Case Report

Excellent Pain Relief Using Spinal Cord Stimulation After Failed Bilateral Stereotactic Anterior Cingulotomy in a Patient with Intractable Neuropathic Pain of the Lower Extremities Following Surgery of Tarlov Cysts

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Background: *Neuropathic pain is a common consequence following disorder or injury of the nervous system. Neurosurgical treatment is a proper option in patients with intractable neuropathic pain.*
Objective: *To report outcome of stereotactic bilateral anterior cingulotomy (SBAC) and spinal cord stimulation (SCS) in a patient with intractable neuropathic pain.*
Material and Method: *A female patient suffered from intractable neuropathic pain on bilateral lower extremities after multiple operations for Tarlov cysts and lysis of surrounding adhesions. The patient underwent SBAC and SCS.*
Results: *After SBAC, pain intensity assessed by visual analogue scale (VAS) was decreased from 9-10 of 10 to 0-1 of 10. The patient could return to work and functional status was dramatically improved. Three months after SBAC, she developed recurrent neuropathic pain. Second SBAC resulted in transient pain reduction. One year later, the patient underwent SCS after an approval of reimbursement of expense for SCS equipment. After SCS, the VAS score was reduced from 8 of 10 to 0-1 of 10. Excellent outcome was maintained until the present.*
Conclusion: *Both of SBAC and SCS yield good outcome in the treatment of intractable neuropathic pain. Recurrent pain may occur after SBAC. Long-term pain relief can be maintained using SCS.*

Keywords: *Intractable neuropathic pain, Stereotactic bilateral anterior cingulotomy, Stereotactic surgery, Anterior cingulotomy, Spinal cord stimulation, Tarlov cyst, Perineural cyst*

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Chronic pain is a common problem found in neurological or neurosurgical practice. A distinctive entity of chronic pain is neuropathic pain which is caused by damage or disease involving the somatosensory system[1,2]. It is characterized by burning feeling, lancinating sensation, tingling, paresthesia and allodynia[1,3]. Several modalities are used for treatment of chronic neuropathic pain, such as medical treatment, transcutaneous magnetic stimulation, repetitive transcranial magnetic stimulation or cognitive-behavioral therapy[4-8]. Nevertheless, a number of patients who suffer from neuropathic pain cannot be relieved by aforementioned treatments and are classified as intractable neuropathic pain. Surgical intervention plays a major role in this particular group of patients.

Fundamentally, neurosurgical intervention for chronic intractable pain is stratified into ablative procedure and neuromodulation therapy[9]. Ablative surgery, such as dorsal root entry zone lesion or anterior cingulotomy, is an effective option in suppression of severe pain. Over recent decades, neuroablative procedure has been decreasingly utilized and superseded by neuromodulation, particularly spinal cord stimulation (SCS), peripheral nerve stimulation (PNS), and deep brain stimulation (DBS)[10].

The present article is a case report of

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utilization of an ablative procedure, stereotactic bilateral anterior cingulotomy (SBAC), combined with a neuromodulation, SCS, in a patient with refractory neuropathic pain caused by neural fibrosis following surgery of Tarlov cysts. Timeline of the treatment is presented and rationale for the use of surgical interventions is discussed.

Case Report

A 38-year-old female presented with radicular pain on both legs in 2010. Lumbosacral spinal magnetic resonance imaging (MRI) revealed Tarlov cysts (perineural cysts) of the sacral nerve root. The patient underwent S2-S4 laminectomy with unroofing of Tarlov cysts at a regional hospital. Postoperatively, her pain had not improved. Six re-operations attempting to decompress the spinal nerve roots and lysis of adhesions were performed at the same hospital. After the last operation in 2013, she still had severe radicular pain on bilateral lower extremities and gluteal regions. Burning pain with allodynia was the major pain characteristic. This neuropathic pain occurred continuously, was predominant on the right lower limb and aggravated by prolonged sitting or standing. Pain intensity assessed by visual analogue scale (VAS) was 2 of 10, 6 of 10, and 8 of 10 for minimal, average and maximal pain intensity score, respectively. It interfered with daily living and impaired function and ambulation of the patient. She could not walk and work as usual, and required an indwelling urinary catheter.

The patient was referred for pain management at Siriraj Pain Management Unit in 2014. After one year of appropriate medical treatment (pregabalin, oxcarbamazepine, methadone, celecoxib and venlafaxine), her pain was not improved. Therefore, surgical intervention was considered for treatment of intractable neuropathic pain in this case. Physical examination revealed intact motor and sensory function of both lower limbs. No pathologic reflex was found. Pain area is shown in Fig.1A. Allodynia was present on bilateral L4 to S1 dermatomes. Lumbosacral MRI showed marked fibrosis within the spinal canal at S1-S3 vertebral levels (Fig.1B-D) without recurrence of Tarlov cyst. Owing to MRI findings, attempting surgery on the region with fibrosis was not helpful for pain relief, so we looked at neurosurgical treatment of intractable pain. Because neuropathic pain involved both lower limbs and the patient still had intact neurological function, spinal cord stimulation (SCS) was a good option; however, the patient could not afford the cost of implanted electrodes and pulse generator. We, therefore, considered other therapeutic options which did not impair motor, sensory and gait function and had no hardware cost. Eventually, SBAC was chosen. After informed consent for risk and benefit of SBAC was given, the patient decided to undergo the surgery.

SBAC was performed in awake condition. After application of Leksell Stereotactic Frame System, images obtained from head computerized tomography were fused with those of cranial MRI for target localization. The targets for lesioning procedure were located at the anterior cingulated cortex 20 mm posterior to the anterior end of the lateral ventricle (Fig. 2A) and 5 mm lateral to the midline (Fig. 2B). A burr hole was drilled at 20 mm lateral to the midline. Radionic RF Lesion Generator System was used for lesioning procedure. A stereotactic thermocouple (TC) electrode with 1.8-mm diameter and 3-mm uninsulated tip for lesioning was inserted into the target (0 mm) through the burr hole, and then the target was coagulated with a temperature of 80°C for 80 seconds. After cooling, the TC electrode was withdrawn 2 and 4 mm (-2 mm and -4 mm), respectively. Coagulation with the same temperature and duration was done at the individual location of electrode withdrawal. The TC electrode was moved to the anterior and posterior tracks 5 mm paralleled to the first track. In the individual tract, coagulation was performed at 0, -2 and -4 mm, respectively. Nine lesions

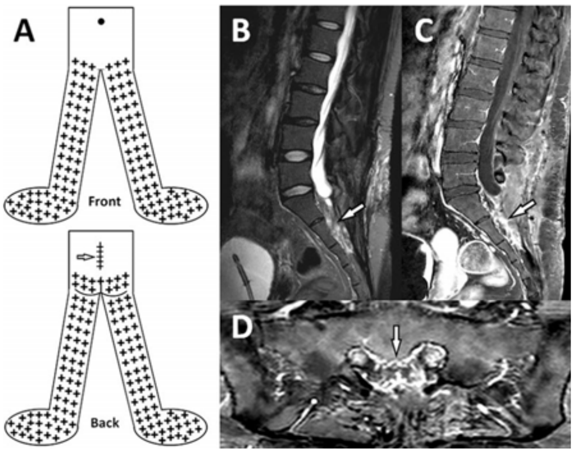

Fig. 1 Clinical and radiographic findings: (A) Area of neuropathic pain in front (anterior) and back (posterior) views, surgical scar (arrow); (B) lumbosacal spinal MRI in sagittal T2WI, (C) sagittal T1WI with contrast and (D) axial T1WI with contrast showing marked fibrosis (arrow) within the S1-S3 spinal canal.

were created on a unilateral side of the anterior cingulate cortex. The same lesioning procedure was performed on the contralateral anterior cingulum. Immediately following the bilateral procedures, neuropathic pain dramatically improved and the patient could moved her legs very well. The VAS score was reduced from 9-10 of 10 to 0-1 of 10. No cognitive adverse effect was found after the operation. Postoperative MRI showed proper size and location of bilateral lesions (Fig. 2C-D). The patient could walk independently and return to work. She could also return to ride a bicycle and a motorcycle.

Three months following SBAC, the patient developed recurrent neuropathic pain with the right side predominance. Pain intensity gradually increased with time and neuropathic pain disturbed daily living (the VAS score 8 of 10). SCS was reconsidered for long-term pain relief. We tried to reimburse the cost of SCS to the patient; however, it required a long period for consideration by an authorized committee. Because the patient developed recurrent intractable pain, and the reimbursement was being under consideration, the second SBAC was performed at 4 months after the first SBAC. We used the same surgical technique and targets. Postoperative outcome was favorable. The VAS score was decreased to 2 of 10. One month later, she developed recurrent neuropathic pain on the right lower limb with the VAS score of 5 to 7 of 10. The patient could ride a motorcycle, but could not ride a bicycle.

One year later, in 2016, the reimbursement of expense of SCS hardware was approved. So the patient underwent SCS which was divided to two sessions. The first operation was implantation of 8-contact electrodes for stimulation in a trial period. The procedure was done in awake condition and prone position. Percutaneous electrodes were placed into the epidural space bilaterally. The contacts of electrode were located at T9-T11 vertebral levels (Fig. 3A and 3B). Intra-operative stimulation showed good pain relief on both lower extremities. The electrodes were externalized for a trial period of SCS. After adjustment of stimulation parameters in the trial period, excellent pain relief was achieved and the patient satisfied with the results. The VAS score was reduced from 8 of 10 to 0-1 of 10.

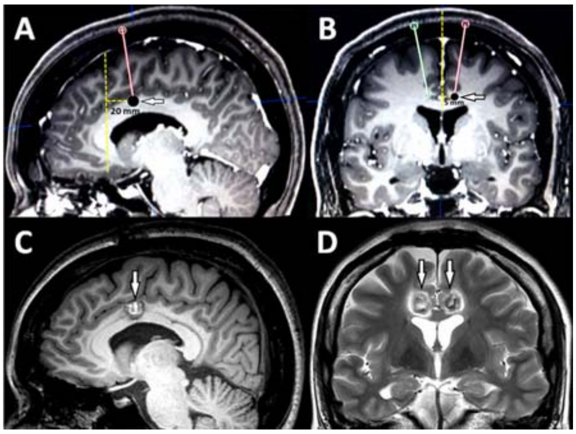

Fig. 2 Preoperative planning for stereotactic bilateral anterior cingulotomy and postoperative MRI: (A) preoperative sagittal T1WI showing a target for anterior cingulotomy (arrow) located at 20 mm posterior to the anterior end of lateral ventricle; (B) preoperative coronal T1WI showing the target (arrow) located at 5 mm lateral to the midline; (C) postoperative sagittal T1WI showing a large lesion at the anterior cingulum (arrow); (D) postoperative coronal T2WI showing large lesions at the bilateral anterior cingulate cortex (arrow).

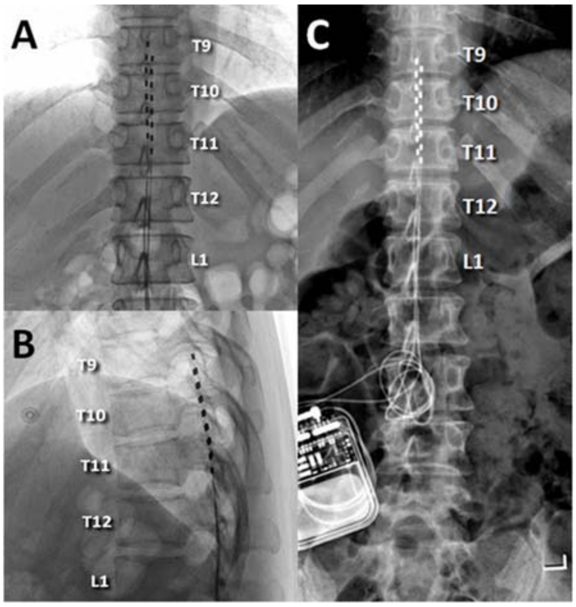

Fig. 3 Spinal radiograph following implantation of electrodes for spinal cord stimulation: (A) anteroposterior spinal radiograph in the trial period showing bilateral electrodes located at the T9-T11 vertebral levels; (B) lateral spinal radiograph in the trial period showing the electrodes located at the posterior portion of the spinal canal; (C) anteroposterior spinal radiograph following implantation of a pulse generator showing both electrodes connected to the pulse generator.

Therefore, the patient underwent the second procedure which was implantation of a pulse generator. Postoperative radiograph showed good position of the electrodes and pulse generator (Fig. 3C). Excellent neuropathic pain relief (the VAS score of 0-1 of 10) has been maintained until the present (1 year postoperatively).

Discussion

Chronic pain affects physical, psychological and social aspects, and daily living of patients[11-14]. Neurosurgical intervention has a major role in the treatment of refractory cases who have failed to conventional therapies[15]. Over recent decades neuromodulation has been increasingly used, whereas utilization of neuroablative procedure has been fallen. However, in our perspective, neuroablative procedure is still useful and effective in well-selected patients, and is a good alternative when patients cannot afford the high cost of hardware in neuromodulation therapy, particularly in developing countries.

Our reported case suffered from severe neuropathic pain on both lower extremities most likely caused by extensive fibrosis within the spinal canal of the sacrum. The patient underwent multiple re-operations which attempted lysis of the fibrosis. In our opinion, reoperation again is not helpful for relieving neuropathic pain and may promote more extensive fibrosis on and around the surgical site as well as increases risk of neurological morbidity, such as sacral nerve injury, permanent urinary or fecal incontinence. Thus neurosurgical procedure for intractable pain is the most appropriate measure in this patient. In our point of view, the best surgical option for this patient is SCS; however, the patient could not afford the cost of hardware used for SCS as well as other methods of neuromodulation therapy. We, therefore, attempted to choose the most optimal ablative procedure. Because the patient had no neurological deficit, the chosen ablative procedure should not impair motor, sensory, and proprioceptive functions. Although ablative neurosurgical procedures interrupting the nociceptive pathway do not impair motor function, they significantly affect somatosensory and proprioceptive recognitions. Lesioning procedure on the dorsal root entry zone or "DREZ lesion" has been used for treatment of intractable pain caused by various etiologies[16-22]. Nowadays, this procedure has proved to be effective in patients with brachial plexus avulsion pain[23-28]. It can also be used in the treatment of neuropathic pain caused by peripheral nerve origin[17,18,20,22], such as spinal nerve root pathology found in our patient. Nevertheless, pain relief following DREZ lesion must be exchanged by deterioration of somatosensory and proprioceptive functions on body parts supplied by lesioned spinal cord segments. Moreover, painful area of our patient is extensive; DREZ lesion for covering the entire painful area absolutely impairs major functions of the lower extremities. In the same way, disruption of the spinothalamic pathway within the spinal cord or "cordotomy" impairs somatosensory function of the body below the level of spinal cord ablation. Ipsilateral motor deficit, urinary incontinence, and the development of new post-cordotomy pain were also reported[29]. Additionally, cordotomy has a high rate of fading of analgesia with time which results in recurrent pain[30], so this lesioning procedure is reserved for only patients with intractable cancer pain with short life expectancy. Consequently, neither DREZ lesion nor cordotomy is appropriate for our patient.

Looking back to ablative procedures without interference of motor, somatosensory and proprioceptive functions, SBAC is one of them. SBAC has been long used for the treatment of intractable pain and several types of psychiatric disorder[31-37]. This procedure interrupts function of the limbic pathway which plays a major role in pain perception and emotional aspect[38]. Even though SBAC does not affect motor, somatosensory and proprioceptive functions, it carries a risk of postoperative cognitive decline and urinary incontinence. However, these operative morbidities are minimal and temporary, and can recover spontaneously with time[34,39]. Therefore, we considered SBAC as the most optimal ablative surgery for our case. Our result showed an excellent outcome of SBAC in the treatment of intractable neuropathic pain. Another major drawback of SBAC is postoperative recurrent pain[40,41] which we already informed the patient about it before the surgery, and eventually the patient had postoperative recurrent neuropathic pain. Fortunately, our patient never developed cognitive adverse effect following a couple of SBAC. Re-operation was not helpful for significant pain relief in previous report and also in our case[41].

As we mentioned that SCS is the best option in our patient, this procedure was performed after the second failure of SBAC and the approval of hardware reimbursement. Our result of SCS confirmed its efficacy in treatment of severe neuropathic pain caused by spinal nerve root pathology. To date and the best of our knowledge, this is the first case report of intractable neuropathic pain treated by a combination of SBAC and SCS in Thailand.

Conclusion

Neurosurgical intervention is alternative to medical treatment in patients with chronic intractable pain. SBAC renders good outcome in the treatment of refractory neuropathic pain; however, recurrent pain can occur with time. SCS is effective for long-term relief of neuropathic pain.

What is already known from this topic?

Neuropathic pain is a major consequence which occurs as a result of insult to the nervous system. In refractory cases, multimodal treatment, including surgical intervention, should be considered.

What this study add?

Neurosurgical treatment is a good alternative for chronic pain which has not responded to conventional therapies. Both SBAC and SCS are effective in the treatment of intractable neuropathic pain. Patients undergoing SBAC may develop recurrent pain with time. SCS yields favorable long-term pain relief in good surgical candidates with successful trial. This case is the first report of neuropathic pain treated by a combination of SBAC and SCS in Thailand.

Potential conflicts of interest

None.

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การลดอาการปวดได้อย่างยอดเยี่ยมโดยการผ่าตัดกระตุ้นไขสันหลังภายหลังการผ่าตัดเพื่อทำให้เกิดรอยโรคในสมองบริเวณ cingulate gyrus ส่วนหน้าไม่ได้ผลในผู้ป่วยที่มีอาการปวดที่เกิดจากระบบประสาทของขาสองข้างภายหลังการผ่าตัดรักษาถุงน้ำรากประสาทไขสันหลัง

นนทพล ปียวัฒนเมธา, บรรพต สิทธินามสุวรรณ, ปราโมทย์ เอื้อโสภณ

ภูมิหลัง: *อาการปวดที่เกิดจากระบบประสาทเป็นภาวะที่พบได้บ่อยหลังความผิดปกติหรือการบาดเจ็บของระบบประสาท การผ่าตัดทางประสาทศัลยศาสตร์เป็นทางเลือกหนึ่งที่เหมาะสมในผู้ป่วยที่มีอาการปวดที่เกิดจากระบบประสาทซึ่งไม่ตอบสนองต่อการรักษา*

วัตถุประสงค์: *เพื่อรายงานผลลัพธ์การรักษาอาการปวดโดยการผ่าตัดเพื่อทำให้เกิดรอยโรคในสมองบริเวณ cingulate gyrus ส่วนหน้าและการผ่าตัดกระตุ้นไขสันหลังในผู้ป่วยหนึ่งรายที่มีอาการปวดที่เกิดจากระบบประสาทซึ่งไม่ตอบสนองต่อการรักษา*

วัสดุและวิธีการ: *ผู้ป่วยหญิงหนึ่งรายมีอาการปวดที่เกิดจากระบบประสาทซึ่งไม่ตอบสนองต่อการรักษาที่ขาทั้งสองข้าง ซึ่งเกิดภายหลังการผ่าตัดหลายครั้งเพื่อรักษาถุงน้ำรากประสาทไขสันหลังและตัดพังผืดที่กดรัดรากประสาทไขสันหลัง ผู้ป่วยได้รับการผ่าตัดเพื่อทำให้เกิดรอยโรคในสมองบริเวณ cingulate gyrus ส่วนหน้าและการผ่าตัดกระตุ้นไขสันหลัง*

ผลการศึกษา: *ภายหลังการผ่าตัดเพื่อทำให้เกิดรอยโรคในสมองบริเวณ cingulate gyrus ส่วนหน้า ความรุนแรงของอาการปวดซึ่งประเมินโดย visual analogue scale ลดลงจาก 9-10 คะแนนจากเต็ม 10 คะแนนเหลือ 0-1 คะแนนจากเต็ม 10 คะแนน ผู้ป่วยสามารถกลับไปทำงานและสมรรถภาพของผู้ป่วยดีขึ้นอย่างชัดเจน สามเดือนหลังได้รับการผ่าตัดวิธีดังกล่าวผู้ป่วยเริ่มมีอาการปวดกลับเป็นซ้ำจึงได้รับการผ่าตัดด้วยวิธีเดิมเป็นครั้งที่ 2 ซึ่งช่วยให้อาการปวดลดลงแค่เพียงชั่วคราว หนึ่งปีต่อมาผู้ป่วยได้รับการผ่าตัดกระตุ้นไขสันหลังภายหลังจากได้รับอนุมัติค่าใช้จ่ายสำหรับอุปกรณ์กระตุ้นไขสันหลัง ภายหลังการผ่าตัดกระตุ้นไขสันหลังความรุนแรงของอาการปวดลดลงจาก 8 คะแนนจากเต็ม 10 คะแนนเหลือ 0-1 คะแนนจากเต็ม 10 คะแนน ซึ่งผลการรักษาที่ดีดังกล่าวสามารถคงอยู่จนถึงปัจจุบัน*

สรุป: *ทั้งการผ่าตัดเพื่อทำให้เกิดรอยโรคในสมองบริเวณ cingulate gyrus ส่วนหน้าและการผ่าตัดกระตุ้นไขสันหลังให้ผลลัพธ์ที่ดีในการรักษาอาการปวดที่เกิดจากระบบประสาทซึ่งไม่ตอบสนองต่อการรักษาอาจพบอาการปวดกลับเป็นซ้ำภายหลังการผ่าตัดเพื่อทำให้เกิดรอยโรคในสมองบริเวณ cingulate gyrus ส่วนหน้า การผ่าตัดกระตุ้นไขสันหลังช่วยลดอาการปวดในระยะยาวได้*