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CASE REPORT

Postpartum hypogalactia in a woman with Brown–Séquard-plus syndrome: a case report

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Study design: A case report.
Objectives: To present a case of postpartum hypogalactia in a woman with Brown–Séquard-plus syndrome (BSPS).
Setting: Outpatient spinal cord injury (SCI) clinic.
Case report: A 33-year-old woman with C4 AIS D tetraplegia (American Spinal Injury Association Impairment Scale) was followed at the outpatient SCI clinic for the past 5 years. Her right side presents with increased tone, increased spasticity and decreased sensitivity to light touch. Conversely, her left side presents with minimal changes in tone and no motor function deficits, but decreased sensitivity to pinprick and temperature sensation. These findings are consistent with BSPS. After inpatient rehabilitation, she was engaged, married, and 8 months ago delivered a healthy child. After an uncomplicated delivery, breastfeeding was attempted, but a significant lack of lactation was noted the first month postpartum from the right breast. Despite the implementation of measures to increase lactation, the lack of lactation from the right breast persisted, and required initiation of formula feeding. The right breast in this case lost not only sensory proprioception, but also autonomic control, which could contribute to this instance of asymmetric lactation.
Conclusion: In addition to motor and sensory dysfunctions following SCI, autonomic dysfunctions are commonly seen in individuals with these devastating injuries. The lactation on the right side, which had interrupted descending spinal autonomic pathways, was decreased by approximately 83%. This case provides us with interesting information regarding attention that clinicians should be paying when discussing the breastfeeding options for women with SCI.
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Keywords: autonomic function; lactation; hypogalactia; Brown–Séquard-plus syndrome; spinal cord injury

INTRODUCTION

Typical Brown–Séquard syndrome is seldom reported and characterized by hemiplegia, ipsilateral hypoesthesia and contralateral analgesia. However, Brown–Séquard-plus syndrome (BSPS), with bilateral asymmetric involvement of upper and/or lower extremities, is clinically much more common. It is defined as an incomplete spinal cord injury (SCI) syndrome with ipsilateral weakness and contralateral loss of both pinprick and temperature sensation.[1]

As a result of injury in people with BSPS, we observe motor and sensory dysfunctions due to disruptions of the corticospinal and spinothalamic spinal tracts. However, due to damage of the descending reticulospinal autonomic pathways, various autonomic dysfunctions also can be observed. Horner's syndrome may be present, attributed to the involvement of ipsilateral descending sympathetic fibers within the cervical spinal cord.[2] Lactation function is also known to be under autonomic (sympathetic) control. However, little is known about the effects of SCI on lactation and breastfeeding. With this case, we are presenting our observations of a young woman who had sustained BSPS and presented with postpartum hypogalactia.

CASE REPORT

A 33-year-old woman with C4 AIS D tetraplegia (American Spinal Injury Association Impairment Scale) was followed at an outpatient SCI clinic following her discharge from inpatient rehabilitation after being involved in a motor vehicle accident. Her injury was classified as BSPS, which affected the right side of motor function, including muscle weakness and spasticity. Motor examination revealed no motor weakness on the left upper extremity (total motor score 25), while there was slight weakness present on the right (total motor score 21).

Four years following her SCI, she successfully delivered a healthy baby boy. During the first month after delivery, she realized that there was a significant lack of lactation from her right breast. A trial of feedings with equal exposure to each breast was performed but unsuccessful. The pediatrician recommended that she use a breast pump to collect milk several times a day in order to feed the baby, yet each time the volume produced from left breast was about 60 ml, whereas the volume of milk from the right breast was varied between 5 and 10 ml. Though the mother diligently followed all recommendations, maintained good nutrition and acquired good feeding techniques that would keep her prolactin level within normal range for a

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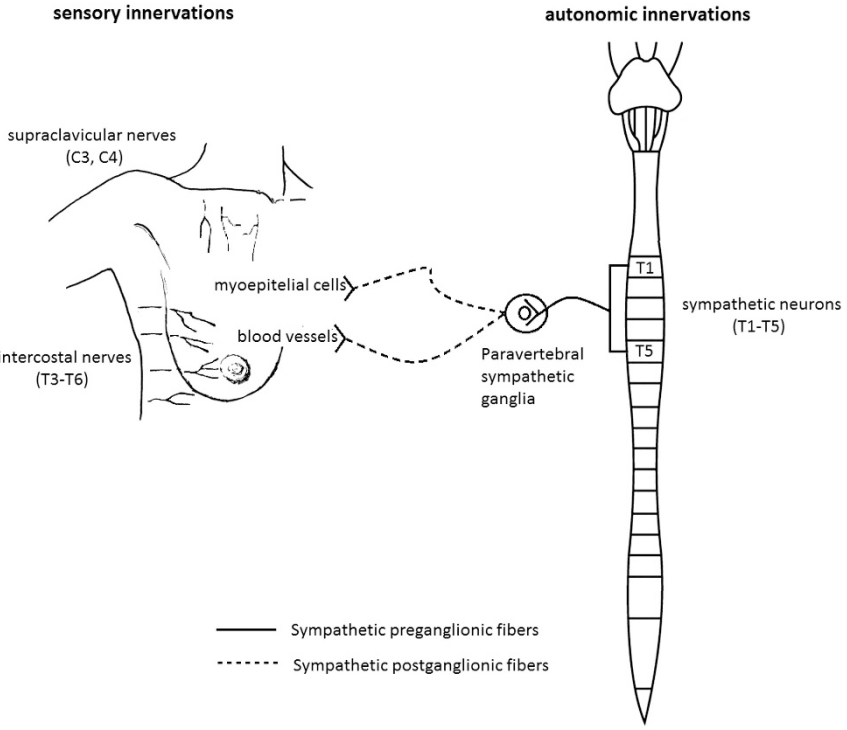

Figure 1 Sensory and autonomic innervations of the breast.

lactating female, unfortunately there was no improvement in lactation from the right breast and she eventually switched to feeding with formula.

DISCUSSION

We are presenting the case of a young female who is presenting with postpartum hypogalactia in the presence of BSPS. The lactation on the right side with interrupted descending spinal autonomic pathways was decreased by approximately 83% (10 versus 60 ml on the left).

It was previously documented that, there is asymmetric milk production present in the able-bodied population.[3,4] Based on the data provided by Cox *et al.*,[4] it is evident that milk production from the right breast (50 ± 17 ml) was higher than from the left (38 ± 12 ml). There was approximately a 24% difference in the milk production between right and left breast in able-bodied women during the first month of lactation (comparable lactation period in our case). However, this was not consistent with findings in our case; the right breast produced 83% less milk than the left in our case during the first month of lactation.

The breast tissue receives both sensory and autonomic innervations (Figure 1). The efferent autonomic innervations of the mammary glands are entirely sympathetic in origin and originate from the sympathetic preganglionic neurons of the upper thoracic spinal cord (T1–T5). The primary sensory innervation is provided through the third, fourth, fifth and sixth intercostal nerves. The upper portion of the breast skin also receives innervation from the supraclavicular nerves (C3–4). In addition to neuronal control (sensory and autonomic), hormonal stimulation is also crucial for lactation. A balanced coordination between sensory, autonomic and hormonal control is required for the peripheral stimulation, central mechanisms and myoepithelial cells to work together in order to produce and eject milk at an appropriate time.[5] Unfortunately, in this case these mechanisms were disrupted on the right side by the SCI that resulted in BSPS. However, there was partial preservation of light touch sensation present in the chest area in our case. This would suggest that in the presence of partial preservation of the sensation, as in this case, only lack of sympathetic control would be responsible for the asymmetric lactation. A screen for low prolactin could be warranted if decreased lactation is present in women with SCI, considering up to 60% of individuals with SCI also have a concomitant traumatic brain injury[6] that could associate with low prolactin levels.[7] However, it would be expected that each breast will be exposed to the same concentration of blood prolactin, and thus asymmetry in milk production probably could not be explained by hormonal changes alone.

The International Standards to document remaining Autonomic Functions after SCI (ISAFSCI) were developed in 2009.[8] These standards identify the autonomic function of blood vessels, heart, respiratory tract, sweat gland, bowel, urinary bladder and sexual function. However, neural control of mammary glands and lactation has not been included in the standards yet.

CONCLUSION

Women represent a minority of the SCI population. As such, very little is known about autonomic dysfunctions that could affect women's health during pregnancy or childcare. This case provides us with interesting information regarding attention that clinicians should be paying when discussing the breastfeeding options for women with SCI. The level and the severity of disruption to central sympathetic control of the mammary glands could play a crucial role in the ability for women to be able to breastfeed postpartum. Further research is needed in this poorly examined area. Additionally, lactation, as a part of autonomic function, should be supplemented into the autonomic standards.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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