure 4. Hypothetical schematic representation of asymptomatic and symptomatic sacral cyst. Asymptomatic cysts exhibit cerebrospinal fluid communication between the dural canal and cyst, maintaining internal pressure equal to that in the subarachnoid space. However, in symptomatic cysts, the check-valve mechanism is established by adhesion between the dural canal and cyst. Increased internal pressure compresses nerve fibers running along the cyst wall or other nerve roots, causing symptoms.

have revealed that nerve fibers or neurons are present in the cyst wall, and cyst resection may cause neurological deterioration. For neck occlusion, Elsawaf et al. suggested using a local fat graft or gelatin sponge[28], whereas Cantore et al. reported a technique using a titanium clip[29]. Good outcomes were reported in both studies; however, recurrence was not investigated in imaging studies. The fenestration and release of adhesions can minimize neural damage[30]. Although this technique achieves free cerebrospinal fluid communication resulting from the destruction of the check-valve, our results demonstrated no cyst recurrence on MRI. The clinical results were positive, and the surgical method was considered simple, minimally invasive, and effective.

The present study has several limitations. First, the number of patients included in this study was low. However, the methods for the diagnosis and treatment of multiple sacral cysts have not yet been standardized. We believe that this case series treated with a surgical method based on a certain diagnostic criterion may be helpful for the further development of diagnosis and treatment methods. Second, although all the patients exhibited improvements in symptoms, complete improvement was not realized. Therefore, the cysts without "filling defect sign," "delayed filling sign," and/or "retention sign" could have been symptomatic. Further studies are required to validate the method proposed in the present study and establish standard diagnostic and treatment

procedures.

Conclusions

This case series presents diagnostic and surgical approaches for multiple sacral perineural cysts and their outcomes, emphasizing the utility of delayed CT myelography in diagnosing symptomatic cysts. All cysts with filling defects or retention signs exhibit neural adhesions at the neck. Microsurgical fenestration and the release of adhesions are effective for symptom improvement without recurrence.

Conflicts of Interest: The authors declare that there are no relevant conflicts of interest.

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Ethical Approval: The study protocol was approved by Institutional Review Board of Osaka Metropolitan University (No. 3170).

Informed Consent: The institutional review board waived the requirement for consent because of the retrospective design of the study.

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