**Streptococcus dysgalactiae-related intrathecal baclofen therapy infection: how to avoid withdrawal?**

V. De Larminat¹, S. Zayet², T. Klopfenstein² and M. Idelcadi¹  
¹) Pain Department and ²) Infectious Diseases Department, Nord Franche-Comté Hospital, Trevenans, France

**Abstract**

Intrathecal baclofen therapy is commonly used for neurologically spastic patients. One of the major complications is hardware infection, which generally requires urgent removal of the pump and the intrathecal catheter, with the risk of severe baclofen withdrawal. We have recently been facing this situation and propose another solution with adapted antibiotic therapy, removal with immediate replacement of the intrathecal catheter, initially connected to an implanted port to continue baclofen administration. A new pump was secondarily implanted, after successful treatment of acute bacterial meningitis due to *Streptococcus dysgalactiae*.

© 2021 The Author(s). Published by Elsevier Ltd.

**Keywords:** Infection, intrathecal baclofen therapy, meningitis, sepsis, *Streptococcus dysgalactiae*

**Original Submission:** 12 March 2021; **Accepted:** 16 March 2021  
**Article published online:** 24 March 2021

**Clinical case**

A 69-year-old woman has been paraplegic since the age of 19 years, after a public road accident. She was implanted for intrathecal therapy in 2008 with an Ascenda catheter and Synchromed II pump (Medtronic, Dublin, Ireland), initially for baclofen alone, and then associated with morphine for neuropathic pain. Doses were unchanged for 10 years, with daily administration of 300 μg of baclofen and 400 μg of morphine (once a day). Fillings were scheduled every 10 weeks. She is a very autonomous and dynamic woman, performing urinary catheterizations herself, daily re-education and transfers in her wheelchair. Pump replacement was performed in 2015, without any trouble. Progressively thereafter, a lymphatic collection settled around the pump, requiring regular aspirations of up to 200 mL of yellow liquid before filling, to gain access to the filling port. Gradually, after several months, the diffusion dried up, simplifying subsequent fillings, from 2017.

On 22 June (day 1) the woman presented to the Emergency Department with a 24-hour history of fever, chills and fatigue. On admission, neurological examination showed a Glasgow Coma Scale score of 15/15 (E = 4, V = 5, M = 6) with no neck stiffness or focal abnormalities. She only complained about nausea and a cloudy liquid issuing from a small hole (through which the pump was apparent) in the area of the pump with an indurated zone surrounding it. Quick sequential organ failure assessment score (qSOFA) was evaluated on two occasions. Routine laboratory findings showed elevated C-reactive protein (150 mg/L).

Antibiotherapy was begun immediately after taking blood samples and a fluid sample from the pump scar for cultures. Lumbar puncture was deferred because of anticoagulant treatment (fluindione) introduced years ago for repetitive phlebitis. Antimicrobial drugs were vancomycin (15 mg/kg loading dose then 60 mg/kg/24 h given as a continuous infusion) and cefotaxime (300 mg/kg/24 h by intravenous infusions six times daily).

**Introduction**

Intrathecal baclofen infusion is an effective solution for neurological patients with spastic disorders that are often difficult to control with oral administration [1–3]. This remains an invasive procedure that requires surgical implantation and regular filling with sterile preparation. One of the major complications is material infection [4,5], which generally requires urgent removal of the pump and the intrathecal catheter, with the risk of severe baclofen withdrawal. We have recently faced this problem, and proposed another management of this situation. To our knowledge, we report herein the first case of intrathecal baclofen therapy infection caused by *Streptococcus dysgalactiae*, in France.
The challenge was to treat an infection on neurological material that required its urgent removal, and, if performed, withdrawal of baclofen, which could also be life-threatening.

As the infection tolerance was excellent with very few general and meningeal signs, with rapid decrease of fever and biological inflammatory syndrome within the first 24 h (C-reactive protein 80 mg/L), it was proposed attempt medical treatment after local surgical cleaning of the site of the pump on 23 June (day 2). A cerebrospinal fluid (CSF) sample was collected, thanks to the direct access to the catheter at the top of the pump.

Analysis of the purulent CSF revealed an elevated white blood cell count of 660/mm³ with 90% neutrophils with elevated protein concentration (4.31 g/L) and low glycorrhachia (glucose concentration 0.8 mmol/L).

Blood cultures were negative. Direct examination of CSF revealed Gram-positive cocci and the culture isolated a Strep-
tococcus dysgalactiae. PCR amplification and sequence analysis determined the same pathogen. The S. dysgalactiae was a strain susceptible to all antibiotics on antimicrobial susceptibility testing. Culture of a scar swab also isolated an S. dysgalactiae. Follow-up CSF culture after administration of antibiotics was sterile. After these results, antibiotics were switched to amoxicillin (200 mg/kg by six intravenous infusions daily) and rifampicin (900 mg daily, orally). The patient was initially hos-

pitalized in the intensive care unit for 3 days, and then trans-

ferred to the infectious diseases department.

Progress was initially favourable, with decrease of fever, disappearance of nausea and headache, and biological improvement with regression of the inflammatory syndrome. Never-

theless, the persistence of a febricula and the absence of biological normalization led us to perform the ablation of the pump and intrathecal catheter on 30 June (day 9). During the same operation, a new intrathecal catheter (Ascenda 8780, Medtronic) was placed, tunnelled to the right hypochondrium and linked to an implanted port (Celsit). This port was connected to an external pump, which allowed the continuous administra-
tion of the intrathecal therapy at the usual doses for this patient.

On 8 July (day 17), replacement of the implanted port by a new Synchromed II pump was decided on, considering the favourable clinical and biological evolution after 16 days of effective anti-

biotherapy. The port was disconnected from the catheter, which was tunnelled up to the new position of the implanted pump, on the opposite abdominal side to the initial site. The patient was discharged for outpatient follow up after 18 days of hospitaliza-
tion. Antibiotic therapy was prolonged at home up to 15 full days after the removal of septic material. Two months after the end of the antibiotics the woman was seen (for filling of her pump); she was cured of this acute episode, despite some difficulties in total recovery of her autonomy.

Discussion

Intrathecal therapy through an implanted pump was first described in 1981 by Onofrio et al., for morphine delivery in chronic cancer pain [6]. Since then, intrathecal administration through an implanted device has been expanded to other drugs such as other morphinomimetics, local anaesthetics, ziconotide, all for analgesic purposes, and baclofen for chronic spasticity [7].

Intrathecal baclofen is now a well-known route of adminis-

tration for the management of widespread spasticity [1], whether unresponsive to conservative treatments, or for pa-
tients with intolerable medication side effects at their thera-
pic dosage [3]. This hydrophilic drug acts on receptors in the dorsal grey matter of the spinal cord, and direct infusion into the CSF concentrates the drug where it is needed for its therapeutic effect, allowing the dose to be decreased down to 1% of the oral posology, and minimizing the secondary effects.

Baclofen intrathecal withdrawal is a potentially life-

threatening event, which may occur due to different factors such as hardware dysfunction (catheter rupture or plicature, pump malfunction) or problems with reservoir refills (non-

compliance of the patient, dosing errors or refill errors).

Clinical symptoms generally appear within a few hours to days—including high fever, altered mental status, seizures, muscle rigidity with rebound spasticity—and can lead up to severe rhabdomyolysis, multiple organ failure and death. Relay by oral or enteral administration is insufficient to halt the progression of withdrawal syndrome because of poor pene-

tration into the central nervous system of the large doses required to be equivalent to the intrathecal dosage [2]. The usual treatment involves hospitalization in an intensive care unit, sedation and muscle relaxation (with benzodiazepines or curare), which requires intubation for airway protection and artificial ventilation. A promising therapy could be the use of dexmedetomidine, with a superior respiratory safety profile than the association of benzodiazepine with propofol, and the avoidance of intubation [8].

In case the of bacterial meningitis on implanted material, the widespread attitude is to remove all the material as soon as possible, to assure an adapted antibiotic treatment and wait at least 7 days (up to 14 days depending of the microorganism in question) and for sterile CSF before a new implantation. The total duration of antibiotics ranges from 14 to 21 days depending on the evolution and the microorganism responsible [9,10].

The use of a temporary intrathecal port was previously described in case of weaning or mechanical dysfunction of the hardware [11], but never, to our knowledge, when it was associated with sepsis.
Conclusion

This case report demonstrates that temporary use of an external pump connected to a port and intrathecal catheter may be a useful alternative to avoid intrathecal therapy withdrawal, even in the case of sepsis.

It remains to be determined for how long antibiotherapy should be administered before removal of the material and how long we should wait before implantation of a new material. There are only old recommendations with a low evidence level.

Funding sources

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Conflict of interest

The authors have stated that there are no conflicts of interest.

References

[1] Ochs G, Struppler A, Meyerson BA, Linderoth B, Gybels J, Gardner BP, et al. Intrathecal baclofen for long-term treatment of spasticity: a multi-centre study. J Neurol Neurosurg Psychiatr 1989;52:933–9.

[2] Ross JC, Cook AM, Stewart GL, Fahy BG. Acute intrathecal baclofen withdrawal: a brief review of treatment options. Neurocrit Care 2011;14:103–8.

[3] Francisco GE, Yablon SA, Schiess MC, Wiggs L, Cavalier S, Grissom S. Consensus panel guidelines for the use of intrathecal baclofen therapy in poststroke spastic hypertonia. Top Stroke Rehabil 2006;13:74–85.

[4] Koljonen PA, Chan SHS, Liu T, Ho ACC, Chim S, Tsoi NS, et al. Intrathecal baclofen pump infection with meningitis: effective treatment by radical debridement and intrareservoir baclofen-vancomycin co-infusion. Neuromodulation J Int Neuromodulation Soc 2021 Feb 3. Epub ahead of print.

[5] Balaratnam MS, Donnelly A, Padilla H, Simeoni S, Bahadur S, Keenan L, et al. Reducing intrathecal baclofen related infections: service evaluation and best practice guidelines. Neuromodulation J Int Neuromodulation Soc 2020;23:991–5.

[6] Onofrio BM, Yaksh TL, Arnold PG. Continuous low-dose intrathecal morphine administration in the treatment of chronic pain of malignant origin. Mayo Clin Proc 1981;56:516–20.

[7] Deer TR, Pope JE, Hayek SM, Bux A, Buchser E, Eldabe S, et al. The Polyanalgesic Consensus Conference (PACC): recommendations on intrathecal drug infusion systems best practices and guidelines. Neuromodulation J Int Neuromodulation Soc 2017;20:96–132.

[8] Gottula AL, Gorder KL, Peck AR, Renne BC. Dexmedetomidine for acute management of intrathecal baclofen withdrawal. J Emerg Med 2019 Nov 20. Epub ahead of print.

[9] Affana CK, Vu M, Riel-Romero R, Nanda A, Sun H. A rare case of pseudomonas meningitis following intrathecal baclofen pump placement in a 28-year-old paraplegic patient. Interdiscip Neurosurg Adv Tech Case Manag 2018;13:82–4.

[10] Tunkel AR, Hartman BJ, Kaplan SL, Kaufman BA, Roos KL, Scheld WM, et al. Practice guidelines for the management of bacterial meningitis. Clin Infect Dis Off Publ Infect Dis Soc Am 2004;39:1267–84.

[11] Duhon BS, MacDonald JD. Infusion of intrathecal baclofen for acute withdrawal. Technical note. J Neurosurg 2007;107:878–80.