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Case Report

Tension Pseudomeningocele Associated with Retained Intrathecal Catheter: A Case Report with a Review of Literature

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Catheter related complications are not uncommon with permanently implanted intrathecal drug delivery systems. Pericatheter leak of cerebrospinal fluid usually responds to conservative treatment.

We report a case of tension pseudomeningocele due to retained lumbar intrathecal catheter. It is essential to be aware of this rare complication and we recommend appropriate neurosurgical involvement in the management of pseudomeningocele to avoid potential and catastrophic complications.

This case presents one of the rare complications associated with the implantation of IDDS. If a patient with IDDS develops a complete fracture of the catheter at the spinal end, all attempts should be made to define the 2 ends of the catheter and establish the continuity with a titanium connector. If the spinal end of the catheter is retracted deep into the interspinous ligaments and not recoverable, avoid entering the intrethecal space at the same level. If a patient develops pseudomeningocele in the postoperative period of IDDS and conservative methods including autologous epidural blood patch fail, we recommend an MRI of the spine for a detailed study along with prompt neurosurgical consultation.

Key words: Intrathecal catherer, retained, broken intrathecal catheter, pericatheter leak, lumbar swelling, psedomeningocele, duroplasty, ventriculoperitoneal shunt

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Intrathecal drug delivery systems (IDDS) have been the standard methods of treatment of chronic pain of nonmalignant and malignant etiologies for more than 2½ decades (1-5). While the Food and Drug Administration has approved morphine and baclofen for intrathecal therapy of chronic pain and spasticity, other opioids such as fentanyl, hydromorphone, sufentanil, and meperidine, and nonopioids including bupivacaine, clonidine, and zeconitide are used to treat patients with different pain syndromes not well controlled with oral or transdermal opioid and nonopioid therapies (6-9).

Catheter related complications include pericatheter cerebrospinal fluid (CSF) leak, catheter breakage, disruption, occlusion, slippage, hub fracture, or dislocation occur in 3 – 6% of IDDS implant cases (10-12).

Pseudomeiningocele is the extradural collection of CSF due to a breach in the dural-arachnoid layer and they occur after incidental durotomy during lumbar spine surgery. Pediatric pseudomeningoceles

have been reported following the implant of IDDS for baclofen infusion (10). We report a detailed case of tension pseudomeningocele in an adult patient due to a broken and retained lumbar intrathecal catheter. This case focuses on the need for recognition of a rare and semi-emergency clinical condition that necessitates prompt neurosurgical involvement to avoid catastrophic complications from meningitis due to an external CSF leak.

Case Report

Seven years ago, a 45-year-old thin-built male who was 6'2" in height and weighed 149 pounds, presented to our pain center for management of chronic pain in the upper and lower extremities from Complex Regional Pain Syndrome (CRPS) Type I. The patient underwent the implant of an intrathecal catheter and SynchroMed EL infusion pump in Michigan and was on simple continuous intrathecal infusion of morphine sulfate (MSO4).

The patient was diagnosed with CRPS 14 years earlier secondary to blunt trauma. He received a series of local anesthetic sympathetic blocks and later on spinal cord stimulation with epidural electrodes and a radiofrequency receiver unit of the Medtronics system for electroanalgesia. A year later the system was explanted for inadequate pain control. Six years later, an Intrathecal catheter and SynchroMed EL pump were implanted.

Over the subsequent 5 years of follow up at our pain center, the patient was managed with monthly titration of intrethecal MSO4 infusion from 3 mg/day to 16 mg/day along with bupivacaine 4.7 mg/day. Five years after the initial pump implant, the SynchroMed EL was replaced with a SynchroMed – II for the end of pump battery life. Intraoperatively, patency of the intrathecal catheter was confirmed by free aspiration of CSF. Three months later, the patient continued to report inadequate pain relief. A side port of the pump was accessed with failure to aspirate CSF. A fluoroscopic side port study with injection of Omnipaue 300 also failed to visualize the intrathecal spread of contrast. However, contrast extravasation was noted away from the pump head close to the lumbar para spinal area. With diagnosis of partial disconnect or rupture of catheter, the patient was scheduled for surgery.

After exploration of the pump reservoir, a repeat side port access failed to aspirate CSF and there was no kink or disconnection of the catheter. When the lumbar area was explored, the spinal end of the catheter was found completely broken distal to the anchor in the para spinal muscle tissues and the catheter could not be retrieved from the depth. Although the surgery was electively scheduled, Medtronics representatives could not be available for the catheter replacement. Hence the decision was made to replace the pump back in the original subcutaneous abdominal wall pocket and to reschedule at a later date for intrathecal catheter implant to maintain continuity with the in-situ pump reservoir. On the twenty-sixth post-op day, the patient presented with wound discharge, swelling, and a partially exposed pump reservoir that required an emergency removal.

Four months later the patient underwent the implantation of an intrathecal catheter and SynchroMed – II programmable pump. The patient had an uneventful immediate postoperative period with suture removal on the tenth day. In the seventh week he developed a small cystic fluctuant swelling in the lumbar area that increased over next 3 days to a tense tender swelling measuring 4 x 4 x 3 inches (Fig. 1). With an initial diagnosis of pericatheter CSF leak, an autologous epidural blood patch was performed under fluoroscopy via the L3/4 interlaminar space and the patient was recommended the use of an abdominal binder and strict bed rest. In spite of the above measures, the swelling increased in size and upon aspiration under sterile conditions, 30 mL of clear fluid was drained that was negative for cytology with a glucose level of 48 mg/dl. As the fluid rapidly re-accumulated within just a few hours to its original size, a fluoroscopic contrast study was done via the pump's side port. The study showed intrathecal spread of contrast without any leak along the extra spinal course of the catheter.

With a clinical diagnosis of a pericatheter CSF leak, the lumbar area was explored and the CSF leak around the catheter was confirmed. Purse-string silk sutures were carefully placed around the catheter gathering the surrounding paraspinal tissues and the incision was closed in 2 layers with the application of a pressure dressing. The patient was discharged home with strict recommendations to use an abdominal binder and bed rest.

The CSF continued to leak into subcutaneous tissues with the recurrence of a tense tender tennis-ball sized cystic swelling without external leakage of CSF (Fig. 2). At this point an urgent neurosurgical consultation was obtained and a diagnosis of CSF fistula

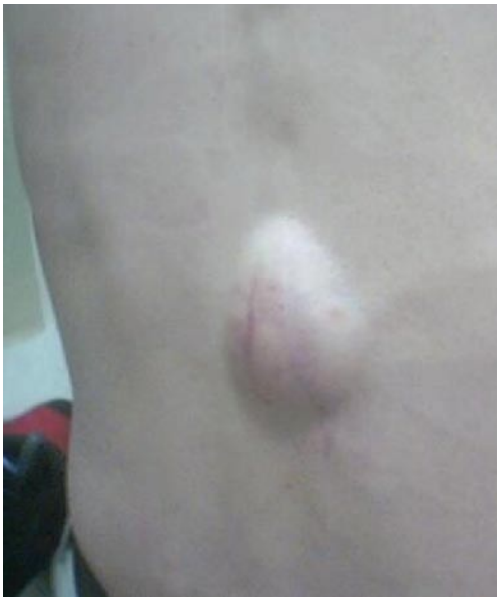

Fig.1. *Tense cystic swelling measurig 4 x 3 x 3 inches developed 7 weeks after IT catheter and SynchroMed II Pump implant. On close observation the subcutaneous course of the catheter in the right paraspinal area is appreciated.*

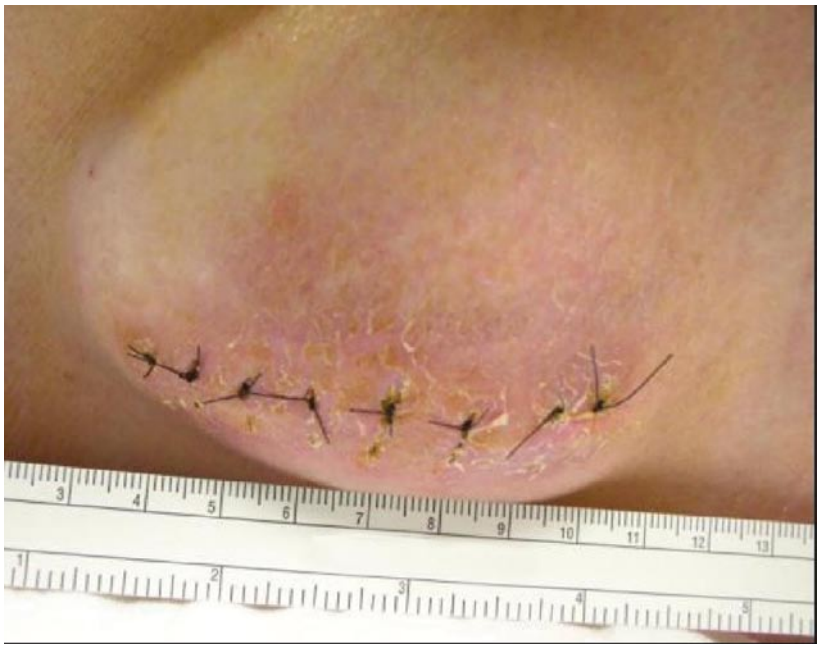

Fig. 2. *Recurrent tense swelling with impending rupture. Tension pseudomeningocele developed in the lumbar area after a repeat IT catheter and SynchroMed II implant.*

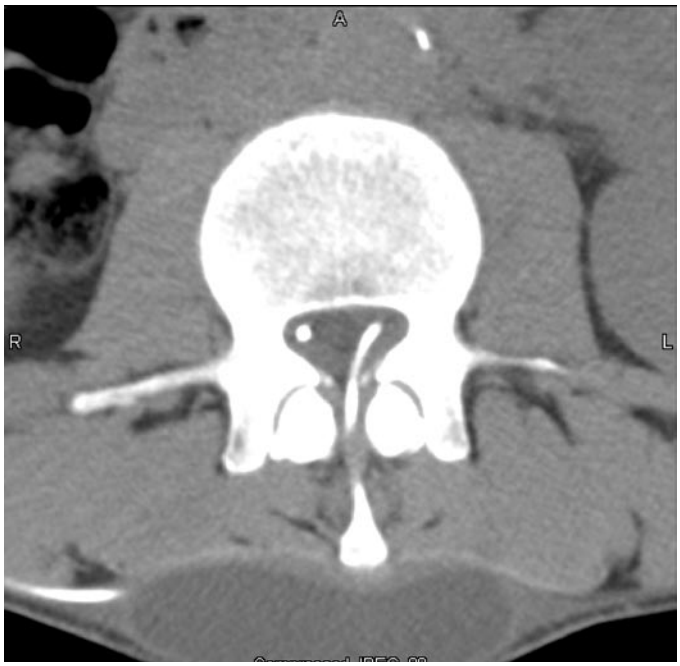

Fig. 3. *CT scan of the lumbar spine demonstrating subcutaneous swelling and 2 intrathecal catheters with one of them passing through the interlaminar space.*

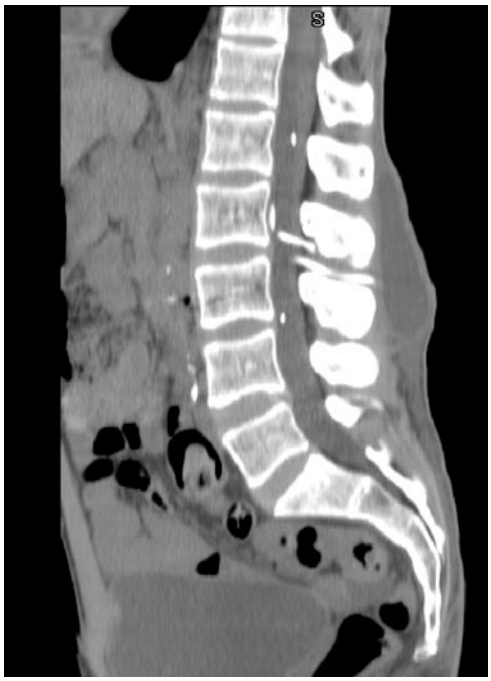

Fig. 4. *CT scan of lumbosacral spine sagittal view showing entry of 2 catheters at L2/3 level. Lumbar subcutaneous pseudomeningocele extending from L1 to L3 level.*

with tension pseudomeningocele was made. There was a great concern with potential imminent rupture of swelling leading to CSF contamination and meningitis. Emergency computerized axial tomography of the thoracic and lumbar spine showed the presence of 2 intrathecal catheters entering the thecal sac at the L2-3 inter laminar space (Figs. 3 and 4). After discussion with the neurosurgeon, the decision was made to remove the infusion pump along with the intrathecal catheters at the L2-3 level. In surgery, an L2 laminectomy was performed and pericatheter CSF leak was noted. The 2 catheters were removed from the intrathecal space followed by duroplasty. Although duroplasty appeared water tight in the recumbent position, it was anticipated that Valsalva and other maneuvers might stress the duroplasty resulting in a recurrence of the fistula. Therefore, an external lumbar drain at the L4-5 level was placed.

The follow-up examination of the patient was alarming showing a CSF leak into the subcutaneous tissues threatening to repeat the process that led to surgery. This was believed to be primarily due to the patient's poor compliance with his bed rest orders. Restlessness and a heavy smoking habit resulted in frequent ambulation of the patient with a kink of the external drainage system and its malfunction. Two days post-operatively an internalized lumbo-peritoneal shunt (NMT Corporation, Duluth, GA) was placed. This device had the immediate effect of increasing compliance with bed rest orders as the patient did experience low pressure headaches when fully upright. As the days went on, the low pressure headache subsided and the patient became increasingly ambulatory with no recurrence of CSF collection and complete healing of the skin incision. We decided to leave the lumbo-peritoneal catheter in long-term as it is not inconceivable that its removal could result in a new CSF collection at the lower lumbar level. Currently the patient's pain symptoms are managed with oral opioid medications.

Discussion

Over the past 25 years, neuroaxial analgesia using implantable IDDS with continuous infusion of opioids ± adjuvant medications has become one of the standard methods of treatment of chronic pain for non-malignant and malignant etiologies (1-5) and is associated with intrathecal catheter related complications ranging from 3 to 6% (10-13).

Use of a 15-gauge Tuohy needle for dural puncture followed by the intrathecal placement of 1.4 mm outer diameter Medtronics intrathecal catheter (InDura) leaves potential for pericatheter leak of CSF after removal of the Tuohy needle. Snuggly fitting a purse-string suture around the catheter with non-absorbable silk decreases the incidence of postoperative pericatheter CSF leak, reported in 2.0% of cases (13,14) after IDDS implant. In most cases, the resulting postural headaches are managed with conservative measures like bed rest and increased fluid intake along with caffeine. If symptoms still persist, an autologus lumabr epidural blood patch under fluoroscopy is recommended (10,15,16). InDura catheters are soft, elastic, and flexible, and are made up of silicon with a radio-opaque tip. For better delineation of catheter continuity, a contrast study is recommended. As the catheters are soft, they are prone to fracture and breakage in up to 5.1% of cases (13). If the breakage is close to the hub as evidenced by the contrast study, the catheter is trimmed beyond the breakage and reconnected to the hub. If the partial or complete catheter breakage is noted at the spinal area, after resection of the damaged segment, the 2 ends of the catheter are connected using a titanium connector provided in the Medtronic catheter repair kit.

In this patient, the catheter completely fractured distal to the anchor and was retracted into deeper interspinous tissues and remained asymptomatic until the second catheter, that was accidentally juxtapositioned to it in L2/3 space, triggering a possible ball-valve effect creating a persistent pericatheter leak, thereby leading to the development of a pseudomeningocele under tension.

Postoperative CSF collection around the intrathecal catheter is addressed, in pain management literature, as a "CSF leak." If the subcutaneous swelling is large enough, as in this case, the swelling is called "CSF hygroma" (13,17). We believe it is a misnomer as typically "hygroma" refers to a benign, distended, lymph filled cavity, usually seen in children (17). Review of the literature refers such swellings to be appropriately called as pseudomeningoceles because they result from extradural collection of CSF due to the breach in the dural-arachnoid layer and they are, at least in the initial stages, not lined by arachnoid membrane.

In a single institutional retrospective analysis of 314 intrathecal catheter and pump-related procedures, performed in 195 pediatric and adult patients requiring intrathecal baclofen therapy, Vander et al (10) encountered 48 catheter revisions (38 pediatric and 10 adult patients) for catheter-related complications. These complications included hub fracture or dislocation, breakage or disruption, occlusion, and slippage or pullout. The pediatric population had 37% incidence of catheter slippage or pull out compared to 30% in adults. They inferred that the higher incidence of catheter complications, including 7 cases of pseudomeningocele in children, was due to lesser subcutaneous tissue and muscle mass, smaller overall size, and less distance between the pump and the catheter entry point at the dura. Pressure dressing, use of acetazolamide, and autologous epidural blood patch helped in the resolution of pediatric lumbar pseudomeningoceles.

In a multicenter study of 209 patients with IDDS, Follett and Naumann (12) reported 49 catheter system complications in 37 cases that included 7 complications related to the catheter per se and 42 related to the implantation procedure. They concluded that the incidence of complications are similar between one piece versus 2-piece Medtronics intrathecal InDura catheters.

Literature lacks information on the consequences of broken and retained intrathecal catheters of the Medtronics system. However, management of such catheters relies on similar guidelines applied to broken and retained epidural catheters. Catheter breakage is reported infrequently following the epidural placement for obstetric and lower extremity anesthesia (18) and similarly Racz catheters could break during caudal epidurolysis (19,20). In most cases retained epidural catheters are asymptomatic, but when symptomatic due to lumbar canal stenosis (21) or causing radicular pain (22) surgical removal is indicated. Similarly, intrathecal catheter tip granulomas causing symptoms due to compression of spinal cord or nerve roots necessitate decompressive laminectomy at an appropriate level with removal of intrathecal catheters (23).

Spinal psuedomeningoceles results from an extradural collection of CSF due to a breach in the dural-arachnoid layer. While true meningocele is always lined by an arachnoid membrane, pseudomeningocele, at least initially, may not be arachnoid lined, and therefore, it is not a true meningocele (24). Psuedomeningoceles are often described in association with CSF fisutulas that result from extradural fluid communication with a cavity like pleura or a direct communication to the exterior (25). By far, the majority of pseudomeningoceles result from iatrogenic incidental durotomy. Other causes include trauma and congenital abnormalities (26-28). Iatrogenic durotomy occurs in between 0.3 and 13% cases of spinal surgery, more frequently with lumbar laminectomy (29,30) with reported incidences of pseudomeningocele between 0.07 and 0.8% (28,31). Avlusion injuries of the brachial and lumbo-sacral plexus could result in the stretching of the nerve roots, subsequent tearing of arachnoid and dural sleeves, and development of a CSF leak and pseudomeningoceles (32). Congenital conditions such as Marfans syndrome and neurofibromatosis have been associated with asymptomatic dural ectsia in up to 67% of cases (33,34).

Pseudomenigoceles are often asymptomatic and may present as fluctuant swellings in the neck thorax or lumbar area. The time interval between the spine surgery and onset of symptoms may vary from months to years (35). Patients may present with postural headaches, localized back pain, or nerve root entrapments producing radicular symptoms (36-38). If the communication between the pseudomeningocele and the intrathecal space is wide enough, the patient may present with a postoperative fluctuant mass that enlarges with Valsalva maneuvers such as coughing or sneezing (24). In this patient, the size of the lumbar swelling increased due to spreading subcutaneous and sub fascial CSF collection and did not transmit increased CSF pressure on Valsalva maneuvers. The retained intrathecal catheter with the entrapment of a broken end in the interlaminar and interspinous level would act as a ball-valve mechanism with the narrow opening allowing CSF in one-way, resulting in tense nonfluctuant swelling (39). In our patient, the capacity for the pseudomeningocele to achieve such high pressure and tension is thought to be secondary to the ball-valve mechanism and perhaps associated arachnoiditis. When the fistulous tract communicates to the exterior, decreased CSF volume along with lowered intracranial pressure results in severe headache from traction on the meningeal blood vessels. External CSF leak results in a halo sign — a light brown halo that surrounds a central stain on bedding or another absorbent surface. This is an ominous sign due to external communication of intrathecal space and is a surgical emergency. In this case report, the multilayer interrupted closure of subcutaneous tissues and skin resulted in prevention of external CSF leak; however, a continuous internal CSF leak leads to the recurrence of tense cystic subcutaneous pseudomeningocele. In rare cases, the authors have described patients with progressive myelopathy due to spinal cord herniation into the pseudomeningocele with cardiac and respiratory effects (39,40).

Clinical diagnosis including history and physical examination is reinforced by several imaging modalities and the analysis of CSF. Postoperative seroma, liquefied hematoma, wound infection, and abscess should be ruled out to reach a definite diagnosis. Magnetic resonance imaging typically demonstrates CSF as low signal intensity on T1-weighted images and high signal intensity on T2-weighted images with a demonstrable level of communication with the thecal sac as well as spinal cord compression or nerve root entrapment (24,41). Computerized Tomographic Myelography visualizes the location of the pseudomeningocele relative to a surgical site because of its superior bone imaging quality compared to MRI (24,42). Retrograde radionuclide myelography and digital subtraction myelography have been successfully used to diagnose pseudomeningocele (43,44).

Laboratory evaluation of aspirated fluid levels of B_2 transferrin, a protein isoform formed by the action of cerebral neuraminidase that is present only in CSF, is a very sensitive and specific test (45). Assessment of CSF glucose levels is an unreliable method, as is comparing the serum and fluid chloride levels (46).

Conservative treatment including bed rest, use of an abdominal binder, and focal compression may be tried in the initial phase of pseudomennigocele of iatrogenic origin (47,48). An epidural blood patch has been successfully used to treat postoperative psuedomeningoceles and CSF fistulas by forming an epidural blood clot, preventing CSF efflux and sealing the dural tear (10,15,16). In our patient an epidural blood patch failed to control the recurrent fluid collection possibly due to the presence of the retained catheter.

Lumbar pseudomeningocele is a rare complication encountered by physicians practicing interventional pain medicine. We strongly recommend neurosurgical consultation when a patient develops gradually

increasing cystic swelling at the site of intrathecal catheter insertion, if the swelling does not respond to epidural blood patch or, if the patient develops CSF fistula and/or new radicular pain symptoms. Surgical repair of the dural defect is the definitive treatment for pseudomeningoceles and CSF fistulas.

Different surgical techniques are described for the repair of dural tears depending on the site and location of the tear or its communication. Some surgeons have treated a small dural communication by merely opening the dura widely to prevent a ball-valve effect, while most surgeons recommend primary closure of the dura using nonabsorbable sutures with a 4-0 to 7-0 tapered needle (24) and in large defects use of additional adjuvant materials — DuraGen, autologous fibrin glue, Tisseal, and BioGlue (46).

Closed subarachnoid drainage decreases the CSF pressure difference between intradural and extradural space and has been successfully used in the treatment of CSF fistulas and pseudomeningoceles with a success rate up to 90% (50,51). Lumbar drains are not without complications and include spinal headaches (60%), meningitis (2.5%), discitis (5%), wound infection (2.5%), transient nerve root irritation, and recurrence (51). Diversion of CSF alone may not be sufficient and is frequently combined with epidural blood patch (49). Ventriculoperitoneal or lumboperitoneal shunt procedures are indicated in cases when the initial surgical repair of dura had failed or in whom surgical repair is not possible due to the location of the dural defect that needs complicated repair (24,52).

Conclusion

This case presents one of the rare complications associated with the implantation of IDDS. If a patient with IDDS develops a complete fracture of the catheter at the spinal end, all attempts should be made to define the 2 ends of the catheter and establish the continuity with a titanium connector. If the spinal end of the catheter is retracted deep into the interspinous ligaments and not recoverable, avoid entering the intrethecal space at the same level. If a patient develops pseudomeningocele in the postoperative period of IDDS and conservative methods including autologous epidural blood patch fail, we recommend an MRI of the spine for a detailed study along with prompt neurosurgical consultation.

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