American Journal of Case Reports

ISSN 1941-5923
© Am J Case Rep, 2016; 17: 957-962
DOI: 10.12659/AJCR.901157

Received: 2016.08.21
Accepted: 2016.10.05
Published: 2016.12.16

Intrathecal Analgesic Drug Delivery is Effective for Analgesia in a Patient with Post-Poliomyelitis Syndrome: A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

ABCDEF **Cornelis W.J. van Tilburg**

Department of Anesthesiology, Bravis Hospital, Bergen op Zoom, The Netherlands

Corresponding Author: Cornelis W. J. van Tilburg, e-mail: vtilburg@ziggo.nl
Conflict of interest: None declared

Patient: Female, 45
Final Diagnosis: Post-poliomyelitis syndrome
Symptoms: Chronic pain
Medication: Fentanyl • Oxycodone • Gabapentin • Naproxen • Paracetamol
Clinical Procedure: Intrathecal analgesic drug delivery
Specialty: Anesthesiology

Objective: Unusual setting of medical care

Background: Post-poliomyelitis syndrome (PPS) is a progressive neuromuscular syndrome, with chronic pain being one of the most prevalent symptoms. We present a case report on intrathecal analgesic drug delivery to diminish chronic, refractory pain in a patient with PPS.

Case Report: In a wheelchair-bound 45-year-old female patient (Caucasian, body mass index [BMI] 20.5) with severe chronic, refractory pain, a Synchromed® II pump (Medtronic, Minneapolis, Minnesota, USA) was implanted after multidisciplinary consultation and a successful trial period. After 8 months, relocation of the pump due to regional pressure problems with surrounding erythema had to occur. A second pump relocation due to pressure problems and skin erosion was needed 18 months after the first relocation, moving from the abdominal wall to the sheath of the rectus abdominis muscle, resulting in resolution of the problems.

Conclusions: In patients with PPS, intrathecal analgesic drug delivery can be an option to treat chronic, refractory pain. Multidisciplinary consultation is necessary to deal with the wide variety of problems in these patients. Skin problems at the site of the pump reservoir can be challenging and time-consuming and, ultimately, can necessitate relocation (or removal) of the device.

MeSH Keywords: Infusions, Spinal • Poliomyelitis • Postpoliomyelitis Syndrome • Infusion Pumps, Implantable • Drug Delivery Systems

Full-text PDF: http://www.amjcaserep.com/abstract/index/idArt/901157

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Background

Post-poliomyelitis syndrome (PPS) is a progressive neuromuscular syndrome, with chronic pain being one of the most prevalent symptoms [1]. Polio now no longer poses a major threat in developed countries, but many people still live with the sequelae [2,3]. Scoliosis is well known to many polio survivors as a result of paralysis or weakness. This may again lead to all sorts of problems such as pain, decreased pulmonary function, decreased digestion, and reduced use of the arms [4]. Survivors may experience a gradual decline in function and strength over many years [5]; other patients experience progressive disability with symptoms like muscle weakness and muscle atrophy, fatigue, pain, and decreased ambulation. These symptoms may occur 30 to 40 years after the original illness [6,7].

Chronic pain is one the most prevalent symptoms in post-polio patients [1,8–10]. In a retrospective article, Gawne and Halstead reported that pain was present in 95% of patients seen at the post-polio clinic [11]. The use of a multidisciplinary team is advocated for pain relief and functional restoration. A variety of medications have been used with varying success. In the case of severe, refractory pain, placement of an intrathecal drug delivery system (ITDD) may provide an alternate route of delivery compared with systemic opioids with their associated side effects [12–14]; ITDD may offer a way to achieve significant and lasting pain relief with the fewest possible side effects [15,16].

Case Report

A 45-year-old female (Caucasian, body mass index [BMI] 20.5) was referred to the pain center by her general physician because of chronic, refractory, and widespread pain, predominantly occurring at the lumbar spine and radiating into the lower limbs. Medical history revealed poliomyelitis at the age of 7, severe scoliosis and extensive orthopedic surgery (Figure 1A, 1B), lower-limb paresis (wheelchair bound) and incontinence as a result of PPS, osteoporosis, arterial hypertension, and a cochlear implant. Sitting down in the wheelchair was difficult because of her severe chronic pain, and as a consequence, she was bed bound. She was treated with transdermal fentanyl 100 µg/h (equivalent to 240 mg of oral morphine per day), oral oxycodone 5 mg (equivalent to 10 mg of oral morphine) prn, naproxen 500 mg twice daily, gabapentin 300 mg twice daily, and paracetamol 1 g four times daily, which unfortunately did not result in sufficient pain relief (verbal Numerical Rating Scale [NRS] for pain 9/10 when sitting down and 5/10 when lying down). We considered the limited possibilities together with the patient and her family and together decided to initiate the flowchart procedures for intrathecal drug delivery (ITDD; Figure 2). Particular attention was paid to technical failure and multidisciplinary consultation. The rehabilitation physician increased her sedentary activities by adjusting the wheelchair. Psychiatric consultation revealed a minor adjustment disorder. After consultation from an urologist, because of

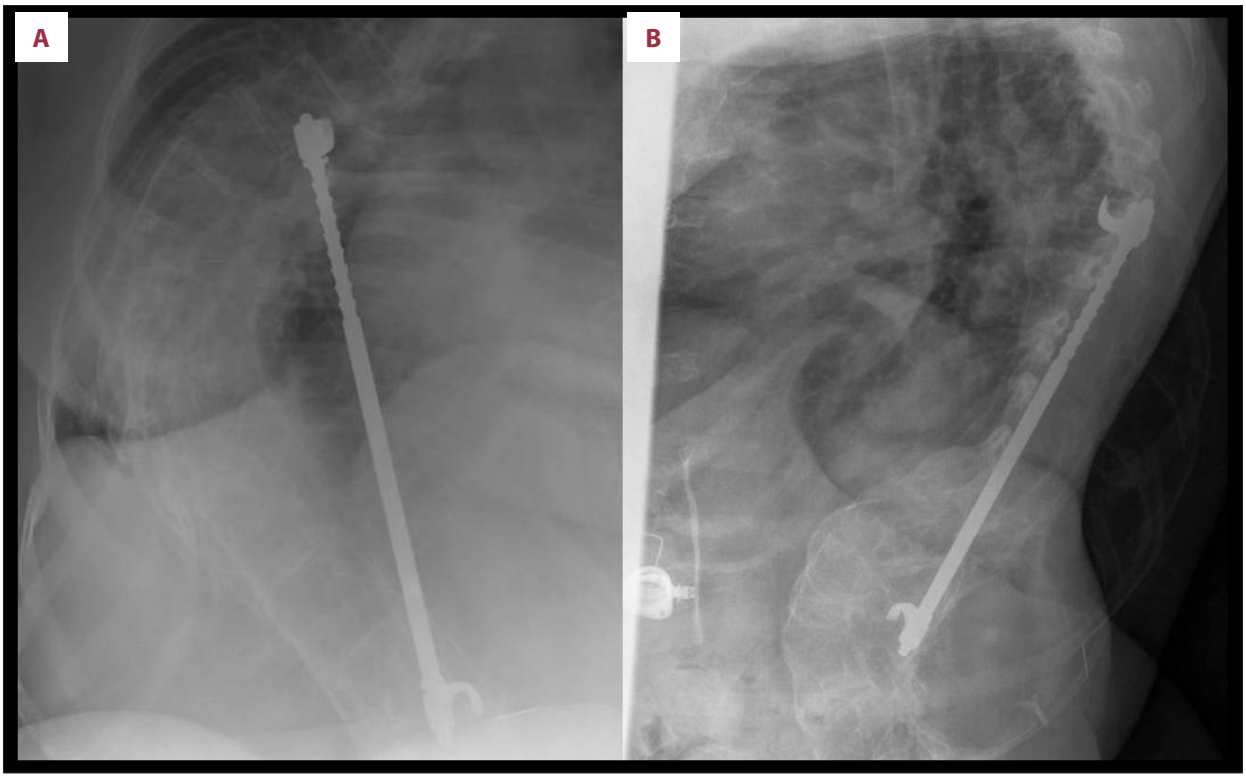

Figure 1. (**A, B**) AP/lateral X-ray of the lumbar spine, showing severe scoliosis and Harrington rod.

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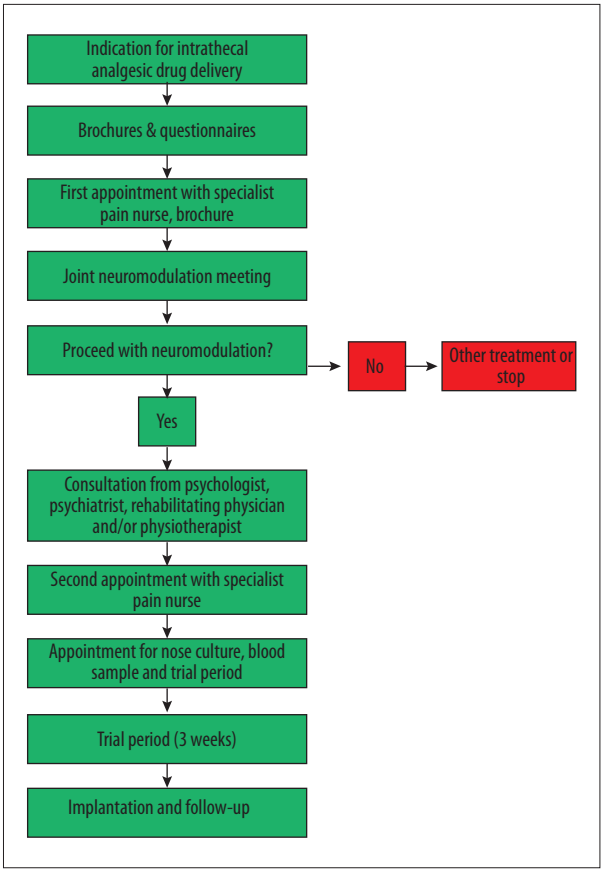

Figure 2. Flowchart for intrathecal drug delivery.

her atonic bladder, the patient received a transurethral catheter, which had to be changed every 6 weeks at home. She went through the ITDD flowchart uneventfully.

After pre-operative screening, an ITDD trial period was initiated, using an Ascenda® spinal catheter (model 8781, Medtronic, Minneapolis, Minnesota, USA), a port-a-cath system (Porthales® 4000M, Medtronic, Minneapolis, Minnesota, USA), and a Crono Five ambulatory infusion pump (Intra Pump® Infusion Systems, Grapevine, Texas, USA). With the patient in the right lateral recumbent position, an intrathecal catheter was inserted (Figure 3); placement of the port-a-cath occurred at the site of pump implantation (left lateral flank), and after that the catheter system was connected to the extension tubing. Despite the spinal deformities, the procedure passed uneventfully. The patient was hospitalized, and an infusion regimen with morphine (Table 1) was initiated, while gradually decreasing oral and transdermal medication. After four days she was discharged and went home, continuing the trial up to three weeks (common practice), in close contact with the pain team. After a successful three-week trial period, the port-a-cath system was replaced by a Synchromed® II programmable infusion pump (model 8637-40, Medtronic, Minneapolis, Minnesota,

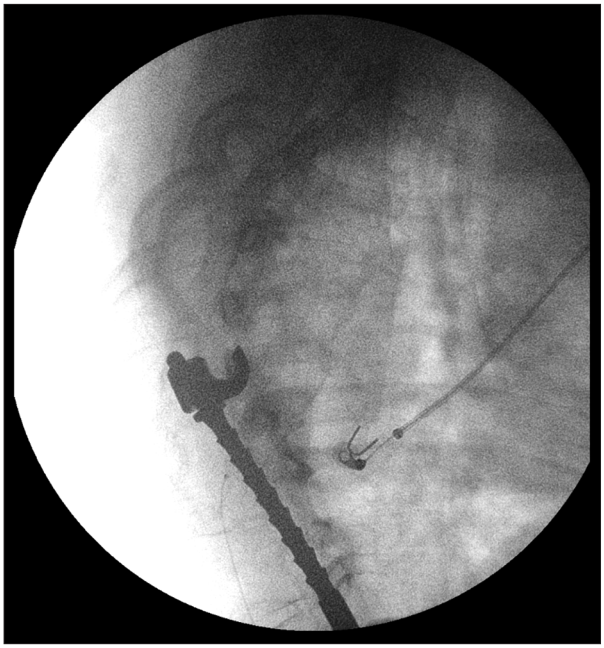

Figure 3. An intrathecal catheter was placed at the thoracic level.

USA), using the same flow and concentration found during the trial period.

During the trial period, verbal NRS for pain decreased from 9/10 to 2/10 with 3.36 mg of spinal morphine (2 mg/mL, 1.68 mL/day). The spinal morphine infusion scheme and verbal NRS for pain after implantation are presented in Table 2.

After 8 months the patient was seen because of skin problems at the pump pocket due to regional pressure resulting from abnormal posture in a wheelchair-bound situation, leading to surrounding erythema at the site of the pump reservoir (Figure 4). We decided to relocate the pump reservoir from the left to the right abdominal wall; this went smoothly and without difficulties.

Eighteen months after the relocation, the patient was seen again seen because of skin problems due to regional postural pressure (letting her right arm rest almost continuously on her belly). Because of skin erosion, the pump reservoir was exposed to open air (Figure 5A, 5B), so we decided on a second relocation, moving towards the left rectus abdominis muscle sheath, a procedure jointly performed with one of our surgeons. This procedure turned out to be a success, and further follow-up was uneventful.

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Table 1. Intrathecal morphine 2 mg/mL titration scheme during the trial period.

Day	Morphine dose (mg/day)	Flow rate (mL/day)	NRS	Fentanyl dose (µg/h)
0	0.48	0.24	9	100
1	0.96	0.48	5	50
2	1.44	0.42	5	0
3	1.92	0.96	5	0
4	1.92	0.96	5	0
7	2.40	1.20	3	0
10	2.88	1.44	4	0
14	3.36	1.68	2	0
17	3.36	1.68	2	0

NRS – verbal Numerical Rating Scale for pain.

Table 2. Intrathecal morphine 2 mg/mL infusion scheme during the follow-up period.

Follow-up	Morphine dose (mg/day)	NRS
Day 1	3.36	2
1 week	3.36	1
1 month	3.69	4
3 months	3.69	2
5 months	4.06	4
6 months	4.06	2
Pump relocation		
1 month	4.06	3
2 months	4.47	2
6 months	4.47	2
10 months	4.97	7
12 months	4.97	4
18 months	4.97	3
Pump relocation		
1 month	4.80	3
2 months	4.80	5

NRS – verbal Numerical Rating Scale for pain.

Discussion

With this case report we would like to draw attention to the complexity of chronic pain management in patients with PPS, as well as the problems that can be associated with this treatment. This case report demonstrates that ITDD can be an option to treat chronic, refractory pain in patients with PPS. Multidisciplinary consultation is necessary to deal with the wide variety of problems in these patients. Skin problems at the site of the pump reservoir can be challenging and time-consuming and, ultimately, can necessitate relocation (or removal) of the device.

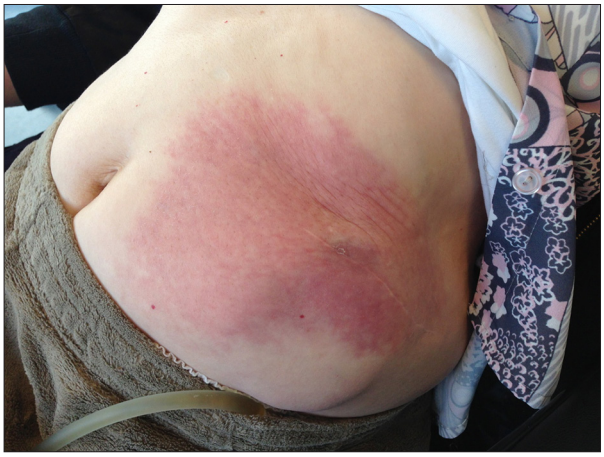

Figure 4. Skin erythema as a result of regional pressure.

Pump pocket complications mainly consist of surgical-site infections, seroma formation, and skin problems such as erythema and erosion [17]. Initial treatment can consist of antibiotic treatment and meticulous wound care, but sometimes more aggressive procedures are needed [18]. Removal of the ITDD system can be considered during the trial period or after implantation when little benefit is being gained at the time these problems occur, or when the infection leads to skin erosion or spreads centrally. Combining the clinical picture with the patient's wishes made us decide to start with antibiotics first, but perhaps immediate removal would have been a better choice. Since an ITDD system represents a lifelong treatment, these discussions are particularly important.

Complications due to regional postural pressure should be considered when implanting an ITDD system in these patients.

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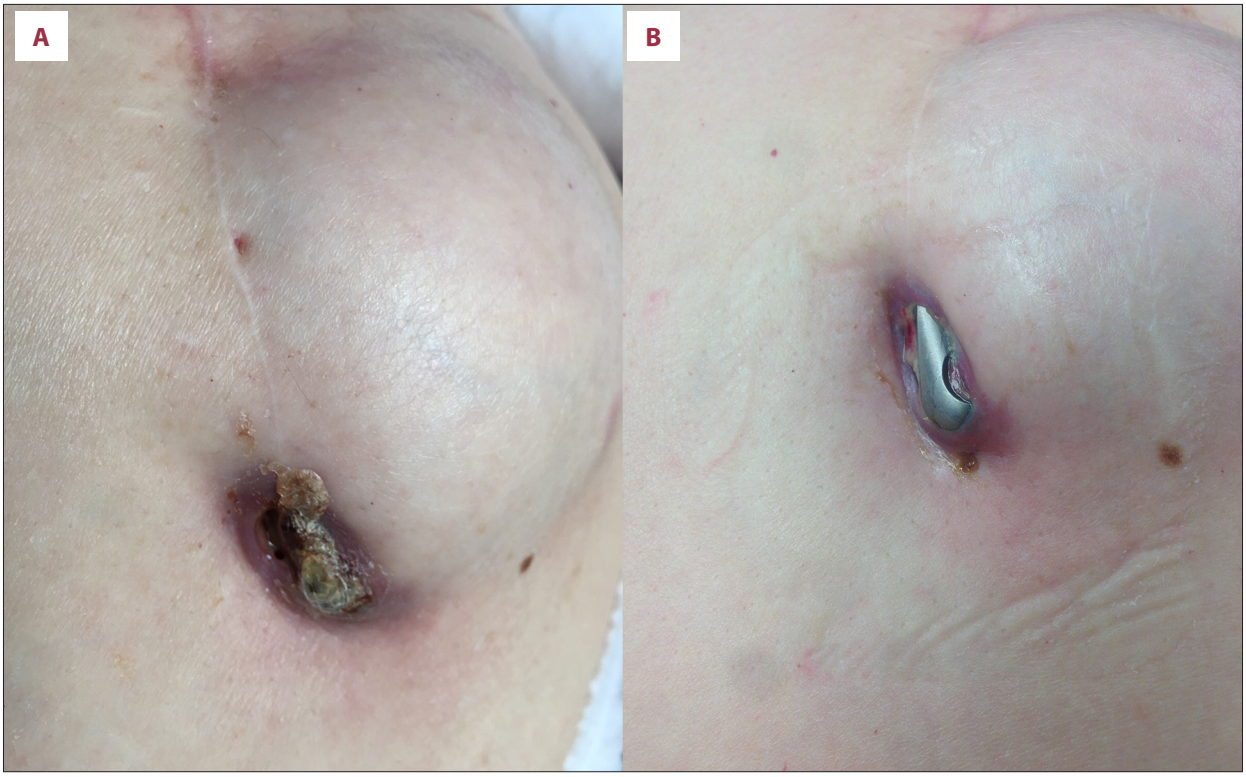

Figure 5. (**A, B**) Skin erosion as a result of regional pressure.

Regular nursing checks, advice on body posture, adaptations to the wheelchair, and putting on a bandage can help decrease the incidence of these complications, but all these methods together did not prevent the occurrence of the situation described in this case report.

Due to its high complexity and limited reimbursement, intrathecal therapy is likely to be underutilized [19]. However, the use of ITDD will grow in the next years, driven by the high incidence of chronic illnesses [20]. Pharmacologic, non-pharmacologic, and hardware-related complications have been described [14] and the physician (and other members of the pain team) should therefore adopt a proactive stance to recognize and consequently treat the complications that may arise. The most common reported side effects of ITDD are constipation (38%), nausea/vomiting (33%), pruritus (26%), sexual dysfunction (26%), and urinary retention (24%) [21]. Frequently reported non-pharmacologic complications are wound infection (12%) and meningitis (2%). Hardware-related complications are equipment revisions (27%), catheter-related problems such as migration, dislodgement, kinking, obstruction, and occlusion (18%), mechanical failure (5%), and pump removal (5%). An uncommon but potentially serious problem is granuloma formation at the catheter tip, for which vigilance in any patient is recommended [22–24].

Given the high percentage of PPS and survivors worldwide, we also need to understand challenges associated with spinal reconstructive surgery. Anterior and posterior correction and fusion of the spine have all been described in detail [25–27]. Spinal reconstructive surgery for poliomyelitis-associated deformity is associated with an overall complication rate of 54%, of which 45% are major complications such as new neurological deficits [28] and the flat back syndrome, which constitutes an inability to stand erect because of forward flexion of the trunk and pain in the low back and/or legs [29]. Since the patient presented in this case report rejected any further spinal surgery, this possibility was not addressed any further.

Conclusions

In patients with PPS, intrathecal analgesic drug delivery can be an option to treat chronic, refractory pain. Multidisciplinary consultation is necessary to deal with the wide variety of problems in these patients. Skin problems at the site of the pump reservoir can be challenging and time-consuming and, ultimately, can necessitate relocation (or removal) of the device.

Acknowledgements

The author wishes to acknowledge the work of Dr. I. M. Cherepanin, surgeon, who was involved in placing the pump reservoir in the rectus sheath, and Dr. J. G. Groeneweg, assistant professor and research coordinator, for proofreading.

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