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CASE REPORT

Neuraxial analgesia in a pregnant woman with Fowler's syndrome and sacral neuromodulation

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ABSTRACT
We report the anesthetic management of a 16-year-old woman with Fowler's syndrome who became pregnant three years after sacral neuromodulation was initiated for treatment of the condition. Multidisciplinary consensus was to switch off the neurostimulator during pregnancy, and attempt vaginal delivery with a neuraxial block. When the patient was admitted for labor, an epidural catheter was placed successfully. The patient had a normal vaginal delivery. Sacral neuromodulation was restarted uneventfully in the early puerperium and the Fowler's syndrome remains well controlled. The baby continues to develop normally three years after delivery.

Keywords: Fowler's syndrome; Sacral neuromodulation; Neuraxial anesthesia

Introduction

Fowler's syndrome, first described in 1988, is a cause of urinary retention in 20 to 30-year-old-women.[1] It is characterized by a failure of the urethral sphincter to relax, which limits the passage of urine. Urodynamic studies show high post-void residual volumes, an increased resting urethral pressure and abnormal urethral sphincter electromyography without neurological or structural disorder. An association between Fowler's syndrome and polycystic ovarian syndrome and hypothyroidism has been described.[1,2] The most effective treatment for urinary retention in Fowler's syndrome is unilateral sacral neuromodulation at S3 level.[2]

Neuromodulation is the alteration of neural activity through delivery of electrical stimulation or chemical agents. The mechanism of action of spinal cord stimulation (SCS) is based on the gate control theory, in which activation of afferent fibers in the spinal cord inhibits ascending nerve traffic at the segmental level. The pathway by which SCS improves nerve function is an area of active research.[3] Neuromodulation is widely used for chronic pain disorders when pharmacological approaches have proved inadequate. Fowler's syndrome, since it predominantly affects women of childbearing age, may be of consequence for anesthesiologists and obstetricians.[1] Controversy exists about whether to keep the stimulator switched on during pregnancy and labor, and will continue until more data about the effects of the electrical current and electromagnetic waves on the fetus become available.[4,5]

Information about the anesthetic and obstetric management of pregnant women with SCS systems is scarce although their use is becoming more frequent.[6] We report the anesthetic management of a pregnant woman with an SCS for Fowler's syndrome.

Case report

A 16-year-old patient with Fowler's syndrome presented at nine weeks of gestation. She had hypothyroidism and vesicoureteral reflux surgically treated in infancy. Before the definitive diagnosis of Fowler's syndrome, she had been treated repeatedly with self-catheterization and bilateral ureteroneocystostomy with no improvement. Fowler's syndrome had been diagnosed three years previously and the patient had been successfully managed with sacral neuromodulation. A sacral SCS (InterStim Medtronic®) was implanted for urethral sphincter stimulation. The implantable pulse generator consisted of a battery attached to a stimulating electrode placed

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through the S3 sacral foramen, to deliver an electric current to the sacral nerve roots. A unilateral tetrapolar electrode was placed in the S3 left root, and the battery was implanted in the left buttock (Fig. 1). After placement, voiding function was restored with no post-void residual urine and there was no urinary tract dilation as demonstrated by ultrasonography.

In the tenth week of pregnancy, a multidisciplinary meeting of urologists, obstetricians and obstetric anesthesiologists was held. At that time, FDA and manufacturer's advice was to switch off the SCS once pregnancy was diagnosed.[4,5,7] Based on these recommendations, this was done and vaginal delivery was planned. After checking battery and electrode location, it was considered there was no contraindication for neuraxial analgesia in labor. When the sacral stimulator was switched off, the patient developed urinary retention and required daily catheterization. She also suffered recurrent urinary tract infections that required hospital admission on four occasions. All were treated with ampicillin and gentamicin, and antibiotic prophylaxis with weekly fosfomycin was started.

When the patient was admitted for labor, an L3–4 epidural catheter was placed under strict asepsis (surgical scrub, cap, mask, gown and sterile gloves) using an 18 gauge Tuohy needle. After a negative test dose with 3 mL 0.25% bupivacaine with 1:200 000 epinephrine, a bolus dose of 10 mL of 0.125% bupivacaine plus fentanyl 75 μg was administered and a continuous infusion of 0.0625% bupivacaine plus fentanyl 2 μg/mL was started at 12 mL/h. This gave good pain relief, but after five hours the patient started to complain of abdominal pain. Three 5-mL boluses of 0.125% levobupivacaine were administered with no improvement. A combined spinal-epidural technique was subsequently performed at L4–5 with a 18-gauge Tuohy and 27-gauge Whitacre needle. Intrathecal bupivacaine 2.5 mg with fentanyl 20 μg were administered and the continuous epidural infusion was restarted at 12 mL/h with good effect. Four hours later vaginal examination showed complete cervical dilation and the patient delivered without complications. Manual extraction of a retained placenta under sedation with propofol was required, and uterine atony responded to uterine massage and oxytocin infusion. Estimated blood loss was 1000 mL. This caused dizziness and hypotension which responded to 1 L of fluid therapy and intravenous ephedrine 10 mg.

After a period in the post-anesthesia care unit, the patient remained hemodynamically stable. Blood results showed a hemoglobin value of 7.2 g/dL with an international normalized ratio of 1.05 and an activated partial thromboplastin ratio of 1.3. There was no active bleeding and the patient was transferred to the ward. The day after delivery the SCS was restarted successfully. She recovered good void control with weekly catheterizations to control residual post-voiding volume. The baby developed normally.

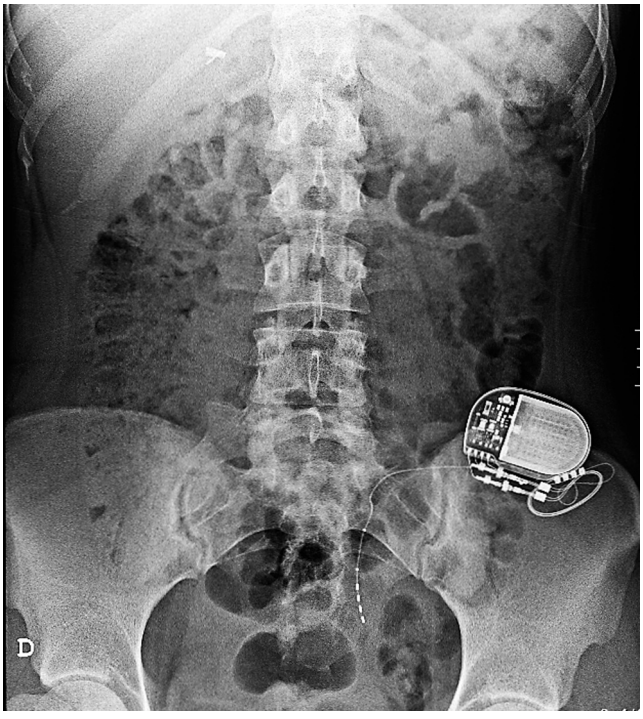

Fig. 1 Battery attached to stimulating electrode placed through sacral foramen S3

Discussion

There are few published cases reports of neuraxial anesthesia or analgesia in pregnant patients with Fowler's syndrome and sacral neuromodulation.[8,9] Epidural usage in labor is increasing, and more patients with uncommon diseases who become pregnant may request neuraxial analgesia.[8,10] The obstetric and anesthetic implications of these conditions may remain obscure.

There is a potential risk of electrode displacement during pregnancy due to changes in lumbar spinal curvature and the pressure exerted by the gravid uterus, and some consideration must be given to the choice of analgesic and anesthetic technique. This should include the entry point and path of the electrodes to the epidural space, the location of the impulse generator and its intermediate connections to avoid displacement or damage with the epidural needle or catheter. Furthermore, the potentially increased risk of infection in patients with foreign elements in the epidural space should be considered.[11]

The preferred method of delivery is unclear, but should be chosen in advance.[4,5] During vaginal delivery it is possible that the device may be displaced due to patient positioning and the effort of pushing. In contrast, devices placed in the abdomen may be damaged if cesarean delivery is performed.[4]

There are a number of case reports of pregnant patients with chronic pain who have cervical, thoracic,

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