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A case of cyst formation at the intrathecal catheter tip of an Intrathecal Baclofen Pump

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Abstract : In 1984, intrathecal baclofen was first tried and intrathecal baclofen therapy (ITB) was covered by insurance in Japan in 2006. While ITB has contributed greatly to the treatment of spasticity, complications related to catheters, pumps, surgery, and programming errors have also been reported. In this report, we describe a case of cyst formation at the tip of an intrathecal catheter tip of an Intrathecal Baclofen Pump.

The patient was a 69–year–old man with spasticity in both lower limbs after decompression surgery for thoracic spinal injury and an ITB pump implantation was performed. During the operation, he had difficulty in lumbar puncture and required several punctures, but the operation was completed without any major problems. After the surgery, the daily dose of baclofen was gradually titrated and the spasticity improved, but on the 24th postoperative day, the spasticity worsened. As a result of direct intrathecal injection of baclofen via lumbar puncture, the spasticity improved markedly, and the catheter trouble was considered to be the cause of the problem. The reoperation was performed. Intraoperative catheter angiography revealed a cyst formation at the tip of the intrathecal catheter, so the intrathecal catheter was removed and the catheter was inserted after another lumbar puncture. After the operation, the spasticity improved markedly. If the spasticity does not improve in spite of increased medication, a cyst may have formed at the tip of the intrathecal catheter, and we should observe the patient carefully.

Key words : Intrathecal baclofen therapy; Postoperative complication; Spasticity; Intrathecal administration

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I. Introduction

Intrathecal baclofen (ITB) therapy has been an accepted treatment for spasticity and dystonia since Penn et al. attempted intrathecal injection of baclofen in 1984 [1]. It was introduced in Japan in 2006 and awareness of this treatment is increasing. Baclofen is a muscle relaxant and a GABA agonist that works at the level of the spine and brain to reduce muscle spasticity and dystonia. It is commonly administered either enterally or intrathecally and aims to improve range of motion, reduce risk of contracture development, decrease pain, and improve quality of life [2]. Patients who receive intrathecal baclofen, rather than enteral baclofen, are less likely to experience adverse effects such as confusion, listlessness, and nausea. In addition, a lower dose is required, and serious adverse effects are less common [3]. When adequate symptom control cannot be achieved by oral admin-

istration of baclofen, or an excessive side effect profile is associated with an effective dose, direct intrathecal administration is an effective method for controlling spasticity and reducing total baclofen dosage.

But we sometimes have significant complications associated with intrathecal baclofen. Motta F, Antonello CE reports at least 1 complication was present in 25% of the patients: 9.3% experienced an infection, 4.9% a CSF leak, 15.1% a problem with the catheter, and 1% a problem related to the pump [4]. Neil Haranhalli, et al reports 13 out of 76 (17.1%) patients primarily treated at their department had 25 complications. Additional 4 patients with pumps placed elsewhere had 6 complications and were subsequently treated by their group. The main complications were: catheter fracture, subcutaneous fluid collection, lumbar wound/CSF infection, lumbar catheter or connector protrusion, pump malfunction, distal catheter migration outside the thecal sac, and baclofen withdrawal [5]. Borowski et al. found that ITB therapy had 31% rate of complications requiring surgical management over a 3–year treatment period [6].

We have performed 13 surgeries for ITB pump implantation in our department since 2015. Here, we report a case of cyst formation at the intrathecal tip of an intrathecal baclofen pump.

II. Case report

A 69–year–old man with no significant medical history was paralyzed in both lower limbs as a result of a thoracic spinal cord injury. Decompression was performed by the previous orthopedic surgeon. His paralysis was not improved and he suffered from spasticity of both lower limbs. His recovery was complicated by oral baclofen therapy, so he was referred to our department for spasticity treatment.

Intrathecal baclofen screening was performed with test doses of 50 μg administered by a single shot intrathecal injection. After successful intrathecal screening, an ITB pump implantation was performed. A lumber catheter was placed with the distal tip at Th7/8. In the surgery, we performed lumber puncture several times because of difficulty, otherwise the procedure was completed with no problems.

An initial daily dose of 50 μg of baclofen (500 μg/mL) was used. The intrathecal baclofen dose was gradually titrated upward to achieve complete control with a dose of 236 μg/day. However, approximately 23 days after the initial implantation, spasticity control deteriorated significantly (**Fig.1**).

He was afebrile. There was no evidence of infection in the wound. His physical examination revealed increased spasticity without any change in other neurological parameters. Inflammatory markers (C reactive protein) were slightly elevated at 3.6 mg/L, and there were no other special findings in the laboratory data. A CT scan of the abdomen showed that intrathecal catheter had not displaced (**Fig.2**).

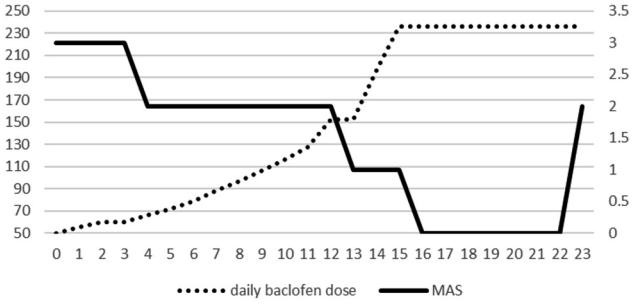

Fig.1 Daily baclofen dose and spasticity.
The daily dose of baclofen was gradually increased to 236 μg/day and the spasticity improved, but on the 23th postoperative day, the spasticity worsened and the Modified Ashworth Scale (MAS) was from 0 to 2.

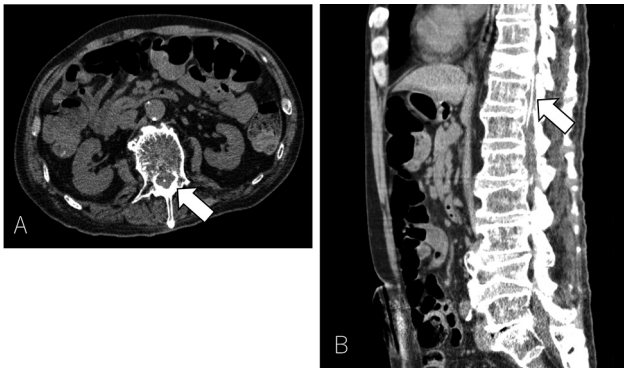

Fig.2 CT scan of abdomen (**A**: axial image, **B**: sagittal image).
CT scan of abdomen was performed when the spasticity worsened. White arrows indicate intrathecal catheter. Intrathecal catheter was located in the spinal canal and we confirmed that the catheter had not deviated. No other abnormal findings were observed.

Baclofen concentration was maintained at 500 μg/mL, and his refills were performed with standard Medtronic kits. Interrogation of the pump indicated that the correct dose of intrathecal baclofen had been delivered. The residual volume remaining within the pump corresponded to the computer estimation.

A catheter contrast examination was performed. Before injecting contrast agent through access port, we applied negative pressure to the syringe. No cerebrospinal fluid was drawn, and we terminated the examination. Intrathecal baclofen screening testing was performed again with test dose of 50 μg baclofen administered by a single–shot intrathecal injection. Spasticity improved significantly.

Therefore, we thought that the deterioration in spasm control was not due to drug tolerance but to a catheter

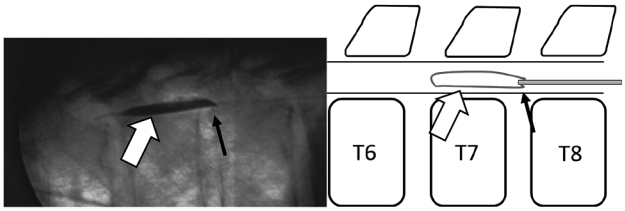

Fig.3 Lumber catheter imaging and illustration.
The left side is intraoperative fluoroscopy and the right side is an illustration of it. White arrows indicate contrasted cyst and arrows indicate a catheter tip. Contrast agent was injected through the catheter with omnipaque. The contrast medium flowed out of the distal tip of the catheter, but did not spread into intrathecal space. A cystic sac was formed at the tip of the catheter and moving flutteringly.

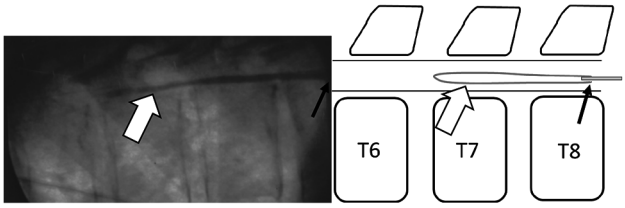

Fig.4 Lumber catheter imaging and illustration after the catheter pulled out.
The left side is intraoperative fluoroscopy and the right side is an illustration of it. White arrows indicate contrasted cyst and arrows indicate a catheter tip. We pulled out a lumber catheter as contrast agent was injected. A lumber catheter was withdrawn smoothly, and the tract was bag–shaped enhanced as we pulled it out.

trouble, and we planned to reimplant the catheter.

Postoperative day 71, we performed reoperation. The pump was removed from under the skin. The pump and peritoneal catheter were clear. The lumber catheter took off the pump and injected contrast agent from the distal tip of the catheter by omnipaque. The contrast medium was spilled from the tip of the catheter, but a cystic sac was formed at the tip of the catheter and contrast medium was not spread (**Fig.3**). The cyst was moving flutteringly, so we assessed the cyst was located in the intrathecal space. We pulled out a lumber catheter. A lumber catheter was withdrawn smoothly, and the tract was bag–shaped enhanced as we pulled it out (**Fig.4**). We thought that a cystic sac covered the lumber catheter. We removed the lumber catheter and performed lumber puncture again. New lumber catheter was inserted. Catheter angiography showed diffusion of contrast medium from the catheter tip into the intrathecal space, and outflow of spinal fluid from the catheter on the pump side was observed.

An initial daily dose of 50 μg of baclofen (500 μg/mL) was used, and we increased daily baclofen dose gradually. 6 days after the reimplantation, daily baclofen dose was 70 μg. Modified Ashworth Scale improved from 3 to 0. He transferred to rehabilitation hospital. His spasticity is currently well controlled. No further problems have developed since the catheter correction.

III. Discussion

Intrathecal baclofen therapy is an accepted treatment for spasticity and dystonia. It decreased muscle tension, improved positioning, and decreased decubitus ulcers. But we sometimes have significant complications that include drug withdrawal, catheter infection, drug overdose, failure, and pump failure. ITB therapy is associated with complications, many of which require additional surgery. We experienced a case of spasm deterioration after surgery. Inflammatory mass in patients receiving baclofen as a sole intrathecal agent is reported. But cyst formation at the intrathecal tip has never been reported.

In 1991, North et al. reported the first case of an IT catheter tip inflammatory mass in a patient receiving morphine. Since then, there have multiple reports of catheter–associated masses. But catheter–associated masses were not described in any patient who had received baclofen as their only IT drug [7]. Deer et al. reported two cases of inflammatory mass in patients receiving baclofen as a sole intrathecal agent [8]. Murphy et al. also reported catheter tip granuloma. Potential mechanisms for catheter tip granuloma formation have included catheter–based mechanisms, such as catheter tip design, placement–associated trauma, final tip position, infection, and silicone hypersensitivity, and drug–related mechanisms, such as impure/contaminated drugs and the action of opioid agonists on immunological function and the blood–central nervous system barrier [9]. North et al. raised the possibility of arachnoiditis as an important etiological factor; however, the reports of granuloma formation in the absence of previous spinal surgery argue against this [7]. Jones et al. reported that an outbreak of granuloma formation was consequent upon drug contamination; however, these results were limited [10]. Preventive measures suggested maintaining the IT opioid dose and concentration at a small level and lumbar versus thoracic placement of the catheter, and any alteration in drug efficacy or neurological status must alert the attending physician to the possibility of a catheter–associated mass, and appropriate investigations must be undertaken [9]. Guidelines for the prevention, diagnosis, and management of catheter tip–associated masses has been established, but the authors point out that baclofen–associated masses have not been reported and that, therefore, the guidelines are predominantly concerned with patients receiving IT opioids [11].

In this case, approximately 3 weeks after the initial implantation, spasm control significantly deteriorated but improved with low baclofen dosage after catheter reinsertion. Cyst formation at the intrathecal catheter tip of an intrathecal baclofen pump is considered to prevent drug delivery properly, so muscle tension increased. We thought that baclofen delivered properly by reoperation and muscle tension decreased. MRI was not taken due to no neurological symptoms, so mass formation was unknown.

One possible cause of cyst formation was post thoracic spine surgery and it may have been prone to cause adhesions. After initial surgery, spasticity did not improve easily, so the baclofen concentration was titrated up. As a result, we administered high dose of baclofen. These adhesion–prone environment and high baclofen dose may have been involved in the formation of the cyst. The mechanisms and preventive measures for mass or cyst formation are not clear, but it is important to suspect mass or cyst formation when spasm control deteriorates.

IV. Conclusion

We experienced a case of cyst formation at the intrathecal tip of an intrathecal baclofen pump. If the clinical response to the infused drug changes, physicians need to increase vigilance and consider catheter removal.

The author has completed a self–report on conflict of interest to the Japanese Neurosurgical Association for the past 3 years. The authors have no conflict of interest to disclose.

References

1) Penn RD et al : Intrathecal baclofen alleviates spinal cord spasticity. Lancet 323(8385): 1078, 1984.
2) Borowski A et al : Complications of intrathecal baclofen pump therapy in pediatric patients. J Pediatr Orthop 30: 76–81, 2010.
3) Pin TW et al : Use of intrathecal baclofen therapy in ambulant children and adolescents with spasticity and dystonia of cerebral origin: a systematic review. Dev Med Child Neurol 53: 885–895, 2011.
4) Motta F et al : Analysis of complications in 430 consecutive pediatric patients treated with intrathecal baclofen therapy: 14–year experience. J Neurosurg Pediatr 13(3): 301–306, 2014.
5) Neil Haranhalli et al : Intrathecal bacrofen therapy: complication avoidance and management. Child's Nervous System 27: 421–427, 2011.
6) Borowski A et al : Complications of intrathecal baclofen pump therapy in pediatric patients. J Pediatr Orthop 30: 76–81, 2010.
7) North RB et al : Spinal cord compression complicating subarachnoid infusion of morphine: case report and laboratory experience. Neurosurgery 29: 778–784, 1991.
8) Deer TR et al : Inflammatory mass of an intrathecal catheter in patients receiving Baclofen as a sole agent: A report of two cases and a review of the identification and treatment of the complication. Pain Med 8: 259–256, 2007.
9) Murphy PM et al : Intrathecal catheter Granuloma associated with isolated Baclofen infusion. Anesth Analg 102: 848–252, 2006.
10) Jones TF et al : Neurological complications including paralysis after a medication error involving implanted intrathecal catheters. Am J Med 112: 31–36, 2002.
11) Hassenbusch S et al : Management of intrathecal catheter–tip inflammatory masses: a consensus statement. Pain Med 3: 313–323, 2002.

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バクロフェン髄腔内注入ポンプの髄腔内カテーテル先端に嚢胞形成した1例

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抄　録：1984年にバクロフェンの髄注が試みられ，その効果を経てバクロフェン髄注療法（ITB療法）は日本でも2006年に保険収載されている。痙縮治療に多大な貢献をしている一方でカテーテル関連，ポンプ関連，手術関連，プログラムエラーなどの合併症も報告されている。その中で，今回髄腔内カテーテル先端に嚢胞形成をきたした症例を経験したので報告する。

症例は69歳男性で胸髄損傷に対して除圧術後で両下肢の痙縮が著明であり，ITBポンプ埋め込み術を施行した。術中は腰椎穿刺が困難で数回穿刺を要した他は大きな問題なく終了した。術後は徐々にバクロフェンの1日投与量をあげ，痙縮は改善していったが，術後24日目に痙縮の悪化を認めた。腰椎穿刺を行いバクロフェンを直接髄注した結果，痙縮は著明に改善したため，カテーテルのトラブルと考え，再手術を施行した。術中にカテーテル造影を行うと嚢胞が髄腔内カテーテル先端に形成されていたため，髄腔内カテーテルを抜去し，再度腰椎穿刺を行ってカテーテルを挿入したところ痙縮は著明に改善した。薬液増加にもかかわらず痙縮の改善に乏しい場合，髄腔内カテーテルの先端で嚢胞を形成している可能性があり，注意深く観察する必要がある。

索引用語：バクロフェン髄注療法；術後合併症；痙縮；髄腔内投与