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Case Report

Management of an intrathecal baclofen pump pocket empyema caused by a proximal vesicocutaneous fistula: A case report

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ABSTRACT

Background: Intrathecal baclofen infusing pumps are nowadays commonly implanted in patients suffering from severe, intractable spasticity with a background of multiple sclerosis. Although intrathecal baclofen therapy is considered a safe therapeutic modality, complications are unavoidable and broadly categorized as mechanical and infectious. In the instance of a pump pocket infection, a surgical explanation of the pump is often necessary to treat the infection.

Case Description: We present the rare case of a 60-year-old woman who was admitted emergently to our clinic with a subcutaneous pump pocket empyema caused by proximal vesicocutaneous fistulas. The patient underwent explantation of the pump and otherwise had an uncomplicated perioperative course.

Conclusion: The surgical explanation of the baclofen pump and antibiotic treatment were sufficient to treat the pump pocket empyema in this instance. To the best of our knowledge, this is the first report of a pump pocket empyema formed in the proximity of a vesicocutaneous fistula.

Keywords: Baclofen pump, Baclofen pump pocket empyema, Spasticity, Vesicocutaneous fistula

INTRODUCTION

Intrathecal pumps are considered a rather safe method of infusing baclofen to treat patients with severe intractable spasticity.[2,3,10] Nevertheless, complications could occur and could be categorized either as infectious or as mechanical. Infection of the pump pocket is one of the most common complications, and in the instance that it is resistant to antibiotic treatment, pump removal may be essential.[7,19]

The scope of our study is to report the rare case of a patient who developed an empyema of her baclofen pump pocket and infection of the surrounding subcutaneous tissues due to a newly formed proximal vesicocutaneous fistula. The patient had her pump implanted due to severe intractable spasticity manifested in the background of multiple sclerosis (MS). She underwent emergent surgical debridement and removal of the baclofen pump, followed by a course of intravenous antibiotics. Her perioperative course was uncomplicated, and she eventually was

discharged home. A literature review (PubMed and Scopus) did not return similar cases where a vesicocutaneous cyst is the primary source of a pump pocket empyema.

CASE DESCRIPTION

A 60-year-old Caucasian female patient had a baclofen infusion pump implanted two years ago to treat severe spasticity and dystonia due to advanced MS. The patient had a medical history of a cesarian section 32 years ago and was diagnosed with MS 21 years ago. MS caused her severe urinary incontinence and neurogenic detrusor overactivity. During the past two years, she suffered from multiple episodes of lower urinary tract infections (UTIs) and required multiple hospitalizations. The UTIs, in conjunction with the history of cesarean section and bladder overfill, lead to the formation of a vesicocutaneous fistula (fistula connecting the urinary bladder to the skin) at the midline of the lower abdominal area 6 months ago [Figure 1a]. Two weeks before her admission to our Clinic, a second fistula was formed, which was in close proximity to the implanted baclofen pump [Figure 1a]. Thus, she developed a high fever and was admitted initially to an Internal Medicine Clinic, diagnosed with a recurring lower UTI. Her urine cultures were positive for three bacterial organisms, *Klebsiella pneumoniae*, *Proteus mirabillis,* and *Escherichia coli*, which were susceptible to first-line antibiotics. Hence, the patient was treated medically and underwent an abdominal computed tomography (CT)-scan, which revealed a collection of fluid surrounding the baclofen pump [Figure 1b]. Unfortunately, due to poor response to the conservative treatment, the patient was referred to our neurosurgical department and due to high suspicion of a pump pocket empyema, she was transferred to our hospital. Her physical examination revealed a tense baclofen pump pocket with overlying redness of the skin [Figure 2a]. Thus, the patient underwent an emergent surgical exploration of the pump pocket. During the operation, empyema was confirmed [Figure 2b], and approximately 80 mL of inflammatory fluid were evacuated from the pocket [Figure 2c]. The baclofen pump and the catheter were removed and sent for cultures, along with a cerebrospinal fluid (CSF) sample. During the operation, urological consultation was requested, but the attending urologist did not consider a surgical intervention for the fistulas in this instance. Thus, the subcutaneous tissues were thoroughly debrided, and the surgical incisions were sutured. The perioperative course of the patient was uncomplicated. She had a 14-day course of meropenem and colistimethate while the fever subsided and the inflammatory markers returned to normal levels. To prevent a sudden increase in her spasticity and baclofen withdrawal syndrome, oral baclofen and a short course of diazepam were administered.[11] The culture of the pump and the proximal catheter developed the same bacteria as her urinary culture; however, the CSF and intrathecal catheter cultures were negative. Hopefully, a central nervous system infection (CNS) was prevented. After two weeks of hospitalization and assessment by the infectious disease team, she was discharged home in good overall condition but with an increased spasticity score measured in the modified Ashworth Scale.

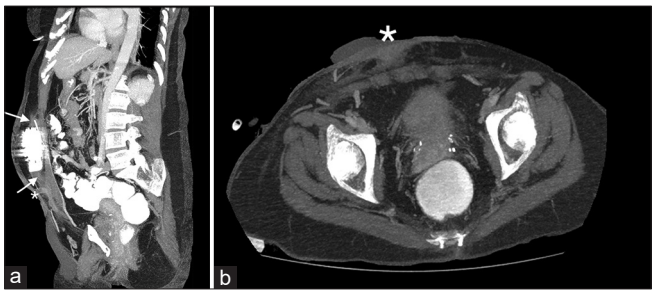

Figure 1: Preoperative abdominal computed tomography scan with intravenous and per os contrast. (a) Sagittal cut demonstrating the subcutaneously implanted baclofen pump, the surrounding pus and inflammation (white arrows) and the exit point of the proximal vesicocutaneous fistula (white asterisk). (b) Axial cut showing the exit point of the suprapubic midline vesicocutaneous fistula (white asterisk) .

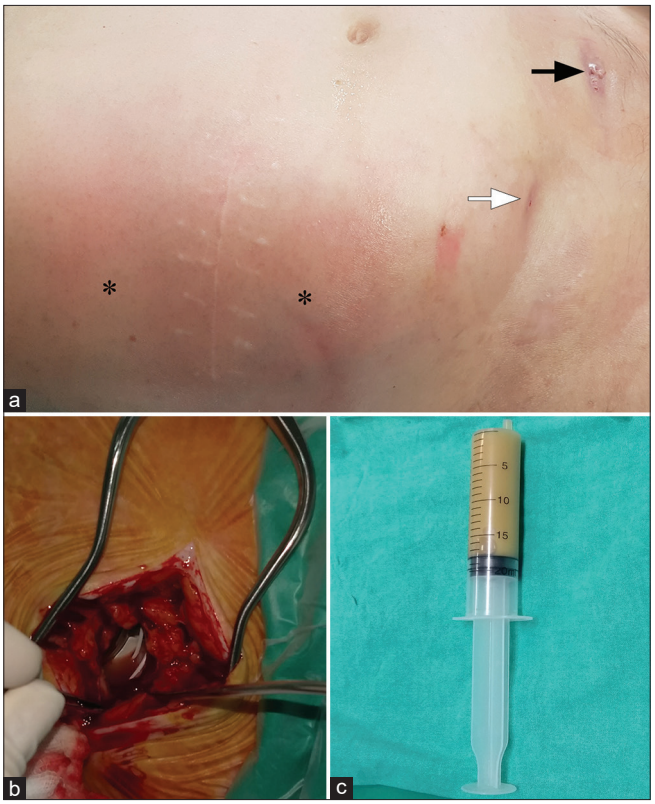

Figure 2: (a) Suprapubic midline vesicocutaneous fistula (black arrow), proximal vesicocutaneous fistula (white arrow) to the pump pocket and skin erythema (black asterisks) surrounding the implantation scar. (b) Intraoperative picture of the pump pocket filled by the empyema. (c) Evacuated pus from the pump pocket.

DISCUSSION

Spasticity manifests after impairment of the upper motor neurons of the corticospinal tract due to abnormal supra-spinal driving of spinal reflexes and affects around 84% of patients suffering from MS.[18] Baclofen is a structural analogue of gamma-aminobutyric acid (GABA), which crosses the blood-brain barrier and binds to pre- and post-synaptic GABA receptors, inhibiting the activity of motor-neurons and inter-neurons.[15] The maximum daily dosage of oral baclofen could be up to 100 mg daily. When this high oral dosage is inadequate to effectively reduce severe spasticity, intrathecal admiration through an infusion pump should be considered. The pump delivers baclofen directly to the CNS, bypassing the blood-brain barrier and thus allowing high concentrations of the drug with daily doses as low as 1% in comparison to the oral dosage.[13,15]Intrathecal baclofen pumps are surgically implanted in the abdominal wall, subcutaneously or subfascially, and a catheter is tunneled from the pump to the lumbar spine, where it enters and rests in the intrathecal space.[6] Our surgical preference is to implant the pump subcutaneously in the left lower abdominal quadrant and insert the catheter through a para-spinal approach. Implantation of baclofen pumps comes with an overall complication rate of 23–25%.[16,17] This rate is variable and depends on factors such as age, type, and severity of the underlying disease.[7,14,17] Complications are categorized into two broad groups: infectious or mechanical.[9,14] Mechanical complications carry a risk of 4–24%,[19] with the most common to be catheter-related.[17] Infectious complications have been reported in the contemporary literature within a range of 5–26%, and they tend to occur earlier than their mechanical counterparts.[4,5,7,9,14] The most commonly encountered infectious agent is *Staphylococcus aureus*.[4,7]

Vesicocutaneous fistulas are very rare clinical entities, which divert the urinary stream from the urinary bladder directly to the overlying skin. Risk factors for the formation of such fistulas include gynecological surgery or traumatic injuries,[1] UTIs and urinary retention (neurogenic or structural).[12,20] Consistently, our patient's medical history included a cesarean section, recurrent UTIs, and bladder distention due to her MS. Most probably, all these factors contributed to the formation of the two vesicocutaneous fistulas. The second fistula occurred in proximity to the pump and eventually caused seeding of the urinary tract infection to the pocket and the surrounding tissues [Figure 2a].

Based on the apparent inflammation of the soft tissues and the skin erythema surrounding the implantation site, as well as the CT findings, the patient was transferred to our clinic for further management. Another alarming factor was the poor response to the antibiotic treatment until then. Since an empyema was suspected, our clinical decision was emergent surgical debridement and explanation of the pump apparatus. Two surgeons were involved in the operation, one on the abdominal side and one on the lumbar side. Each one of them used a different sterile set and was assisted by a different scrub nurse. Intraoperatively, an empyema was confirmed, and 80 mL of pus was drained from the pump pocket [Figures 2b and c]. The catheter was incised in the lumbar side and withdrawn from the abdominal side. This configuration and surgical technique were necessary to contain the infection on the abdominal side. Cultures from the pocket empyema came back positive for *K. pneumoniae*, *P. mirabillis,* and *E. coli*. Since the same bacteria was also found in her urinary cultures, we speculated that the proximal fistula leaked content within the pocket, and the urinary infection spread there, forming the empyema. Fortunately, cultures of the CSF and the intrathecal catheter were negative. Thus, CNS infection was excluded. After a course of antibiotics, the patient was discharged home in good general condition.

CONCLUSION

Patients with implanted baclofen pumps usually have comorbidities or disabilities and other medical problems.[8] In this group of patients, meticulous and prompt medical care is essential to prevent baclofen pump-related complications. In our case, a vesicocutaneous fistula near the pump pocket leaked infected urinary content within the pocket, forming an empyema. Emergent surgical debridement and explanation of the hardware, in combination with the antibiotic treatment, prevented a severe complication, such as a CNS infection. This is an extremely rare complication and actually, to our knowledge, the first described in the literature. In similar instances where a potential infectious source appears near the pump, like the fistula in our case, aggressive management is recommended to contain the infection and thus prevent hardware removal and/or CNS involvement.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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