Management of Pregnancy and Childbirth in a Cervical Dystonia Patient with an Implanted Deep Brain Stimulation System: A Case Report

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Abstract

Deep brain stimulation (DBS) can lead to psychosocial and functional improvement in medically refractory cervical, segmental, or generalized moderate to severe dystonia. After treatment with DBS in women with dystonia, pregnancy can be planned. However, in the literature, there are no standardized clinical guidelines for the management of movement disorder treated with DBS during pregnancy. Herein, we report a 24-year-old female patient with cervical dystonia (CD) who have an implanted bilateral globus pallidus interna (GPI)-DBS. The patient got pregnant during the 5-year follow-up period after DBS surgery and then delivered a healthy baby via cesarean section under general anesthesia. A patient with CD who have a DBS system with a rechargeable battery could be managed safely during pregnancy and childbirth.

Keywords: Deep brain stimulation, delivery, dystonia, globus pallidus interna, pregnancy

What is known
Deep brain stimulation (DBS) can lead to psychosocial and functional improvement in medically refractory cervical, segmental, or generalized moderate to severe dystonia. After treatment with DBS in women with dystonia, pregnancy can be planned. However, in the literature, there are no standardized clinical guidelines for the management of movement disorders treated with DBS during pregnancy.

What is new
Herein, we report a 24-year-old female patient with cervical dystonia (CD) who have an implanted bilateral globus pallidus interna (GPI)-DBS. The patient got pregnant during the 5-year follow-up period after DBS surgery and then delivered a healthy baby via cesarean section under general anesthesia. A patient with CD who have a DBS system with a rechargeable battery could be managed safely during pregnancy and childbirth.

Introduction
Dystonia is a movement disorder characterized by intermittent or constant involuntary muscle contractions. Cervical dystonia (CD) is the most common type of focal dystonia and is commonly characterized by abnormal movement and posture in the neck and head.[1] This lifelong disease can significantly affect an individual’s psychological and physiological function and activities of daily living. Botulinum neurotoxin (BoNT) and deep brain stimulation (DBS) have significant clinical benefits in eligible patients.[2] After DBS, individuals with CD can gain their independence and can easily establish a social relationship.[3] In this study, we presented the management of a CD patient with a history of DBS with a rechargeable battery during pregnancy and childbirth.

Case Description
A 24-year-old woman, with a 3-year history of CD, was admitted to our institution. The patient’s symptoms included involuntary twisting or turning of the neck, which causes an abnormal head position, and they were induced by stress. The patient was found to have sporadic CD. Her Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) score was 46 (pain, 7; disability, 22; and severity, 17). The predominant CD pattern was retrocollis. After obtaining a multidisciplinary council decision, DBS was recommended.

Surgery
The patient underwent bilateral Gpi-DBS surgery in our institution. The electrodes were implanted stereotactically into the posteroverentralateral globus pallidus internus that was...
Table 1: Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) during follow-up

|               | Pre | 6 months | 12 months | 36 months | 48 months | 60 months |
|---------------|-----|----------|-----------|-----------|-----------|-----------|
| TWSTRS-Pain   | 7   | 3        | 3         | 1         | 1         | 1         |
| TWSTRS-Disability | 22  | 9        | 3         | 0         | 0         | 0         |
| TWSTRS-Severity | 17  | 5        | 2         | 1         | 0         | 0         |
| TWSTRS-Total  | 46  | 17       | 8         | 2         | 1         | 1         |

Table 2: Parameters of deep brain stimulation

|               | Voltage | Pulse width | Frequency | Electrodes |
|---------------|---------|-------------|-----------|------------|
|               |         | µsec        |           |            |
| Left          |         |             |           | Monopolar mode |
| After surgery | 4.1 V   | 60 µsec     | 130 Hz    | 1          |
| 6 months      | 4.3 V   | 60 µsec     | 130 Hz    | 1          |
| 12 months     | 4.6 V   | 60 µsec     | 130 Hz    | 1          |
| 36 months     | 4.6 V   | 60 µsec     | 130 Hz    | 1          |
| 48 months     | 4.6 V   | 60 µsec     | 130 Hz    | 1          |
| 60 months     | 4.6 V   | 60 µsec     | 130 Hz    | 1          |
| Right         |         |             |           | Monopolar mode |
| After surgery | 2 V     | 60 µsec     | 130 Hz    | 1          |
| 6 months      | 2.2 V   | 60 µsec     | 130 Hz    | 1          |
| 12 months     | 2.5 V   | 60 µsec     | 130 Hz    | 1          |
| 36 months     | 2.5 V   | 60 µsec     | 130 Hz    | 1          |
| 48 months     | 2.5 V   | 60 µsec     | 130 Hz    | 1          |
| 60 months     | 2.5 V   | 60 µsec     | 130 Hz    | 1          |

cross correlated with the Schaltenbrand and Wahren brain atlas, magnetic resonance imaging, and microelectrode mapping. After the DBS procedure, we determined the most effective combination of contacts for the electrodes at a frequency of 130 Hz, amplitude of 2.5 V, and pulse width of 60 µs on the right side and a frequency of 130 Hz, amplitude of 4.6 V, and pulse wide of 60 µs on the left side.

Postoperative control

After GPi stimulation, almost all symptoms were reduced within 6 months, and the patient did not require medication for the treatment of dystonia. Her TWSTRS score was 1 (pain, 1; disability, 0; and severity, 0) [Table 1]. The pulse generator battery was no longer working after 4 years of implantation. Hence, the patient decided to use a rechargeable battery system (Boston Scientific Vercise). After implantation, no complications were observed. Six months after battery replacement, the patient got pregnant.

Pregnancy period

During pregnancy, the patient and the baby did not experience problems associated with the rechargeable battery system. The battery was charged for half an hour per week during pregnancy. There was no clinical deterioration requiring a change in programming during follow-up [Table 2]. At the 36th week of pregnancy, she gave birth to a healthy baby via cesarean section under general anesthesia. The DBS system was closed during the surgery, and there were no problems with the battery during breastfeeding.

Discussion

CD is the most common type of focal dystonia that can significantly reduce quality of life and can cause a person to isolate from the social environment. Previous studies have shown that CD is common in young women. Whether pregnancy is safe for CD individuals with an implanted DBS system who live independently after DBS has not been validated. In the current case, the patient presented with CD, which was primarily characterized by retrocollis. However, she maintained her social life and established a family after bilateral Gpi-DBS implantation. In a few published articles about the management of pregnancy in patients who underwent DBS, some have experienced anxiety caused by the use of battery. By contrast, some had a problem-free pregnancy. None of the studies provided details about the charging processes during pregnancy. There was no change in the baby’s movements or heart rate during charging periods in our patient. We know that the use of shortwave diathermy, microwave diathermy, or therapeutic ultrasound diathermy (now all referred to as diathermy) in patients with a neurostimulation system implanted is not recommended by manufacturers. But, there is no contraindication for diagnostic ultrasound and there is not enough information about fetal monitoring safety during pregnancy in a DBS implanted patient. There was no problem during fetal monitoring during pregnancy follow-ups in our case. In a previous study, there were no significant changes in dystonia symptoms during pregnancy, and there is no consistent correlation between hormonal changes during pregnancy and severity of dystonia.

However, in few cases, dystonia worsened during pregnancy. Ziman et al. reported that only three of seven patients experienced worsening of dystonia symptoms during pregnancy, and these symptoms resolved within 3 months after birth. Two of these patients were diagnosed with isolated segmental dystonia, while the third was diagnosed with isolated hemidystonia. Meanwhile, in the study of Paluzzi et al., none of the pregnant patients (n = 3) experienced worsening of symptoms. In this case, we performed a detailed examination using a questionnaire during each trimester transition. Results showed that the symptoms did not worsen during pregnancy, and there was no problem with the implanted DBS system during pregnancy. Moreover, no abnormality was observed during fetal monitoring before or after surgery.

A comprehensive study showed that individuals with a DBS device were not affected by the mode of delivery. In a previous study on seven pregnant women, four gave birth via vaginal...
delivery and three via cesarean section. Six of these mothers delivered healthy babies, and there were no complications during the delivery process. However, due to premature rupture of membranes, one gave birth prematurely. Hence, the baby had low birth weight.[6] In light of the current literature, pregnant women with implanted DBS systems should deliver their babies at the hospital, not at home. In our case, cesarean section under general anesthesia was planned based on the request of the patient because she is afraid to give birth via normal delivery. Monopolar electrosurgery is strictly prohibited during any surgery and only bipolar use is allowed in DBS systems.[8‑10] However, it must be kept at least 15 cm away from the battery, extension cables, and electrodes.[8] In our case, the obstetrician did not use a monopolar electrocautery device due to possible risks. Data about women with implanted DBS systems and the use of antibiotic prophylaxis during childbirth are limited.[11] However, due to the fact that the DBS system is a foreign body, the patient received intravenous cefuroxime (1.5 g) immediately after the baby was delivered, and another two doses thereafter. Previous studies comprise case series in which the DBS system was turned off during delivery.[5‑6] In patients with a DBS system, complete closure of stimulation is recommended during any surgical procedure.[8‑10] In our case, the stimulation was shut down just before delivery and reactivated after delivery.

Conclusions
A CD patient with an implanted DBS system with a rechargeable battery could be managed safely during pregnancy and childbirth.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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