Case Report

Subdural catheter migration may lead to baclofen pump dysfunction

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Objectives: To report an unusual cause of intrathecal drug delivery failure in baclofen pump device.

Study design: A case report of an SCI patient treated with intrathecal baclofen, presenting a drug withdrawal.

Setting: Regional spinal cord injuries centre in Geneva (Switzerland).

Methods: We present a case of a 38-year-old male with complete T9 spastic paraplegia for 15 years, treated with intrathecal baclofen for 11 years. He recently presented to our centre with a spastic hypertonic episode, associated with rhabdomyolysis.

Results: Standard investigations were unrevealing. However, a CT scan performed after injecting a radio-opaque solution by the side port of the pump, showed an unexpected catheter migration into the subdural space. Surgical revision reversed withdrawal symptoms.

Conclusions: Subdural catheter migration must be considered in the differential diagnosis of intrathecal drug delivery system failures. We recommend the use of the CT scan after contrast injection, to detect the localization of the distal catheter tip and confirm the normal diffusion into the subarachnoid space.

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Introduction

Spasticity, a phenomena characterized by hyperactivity of the muscle stretch reflex, develops in up to 60% of post-traumatic paraplegics.^{1,2} About one in three patients needs oral antispasmodic drugs to counter a loss of independence in their activities of daily living (ADL) or pain secondary to muscle contraction.^{1,2}

Intrathecal baclofen has been commonly used since 1984 in patients who are resistant to or who do not tolerate oral antispasmodic drugs.³ The decision to implant a drug delivery system is usually taken after a positive test (two points diminution in Ashworth or Spasm frequency scale) obtained after a single injection of 50–100 µg of intrathecal baclofen.^{3,4} The effect on the spasticity is spectacular in most cases with a quick diminution of the muscle tonus.^{4–6} Quality of life usually improves due to decrease of nocturnal spasms and a better level of independence.^{6,7} As therapeutic doses are low, the treatment is usually well tolerated. Serious complications are coma and respiratory failure due to depression of the central nervous system. These com-

plications are rare and are seen mostly with programming errors.^{3,4} The most frequent secondary effects are drowsiness, dizziness, blurred vision and slurred speech.³ The fragile component of the drug delivery system is the intrathecal catheter, subjected to the body movements and prone to kink or shear. Catheter dysfunction is seen in about 40% of the patients and typically occurs within 20 months.⁸ Finding the tear can in some cases be extremely difficult due to the minute dimension of the leak in the catheter.^{5,8–10}

We present the case of a patient with symptoms typical of acute baclofen withdrawal and investigations, which allowed us to detect the locus of the system failure.

Case report

The patient fell on his back when he was 24 years and suffered a fracture dislocation of T8–9 and complete T9 paraplegia. The fracture was treated conservatively. After the resolution of the spinal shock, he developed a spasticity of the trunk and lower limbs, severely limiting his independence. At the time of discharge, he was

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treated with oral baclofen 100 mg/day and diazepam 10 mg/day. After 3 years, the oral antispastic treatment was judged to be insufficient and an intrathecal baclofen test was performed with 50 μ g. Following a successful test dose, a drug administration device (DAD) was implanted (SynchroMed EL-18, MEDTRONICS®, MN, USA) in year 1991. During the first months, spasticity was controlled with an average daily dose of 62 μ g baclofen per day.

Since then, the patient presented multiple drug delivery failures, the first one only 5 months after the implantation. Three times, a leak in the catheter was discovered, once caused by a 'bungee' jumping. After 6 years, the battery of the pump had exhausted and the pump needed to be replaced. On three occasions, the patient suffered spastic hypertonic crisis despite an apparently functional DAD. Those episodes were related to bladder microlithiasis, muscle haematoma, intramuscular needle in the right buttock.

The present hospitalisation was motivated by a hypertonic crisis which developed a few hours after sexual intercourse. His spasticity increased progressively and evolved into uninterrupted tetanic spasms of the abdominal wall and lower limbs, despite 300 μ g per day of intrathecal baclofen. Transfers and sitting into the wheelchair became impossible because of the spontaneous contraction of the extensors and his sleep was perturbed. Upon hospitalisation, the patient was found exhausted, suffering from severe spasms (modified Ashworth and spasm frequency scale at 4/4). Exhaustion increasingly gave place to uncontrolled anger. Symptoms did not respond to oral diazepam (10 mg/day) and baclofen (100 mg/day).

We found no cause for increased nociceptive stimulus. A systematic analysis of the DAD showed no programming abnormality. Two bolus of 100 and 200 μ g of baclofen were given with no clinical effect and radiological evaluation showed no rotor movement failure.

An evaluation of the catheter with 4 ml of radio-opaque solution injected in the side port showed no evidence of leakage. However, we had difficulties in seeing the diffusion of the solution at the tip of the catheter.

Oral dantrolene (2 $\times$ 50 mg/day) was given in order to prevent worsening of the rhabdomyolysis. The effect of the dantrolene was beneficial (modified Ashworth score 2/4) but inconstant and hence, a surgical revision was carried out. During the operation, the intra- and extrathecal catheter were disconnected, showing normal backflow of the CSF on one side, and normal flow of baclofen after a bolus on the other. It was decided not to change the catheter and both sides were reconnected. The day after the operation hypertonia relapsed, despite an increase of intrathecal baclofen to 430 μ g/day, a second radiological evaluation of the catheter was performed with 8 ml of solution, this time showing clearly the leak of the radio-opaque product at the level of the catheter's tip in the subdural space (Figure 1). The CT scan performed immediately after the injection demonstrated that the distal catheter was dislodged

Figure 1 Standard X-ray with injection of contrast product by the side port; leak in the subdural space (arrow).

between the subdural and subarachnoid space. This finding explained the insufficient perfusion of the baclofen (Figure 2). The intrathecal catheter thus was surgically replaced, following a complete resolution of symptoms after 24 h.

Discussion

The efficacy of intrathecal baclofen treatment lies in its ability to bind to superficial GABA-B receptors of the inhibitory interneuron in the posterior horn, and thus to decrease muscle tone.¹¹ In cases of drug delivery failure patients present with a severe rebound of their spasticity, which can lead to rhabdomyolysis and consequently renal failure. Our patient had been already treated three times for a rhabdomyolysis since the implantation of his DAD 11 years ago. Reeves *et al*¹² described these types of complications in a tetraplegic suffering from acute catheter failure. The patient needed to be aggressively treated in the emergency department for rhabdomyolysis, hyperthermia, followed by disseminated intravascular coagulation and cardio-respiratory failure. Symptoms may resemble to the Neuroleptic Malignant Syndrome (NMS).¹² Other symptoms described with withdrawal are seizures, hallucinations and psychosis.¹³ During the period of uncontrolled spasticity, our patient presented repeated episodes of rage without hallucinations, which may have reflected his despair, possibly augmented by baclofen withdrawal.

Figure 2 CT scan showing leak in the subdural space (arrow) and catheter tip (double arrow).

The treatment of spasticity in the case of baclofen withdrawal can be very difficult, both for the patient and the clinician. Patients often do not respond to oral therapy. In our patient, oral baclofen and diazepam induced side effects without reducing the spasms. Oral dantrolene (100 mg/day) partially controlled the spasticity. According to Reeves *et al*,¹² the use of dantrolene i.v. could be a temporary solution in case of severe withdrawal, once the site of dysfunction is detected.

Investigations should be performed in a stepwise manner, first eliminating causes that might increase nociceptive stimuli, then the presence of a drug delivery system failure and finally the possibility of drug tolerance.^{9,14}

After the clinical examination, we suggest to check the programming of the pump (dosage, reservoir volume, flow rate, battery status) and perform standard X-ray to look for kinking or disconnection of the catheter. At this point when nothing wrong is detected, we propose to perform progressive boluses with the pump (100, 200 μ g). According to Penn³ and Le Breton *et al*,¹⁰ the next step could be to perform a bolus of 100 μ g by lumbar puncture to exclude the presence of drug tolerance. The evaluation of the integrity of the catheter is difficult and controversial. Filling the pump with the radiological contrast solution and making a bolus is in many cases not sensitive enough to detect catheter tip anatomy.^{9,10} From our experience the use of the side port, injecting 8 ml of radio-opaque solution is more reliable (Figure 1) to follow the intrathecal catheter and the diffusion of the contrast in the subarachnoid space. The use of the CT scan immediately after the injection allowed us to detect the exact place of the tip of the catheter, localised in part in the subdural space (Figure 2). Dislodgement of the catheter is not rare, and occurs after repetitive pulling and shearing movements on the catheter when the patients move his trunk.⁸

We obtained a backflow of CSF during the surgical revision in our patient which in our opinion is not sensitive enough to exclude a small hole on the length of the intrathecal catheter. In case of doubt, we recommend to replace the intrathecal catheter.

Conclusion

In case of baclofen withdrawal, a step-by-step checking method should help to make the difference between drug tolerance and a drug delivery device failure. The most vulnerable part of the system is the intrathecal catheter submitted to the shear forces of the body during trunk movement. Subdural catheter migration must be considered in the differential diagnosis of intrathecal drug delivery system failures. We recommend the use of the CT scan with contrast material injection in the side port, in order to detect precisely the localization of the distal catheter tip.

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