Case Report

Complete Sacral Spina Bifida

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Abstract

Spina bifida occulta is found in 10-30% of the population, and most commonly occurs at L5. Such bony malunions are usually asymptomatic but rarely may be associated with neurological sequelae. Most rare is complete spina bifida of the sacrum. The authors discuss the characteristics of and potential clinical and surgical issues arising from this dysembryology, and two cases of complete malunion of the posterior elements of the sacrum.

Key words: anatomy, back, spine, surgery, vertebral column
Introduction

Spina bifida is the most common congenital vertebral abnormality and is often considered the most complex birth defect that is compatible with survival. Several variations of the disease exist, ranging from open spina bifida cystica to the more benign closed spina bifida occulta (SBO). SBO refers to a heterogeneous group of spinal dysraphisms in which the lesion is covered by skin and often has a cutaneous marker such as a hairy nevus, capillary hemangioma, or dimpling of the skin. Patients often go undiagnosed and are identified later in life if they develop urologic problems, neurological problems of the feet, or low back pain. SBO is thought to be due to an error in tissue separation during secondary neurulation. This error results in dorsal nonunion of the vertebral laminae. SBO can occur at any level of the spinal column but is most common in the lumbosacral region, especially from L3 to S1.

Lower sacral spina bifida is one of the most common forms of SBO and is often characterized as an anatomic variant instead of as a pathological condition. It is typically thought of as a minor defect and is often not even mentioned by the radiologist when lumbosacral radiographs are interpreted. Typically, the dorsal laminae of S4 and S5 remain underdeveloped, thereby leading to the formation of the sacral hiatus. There is disagreement as to what should be classified as SBO because a large percentage of the population have dorsal openings at S3, S4, or S5. In fact, openings at these levels are often considered normal variations of the sacral hiatus. However, defects in the dorsal laminae of S1 and S2 are less prevalent and occur in only about 2% and 4% of individuals, respectively. A cadaveric study conducted by Albrecht et al. found that the most common location for sacral defects was from S3 to S5, with 100% of their specimens having an open S5. They did not report any specimens with total sacral SBO. Total sacral SBO, in which there is failure of fusion of the dorsal lamina from S1 to S5, is very rare. The rarity of this condition may be due to the ability of the body to form calcifications that eventually close the open sacral canal. The formation of such calcifications is one explanation for why the prevalence of SBO is much higher in children than in adults. Total sacral SBO is a rare anatomic variant that is important to identify especially in the setting of symptomatic lumbar spinal disorders that require intervention of some type. Total sacral SBO may be pertinent to the placement of caudal epidural blocks (CEB) or screws for spinal fusion of L5 to S1 and thus clinically significant for anesthesiologists, orthopedic surgeons, and neurosurgeons.

Case Illustrations

The first case involves a dry bone specimen of the sacrum (Figure 1) from a male skeleton aged approximately 65 years that was identified in the anatomy laboratory. The entire sacrum exhibited spina bifida. No other variations of the sacrum or remaining skeleton were identified. The second case involves a pediatric patient with back pain. A 14-year-old female presented with a three-month history of low back pain without trauma. Her physical examination was normal. Anti-inflammatory medications were prescribed, but the back pain persisted. Subsequent radiographs of the spine demonstrated no fractures or pathology. However, complete sacral spina bifida was noted. A CT of the spine (Figure 2) was performed, and better illustrated this bony malformation. The pain improved significantly after the patient had been on medication for one month. At one-year follow up, the patient is relatively pain-free.
Discussion

The clinical impact of sacral SBO varies. Sacral SBO can be merely an asymptomatic anatomic variant that is incidentally identified on radiologic films or during autopsy, or can be a very serious defect that results in neurological deficits. Symptoms often associated with SBO include low back pain, urologic problems, posterior disc herniation, and neurological deficits of the feet. However, these symptoms occur frequently in the population not affected by SBO. Although these symptoms are not specific for SBO, it is important for the physician to rule out SBO, as a diagnosis of SBO may affect the method of treatment. It is essential for the physician to know the exact anatomic layout of the sacrum for procedures such as CEB and screw placement and for approaches through the dorsal sacrum in general. CEB is a common and effective method used to treat chronic low back pain refractory to more conservative treatment modalities. CEB involves injecting corticosteroids directly into the epidural space at the sacral hiatus. Various studies report the success rate of CEB to be around 70-80%. Successful placement of this type of nerve block is largely dependent on correctly identifying the landmarks of the sacral hiatus. Anatomic variations often lead to failure of CEB placement and increase the likelihood of complications such as dural puncture. A study by Sekiguchi et al. found that anatomic variations, including bony septum, absent hiatus, and complete agenesis of the posterior sacral foramina, account for 7% of CEB failure. Spinal dysraphisms such as SBO also make difficult the achievement of reduction and posterior fusion with screws. The anatomic variation makes anterior fusion more appealing, as it minimizes the manipulation of the spinal cord and other posterior structures.

Conclusion

SBO is a congenital disorder and results from failure of the posterior vertebral foramina to fuse. Although it is commonly asymptomatic and goes undiagnosed, clinicians must keep it in mind when using treatment modalities such as CEB and spinal fusion surgery. Failure to recognize this anatomic variant may result in an increased rate of complications.

Acknowledgments

None

Conflict of Interest

The authors declare that they have no conflict of interest.
References

Figure 1: Bony anatomic specimen from an adult male skeleton. Note the complete sacral spina bifida.
Figure 2: Reconstructed CT of the posterior pelvis. Note the complete sacral spina bifida.