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CASE REPORT

A Case Report of Successful Vaginal Delivery for Cauda Equina Syndrome Combined with Complex Regional Pain Syndrome

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Background: Cauda equina syndrome (CES) is defined as a complex clinical entity associated with simultaneous compression of several or all lumbosacral spine nerve roots. Complex regional pain syndrome (CRPS) is a chronic and non-treatable pain condition resulting in lifetime disability due to a tendency to relapse. Both diseases present with chronic pain and neuropathy. We aimed to report a unique patient with successful pregnancy and delivery although she had both CES and CRPS.

Case Presentation: Here we present a female who was diagnosed as CES with CRPS after fall from a high building about 10 years previously. She became pregnant after two miscarriages, maintained her pregnancy with numerous pain killers and finally delivered a healthy baby vaginally. Interestingly, during her parturition, she was tolerable with labor pain unlike her chronic pain that came from CES and CRPS.

Conclusion: She is a unique and significant case because of the rare condition that has two pain-associated diseased and success of vaginal delivery without any complication during pregnancy. This case may be helpful in counselling attending physicians and may inspire other pre-existing CES or CRPS patients who are afraid to decide on pregnancy.

Keywords: cauda equina syndrome, complex regional pain syndrome, delivery, neuropathy, parturition, pregnancy, CES, CRPS

Introduction

Cauda equina syndrome (CES) and complex regional pain syndrome (CRPS) are both very rare disease entities that affect the quality of life. CES is diagnosed when the lower end of the spinal cord, known as cauda equina within the lumbar spinal canal, is compressed or damaged.[1] The clinical manifestations vary according to the degree of damage, nevertheless, common presentations include motor weakness of the legs and lifelong perineal sensory abnormalities along with bowel, bladder, and sexual dysfunction.[2] Tumors or injuries resulting in fractures in this specific location of the spine can CES. In contrast, the cause of CRPS is not usually specified, although injury, trauma or rheumatoid arthritis are known risk factors.[3] It is a complex disorder with diverse presentations of chronic pain without predisposing circumstances and sometimes leading to psychological problems such as depression.[4] Females are known to be a risk factor for CRPS, thus patients in reproductive age have to consider the possibility of pregnancy.[5] There is little literature about CRPS patients who experienced pregnancy and delivery, however medications to control pain and pain itself are factors that make a pregnancy complicated. There was a case report of a successful pregnancy after spinal cord stimulation and a healthy baby was delivered via cesarean section.[6] Kato et al, reported a case of relapsed CRPS after caesarean section.[7]

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Here we present a case of a pregnant woman with alleged CES combined with CRPS who struggled against the pain during her whole pregnancy and finally ended up with successful vaginal delivery. The Institutional Review Board of Seoul National University Bundang Hospital (B-2403-887-701) has approved to proceed the study protocol and to publish the case details on 15th February, 2024. Written informed consent form to have the case details and accompanying images was obtained from the patient.

Case Presentation

A 31-year-old nulliparous pregnant woman with 6 weeks of gestation was admitted for pain control. Her pain was unpredictable, unbearable, and not localized in certain body parts. She had been diagnosed with CES combined with CRPS after falling from a high building due to a suicide attempt in January 2013. Multiple fractures at lumbar to sacral spine bones including the coccyx were found; thus she had undergone 2-step orthopedic surgeries (step 1: lumbar myelopathy and posterior instrumentation in dorsal and dorsolumbar spine; step 2: posterior fusion in lumbar sacral spine) (Figure 1). Her urodynamic study revealed severe bladder dysfunction with acontractile detrusor, therefore, clean intermittent catheterization was unavoidable for the rest of her life. After about one year of physical therapy and rehabilitation exercise, she could walk by herself with the assistance of crutches. Because CRPS developed after the accident, she had to continue to take strong painkillers (pregabalin, tramadol with acetaminophen, and fentanyl patch) for everyday life. Her thermography with digital infrared showed the definite difference in temperature between the left leg and right leg (Figure 2).

She twice experienced miscarriage and finally became pregnant in 2023. Multidisciplinary team approach including doctors in anesthesiology, rehabilitation medicine, neuropsychiatry, obstetrics, and neonatology was performed for the whole pregnancy period. Chronic pain and allodynia (a type of neuropathic pain that causes extreme sensitivity to touch, even from brushing hair or wearing a soft shirt) in both lower legs combined with paresthesia, temperature change, sweat change, and atrophy in the left leg were her symptoms. Transcutaneous Electrical Nerve Stimulation (TENS) to relieve her pain was applied several times and hospitalization was frequently decided for intensive pain control. Table 1 demonstrates her medication history during pregnancy. She repeated admission and discharge because of the pain, however all her regular prenatal check-ups (almost 10 visits before delivery) including laboratory tests, fetal ultrasound, fetal aneuploidy screening

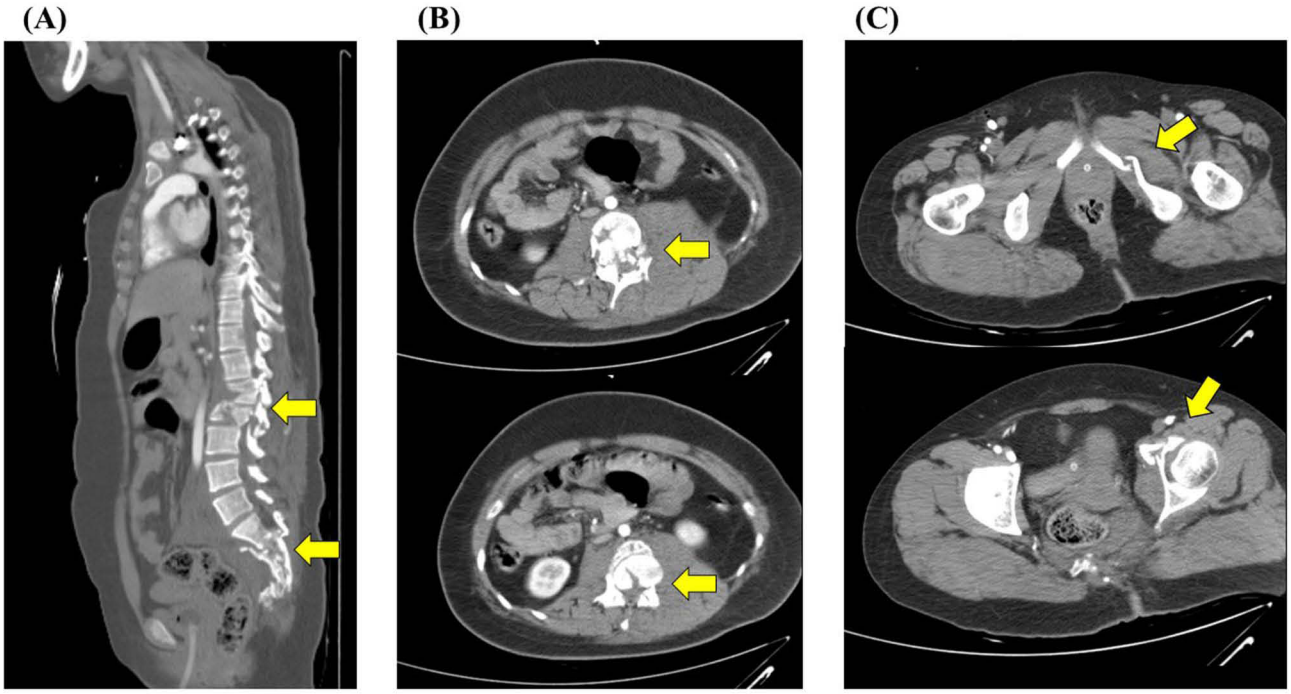

Figure 1 Computed tomography images taken immediately after the fall in January 20th, 2013 (Yellow arrows indicate fractures of spine). (**A**) Sagittal plane image showing multiple comminuted fracture at lumbar-sacral spine, coccyx, left anterior acetabulum and left inferior pubic ramus; (**B**) transverse plane image showing burst fractures with traumatic retrolisthesis in level L1–L2 and multi-level involvement of sacral neural foremen; (**C**) severe central canal and neural foraminal compromise at L1, L2 level.

Figure 2 Digital infrared thermography image showing definite difference in temperature between left leg and right leg, implying complex regional pain syndrome.

test, and gestational diabetes screening test were normal. Even light touch caused by fetal movement and enlarging uterus due to the progression of pregnancy meant uncontrollable pain. The attending obstetrician reassured her that preterm birth could be considered if she could not stand the pain and neonatologists were aware of her status.

Table 1 Medications for the Patient's Pain Control Taken During the Current Pregnancy

Gestational Age	Medication	Remark
–6+3 weeks	Lyrica 150mg (Pregabalin) 1cap 2 times/day PO Ultracet 37.5mg/325mg (Tramadol/AAP) 1 tablet 3 times/day PO	Admitted for pain control at GA 6+3wks
6+4–7+3 weeks	Tylenol ER 650mg (AAP) 1 tablet 3 times/day PO Prn) Pethidine 25mg (Demerol) IV Prn) AAP premix 1g/100mg IV TENS daily	Discharged at GA 7+3wks
7+4–4+6 weeks	Tylenol ER 650mg 1 tablet 3 times/day PO AAP premix 1g/100mg IV per week TENS per week	
15+0wks–20+6wks	Ultracet ER 75mg/650mg 1 tablet 2 times/day PO TENS per month	
21+0wks–29+2wks	Ultracet ER 75mg/650mg 1 tablet 2 times/day PO Ultracet 37.5mg/325m 0.5 tablet 2 times/day PO TENS per month	Admitted for pain control at GA 25+6wks
29+3wks–31+1wks	Ultracet ER 75mg/650mg 1 tablet 3 times/day PO Ultracet 37.5mg/325mg 0.5 tablet 2 times/day PO Prn) Fentanyl 12mcg/h patch per 3 days TENS twice per week	Discharged at GA 31+1wks

(*Continued*)

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Table 1 (Continued).

Gestational Age	Medication	Remark
31+2wks–36+1wks	Ultracet ER 75mg/650mg 1 tablet 2 times/day PO Ultracet 37.5mg/325mg 0.5 tablet 2 times/day PO Prn) Fentanyl 12mcg/h patch per 3 days TENS per week	Admitted for induction of labor at GA 36+1wks

Abbreviations: PO, per os (by mouth); GA, gestational age; AAP, acetaminophen; ER, extended release; prn, pro re nata (as necessary); IV, intravenous; TENS, transcutaneous electrical nerve stimulation.

In May 2023, she was admitted in the delivery unit for induction of labor in the 36th week of gestational age. The delivery mode was decided after thorough counseling with her and her husband. After administration of vaginal suppository prostaglandin, active labor pain accompanied by cervical change (dilatation and effacement) developed, however she could not feel the pain unlike other pregnant women. Epidural anesthesia was not able to be applied due to her past spine fractures and operation history, nevertheless she did not ask for other pain controls during labor. As the cervix dilated fully (10cm) and the fetal head was engaged to her pelvis, she complained of low abdominal pain, but said it was tolerable. Since she could feel the uterine contraction (every three to five minutes), with a much lower degree of labor pain compared to other pregnant women, pushing by herself was possible. With right side medio-lateral episiotomy, a healthy female infant weighing 2.34 kg was delivered vaginally without any complications such as postpartum hemorrhage. No forceps or vacuum were used for the assistance of delivery. Apgar scores of the newborn were 8 and 10 after 1 minute and 5 minutes, respectively. On the next day, her postpartum vital signs were stable with hemoglobin level 9.5 g/dl (previous level; 9.8 g/dl) and the patient's general condition was very tolerable. She was discharged on postpartum day 2.

Discussion

A number of CRPS occur in middle-aged adults with a female predominance, and thus most of the affected women are likely in reproductive age.[8] Because this complex pain syndrome does not usually have a certain etiology, treatment is not targeting the fundamental pathophysiology, but remains in conservative management. Pain control for CRPS was cautiously monitored due to the possibility of side effects and overdosage although in pregnancy, the effect to the fetus has to be considered. In this case, during her pregnancy period, various painkillers were administered much more frequently than other general pregnant women, although none of them was known to be teratogenic. Food and drug administration established five categories (A, B, C, D, and X) to indicate the potential effect of a medication to fetus, nevertheless most of data are not based on definite evidence due to the ethical limitation of experiments or clinical trials to pregnant women. The newborn of this case was healthy at the routine neonatal examinations. Before birth, neonatologists concerned with the possibility of respiratory morbidity because opioids might depress neonatal respiration in a dose-dependent manner.[9]

We could assume the pain from CRPS and CES had been appalling in her whole life that she the degree of labor pain during parturition was tolerable. Therefore, during labor, she did not receive any pain management including epidural anesthesia. The standard treatment protocol for CRPS in non-pregnant people is individualized approach for pain control.[10] Medications such as nonsteroidal anti-inflammatory drugs, opioids and anticonvulsants are primarily used and antidepressants can help reduce pain and sleep disturbance. Corticosteroids are sometimes used to reduce inflammation and physical therapies such as movement therapy or gentle weight-bearing exercises improve circulation and prevent stiffness of muscles. Nerve blocks involve injection of local anesthetics can provide temporary pain relief and cognitive behavioral therapy may be used to develop coping mechanisms. Spinal cord stimulator or biofeedback could be an optimal choice of treatment for pregnant women with CRPS since several case reports showed safety during pregnancy.[11,12] In this case, she received TENS many times especially in the early and later gestational period. In early pregnancy, organogenesis of the fetus has to be considered, so TENS helped reduce the frequency and dosage of medications. In addition, TENS was applied to subside the pain caused by irregular uterine contractions that normally happen during later gestation due to the enlarged uterus. Application of TENS reduced the use of pain control medications.

Table 2 Literature Reviews About Pregnancy and CRPS

Author (Year)	Age	Cause of CRPS (Timing of Diagnosis)	Pregnancy Outcome	Discussion
Segal (1999)[12]	31	Motor vehicle accident (2 years before pregnancy)	Full-term vaginal delivery	SCS was used for the whole duration of labor
Hanson et al (2006)[15]	37	Central catheter line insertion (30 months before pregnancy)	Full-term vaginal delivery	SCS was used during labor
Saxena et al (2009)[16]	30	Viral infection (11 years ago)	Full-term vaginal delivery	SCS seems to be safe in the first two trimesters of pregnancy
Yoo et al (2010)[17]	32	Pedestrian traffic accident (several months before pregnancy)	Missed abortion before 16 weeks of gestation	SCS was used for the pain control in early pregnancy
Kato et al (2013)[7]	31	Venipucture (at age 18)	Cesarean delivery under spinal anesthesia	Continuous burning pain appeared over the abdomen during Cesarean section under spinal anesthesia
Ahmed et al (2016)[11]	24	Work-related injury of hand (2 years before pregnancy)	Full-term vaginal delivery	SCS use during pregnancy is limited, SCS did not affect adversely to pregnancy
Mansour et al (2017)[18]	33	Right arm blunt trauma (13 years ago)	Full-term Cesarean delivery (38 weeks of gestation) under general anesthesia	The previous Cesarean delivery brought the patient exacerbation of CRPS symptoms such as burning pain and allodynia due to inadequate block, general anesthesia was used

Abbreviations: CRPS, complex regional pain syndrome; SCS, spinal cord stimulation.

There is little literature about pregnancy with CES, however, most of them describe newly diagnosed CES during pregnancy or how to manage pregnancy in disc herniation, which is a far wider spectrum of spine disorder. Most of literatures about pregnancy and CES describe how to diagnose CES during pregnancy since pregnancy can mask the disease or how pregnancy affects to the development of CES. Vakos et al, reported a case of a pregnant woman who developed CES at 30th week of gestation.[13] She underwent surgical decompression in a prone position and later delivered her baby through Caesarean section at term gestation. Raj et al, revealed a case of CES caused by the spinal hemangioma at 31 weeks' gestation.[14] She complained of acute urinary retention and neurological features and imaging confirmed spinal hemangioma in T12 level, compressing the posterior dural space and abutting the cord. Thus, diagnosis of CES in pregnancy is difficult and can be masked since back pain is relatively common in pregnancy. There was no report or article describing a pregnancy or delivery of a woman who has CES and CRPS as underlying diseases at the same time, therefore this case report is very unique and meaningful. A few case reports or brief review articles discussed the pain control of CRPS during pregnancy and labor. Table 2 demonstrates the literature reviews about pregnancy and delivery after CRPS has been diagnosed in women with reproductive age.

Pregnancy itself is a serious factor altering both physical and mental condition of a woman and is sometimes related to various morbidities. The patient of this case report already had unsolvable pain and disability that had been sustained for over a decade, however, she succeeded in pregnancy after two times of failures (miscarriage). Her pregnancy was successfully maintained despite many obstacles, and finally resulted in vaginal delivery. In the current literature, this is the first case of vaginal delivery from pre-existing CES combined with CRPS. Close monitoring and frequent discussion among experts from various departments were the key factors for the successful delivery although the most important condition was the patient's motivation and strong will for her baby. There is a definite limitation of this report that case presentations do not provide conclusive evidence of management or treatment effectiveness, therefore future studies should be followed to establish the protocol for women with CRPS and spine disorder when pregnant. Despite of the limitation, this case will be helpful to counsel for attending physicians who experience to meet the pregnant women who have CES and CRPS together and will inspire other pre-existing CES or CRPS patients who are afraid to decide on pregnancy.

Abbreviations

CES, cauda equina syndrome; CRPS, complex regional pain syndrome; TENS, Transcutaneous Electrical Nerve Stimulation.

Data Sharing Statement

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Ethics Approval and Consent to Participate

The study protocol and publishment of the case details were approved by the Institutional Review Board of Seoul National University Bundang Hospital (B-2403-887-701) and written informed consent form was obtained from the patient. All methods involving human data in this study were performed in accordance with the institutional guidelines or the Declaration of Helsinki.

Consent for Publication

Written informed consent for publication of case details and any accompanying images has been obtained from the participant in this study. The signed permission for publication from the patient of this case report is available from the corresponding author.

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Disclosure

The authors declare no competing interests in this work.

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