Baclofen pump catheter leakage after migration of the abdominal catheter in a pediatric patient with spasticity

Amer Dastgir, MD, Nathan J. Ranalli, MD, Theresa L. MacGregor, PhD, and Philipp R. Aldana, MD

Department of Neurosurgery, Division of Pediatric Neurosurgery, University of Florida College of Medicine–Jacksonville, Florida

The authors report an unusual case of intrathecal baclofen withdrawal due to the perforation and subsequent leakage of a baclofen pump catheter in a patient with spastic cerebral palsy. A 15-year-old boy underwent an uncomplicated placement of an intrathecal baclofen pump for the treatment of spasticity due to cerebral palsy. After excellent control of symptoms for 3 years, the patient presented to the emergency department with increasing tremors following a refill of his baclofen pump. Initial evaluation consisted of radiographs of the pump and catheter, which appeared normal, and a successful aspiration of CSF from the pump's side port. A CT dye study revealed a portion of the catheter directly overlying the refill port and extravasation of radiopaque dye into the subfascial pocket anterior to the pump. During subsequent revision surgery, a small puncture hole in the catheter was seen to be leaking the drug. The likely cause of the puncture was an inadvertent perforation of the catheter by a needle during the refilling of the pump. This case report highlights a unique complication in a patient with an intrathecal baclofen pump. Physicians caring for these patients should be aware of this rare yet potential complication in patients presenting with baclofen withdrawal symptoms.

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KEY WORDS baclofen pump; intrathecal; perforation; leakage; baclofen withdrawal; spasticity; catheter migration; functional neurosurgery

BACLOFEN is an agonist for GABA B receptors and is mainly used in the treatment of spasticity.^{15,20} It is an analog of γ -aminobutyric acid, which can inhibit the release of excitatory neurotransmitters in the central nervous system.^{7,30,39} Baclofen can be given orally or intrathecally through the use of an intrathecal pump, which uses a catheter placed in the spinal subarachnoid space.⁴¹ Intrathecal baclofen delivery via a surgically implanted pump placed in the abdomen began in the 1990s as a means of reducing spasticity due to cerebral palsy.

Intrathecal baclofen is far more effective than the oral version. In addition, many patients do not develop a significant tolerance to intrathecal baclofen. Even after years of continued usage, these patients still benefit from the drug's therapeutic antispasmodic effects.¹⁷ Discontinuation of baclofen can result in withdrawal symptoms, especially if the medication has been used continuously over a long period of time. These withdrawal symptoms can mimic alcohol or benzodiazepine withdrawal. Malfunction of the implanted pump or intrathecal catheter results in withdrawal. Symptoms can include but are not limited to dizzi-

ness, disorientation, agitation, hypertonia, spasticity, rigidity, and seizures. Baclofen withdrawal symptoms can be minimized by tapering down the medication.²⁷ Malfunction of the pump hardware and catheter (21%–33%) and infection (9%–10%) are the most common complications in patients. Catheter-related problems are the most common cause of hardware malfunction.^{5,7,8,16,44}

Case Report

History and Examination

This 15-year-old male patient with a history of cerebral palsy had an intrathecal baclofen pump implanted almost 3 years previously for the management of spasticity. He presented to the emergency department with a history of increasing tremors 2 days after the refill of his baclofen pump. The patient's mother reported tremors in his right leg with progression to the other leg and both arms. Physical examination revealed generalized tremors in all extremities increasing with movement.

Plain radiographs suggested a normal-appearing pump

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and intact catheter. Subsequently, the side port of the baclofen pump was accessed, and 2 ml of clear CSF was aspirated. A CT dye study revealed extravasation of radiopaque dye into the subfascial pocket surrounding the pump, with a portion of the catheter seen directly overlying the pump refill port (Fig. 1). The patient was brought into the operating room for exploration and revision of the pump and catheter.

Operation

The patient was taken to the operating room and placed under general anesthesia. The abdominal baclofen pump and catheter were surgically dissected. The pump tubing was found to cross directly over the refill reservoir of the pump and had an active leakage of the drug from a small puncture hole in the pump catheter just proximal to the straight connector (Fig. 2). The pump was removed, and the leaking segment of the catheter along with a few centimeters of proximal and distal tubing were removed and replaced. The pump was then reimplanted with the catheter positioned posterior to the pump.

Postoperative Course

Postoperatively, the patient experienced a reduction in baclofen withdrawal symptoms including spasticity, and he returned to his clinical baseline. The patient was discharged home 48 hours later in good condition.

Literature Review

We performed a literature search using PubMed and the search terms “baclofen pump catheter,” “baclofen pump catheter malfunction,” “baclofen pump catheter leakage,” and “baclofen pump case report.” There were no language exclusions. To include any relevant publications, additional studies were traced by checking the reference lists of selected articles.

Discussion

This case report highlights a previously unreported

FIG. 1. Axial postcontrast CT scan of the abdomen demonstrating intrathecal baclofen pump with extravasation of radiopaque dye into the subfascial compartment anterior to the pump; the catheter is seen directly overlying the refill port. Figure is available in color online only.

FIG. 2. Intraoperative picture of the intrathecal baclofen pump with the pump catheter crossing over the refill reservoir of the pump. Leakage of the drug can be seen from a small hole in the catheter. Figure is available in color online only.

complication of a baclofen catheter perforation during pump refill causing acute baclofen withdrawal in a patient.

Intrathecal baclofen is commonly used to treat spasticity, resulting in a better quality of life for patients.^{1,2,9,11,23,24,26,33,43} Although it has been very effective in treating spasticity, the therapy does entail certain risks,^{4,14,21,22} with approximately 25%–30% of patients experiencing a complication.^{4,22–24} These complications can be from dosing (overdose or underdose), mechanical failure of the pump or catheter, and infection of the hardware or local tissues.^{4,14,21,22,44} Infections, leakage of CSF, and catheter-related issues are the most common complications.^{1,33,44,45} In patients on intrathecal baclofen, withdrawal symptoms can occur when the medication dosage is decreased or abruptly stopped.^{3,6,10,11,14,19–22,25,31,32,37,40,47} Watve et al. observed that intrathecal baclofen withdrawal is frequently due to catheter malfunction.⁴⁶ Baclofen withdrawal can be characterized by reflex spasticity, seizures, central nervous system depression, and hyperthermia, which may progress to disseminated intravascular coagulation and organ failure if left untreated.^{3,10,14,20,21,25,32,47} Baclofen withdrawal symptoms can begin within the first 3 days after slowing or stopping the medication, whether orally or intrathecally.^{10,29,38,39,46,47} These symptoms are likely caused by the removal of the inhibitory tone on the GABA B receptors.^{10,38,46}

Catheter-related complications are the most common type of complication associated with intrathecal baclofen pumps.^{34,35} Catheter complications have numerous etiologies, including breakage, dislodgment, kinks, fibrous blockages, and intraoperative iatrogenic nicks.^{9,34–37} Taira et al. reported that of 400 patients who underwent placement of an intrathecal baclofen pump in Japan between 2005 and 2011, 6 (1.5%) of them had a breakage in the pump catheter.⁴² In another study, 4 of 119 pediatric patients (3%) in the same hospital with an intrathecal baclofen pump suffered a catheter break.¹⁸ Dickerman et al. reported that out of 110 patients at their institution with an intrathecal baclofen pump, 3 (2.7%) required a revision of the pump due to a catheter breakage.¹³ Dickerman and Schneider, in a separate paper, observed a catheter break in the pump catheter neck in one of their patients, on 2 separate occasions 6 months apart.¹² The catheter neck was found to be lying over the iliac crest during both surgeries in this patient, which led the authors to conclude that repetitive compression against the iliac crest was the source of the catheter break.¹² However, to the best of our knowledge there is no documented case of the perforation of an intrathecal pump catheter during the medication-refilling process.

Although many causes must be considered in the differential diagnosis of baclofen withdrawal, pump catheter malfunction must be given ample consideration. In our case, the location of the catheter directly overlying the refill port predisposed it to needle puncture during the refill process. Although the catheter was not positioned there during the original implantation, it somehow migrated over time. Having prior knowledge of this to inform the staff performing the pump refill would have been useful, but we are not sure if this complication would have been permanently avoided given the need for repeated refills. In general, radiography or CT imaging can be performed on a routine basis to check for catheter migration, in particular near the port of the pump, prior to refill. However, given the rarity of the complication documented in this case report and the long-term risks of radiation exposure in childhood, we do not recommend it. This case does illustrate the utility of a CT dye study in the evaluation of a patient with a baclofen pump exhibiting withdrawal symptoms, following negative radiographs and pump side port aspirations.

The technique used during this particular refill was similar to that in previous refills. The patient was placed supine for the procedure, the abdominal region was exposed, and the pump was checked electronically. The area was then cleaned with an antiseptic solution, and the site was isolated for sterility using a drape. A 22-gauge needle was inserted into the refill port of the pump located in the center of the device. Any old remaining medication was drawn out, and the new medication was injected. After removing the needle, a dressing was applied to the puncture site. Finally, the pump was reprogrammed. There was no “rolling” feeling or anything else out of the ordinary during the refilling process. Multiple punctures were not required. In general, baclofen pump refills are performed every 1–6 months depending on the dosage of the drug, concentration of the drug, and size of the pump that the

individual patient had implanted. This modality of treatment is flexible in that the rate of intrathecal infusion can be automatically adjusted throughout the day. Refills are scheduled based on calculations made by the programming device, as was the case for our patient. Baclofen tends to break down if left in the pump for longer than 6 months. Our patient received his refills approximately every 3 months, as was the case with this particular refill.

We theorize that the abdominal catheter migrated from the time of insertion to the time of inadvertent puncture. Although there is not a previous CT scan, the immediate postoperative radiograph from 2012 (Fig. 3) and the scout radiograph from a preoperative CT performed prior to the catheter revision in 2014 (Fig. 4) show the pump in a stable position but with repositioning of the tubing from behind the pump in 2012 to the front of the pump in 2014. Although there is not a formal algorithm documented in the literature, we recommend simple palpation for the presence of the catheter over the refill port by the individual refilling the pump prior to insertion of the needle. The health care provider doing this should pay particular attention to the possibility of the catheter migrating. In the event that the tubing is suspected to be over the refill port or the tubing is palpable anterior to the pump, imaging studies, such as screening anteroposterior and lateral radiographs, are warranted.

Conclusions

This case illustrates the need for physicians treating patients using an intrathecal baclofen pump to be vigilant in considering all possible causes of baclofen withdrawal symptoms in their patients. Surgeons placing these pumps could consider leaving a smaller length of catheter in the abdominal compartment behind the pump to reduce the

FIG. 3. Immediate postoperative kidney, ureter, and bladder (KUB) radiograph obtained on September 13, 2012, after insertion of the pump. There is no evidence of the catheter anterior to the abdominal pump.

FIG. 4. Preoperative KUB radiograph obtained on August 15, 2014, before revision of the pump catheter, suggesting that the catheter is now anterior to the abdominal pump traversing the refill port.

chance of the catheter slipping out from behind the pump and ending up anterior to the pump. Li et al. emphasized minimizing the length of the catheter outside of the spine as a means of decreasing the likelihood of catheter movement.²⁸ Less tubing can lead to an increased likelihood of the catheter slipping out of the intrathecal space as the child grows. However, this must be balanced against the increased risk for catheter migration in the setting of excessive intraabdominal tubing. We believe that our report adds to the existing literature on the complications of intrathecal baclofen pumps, which can aid physicians in optimizing treatment plans for patients who are treated using these pumps by making them aware of potential complications.

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Author Contributions

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Correspondence

Amer Dastgir, Lucy Gooding Pediatric Neurosurgery Center, Wolfson Children's Hospital, 836 Prudential Dr., Ste. 1205, Jacksonville, FL 32207. email: amerdastgirmd@hotmail.com.