Case Report

Sacral butterfly vertebrae in the setting of a sacral fracture and unstable pelvic ring injury: A case report and review of the literature

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ABSTRACT

Butterfly vertebrae are rare developmental anomalies representing failure of formation of the vertebral body. There have only been 8 previous reports of a butterfly vertebra occurring at S1. This is the first described case of a butterfly vertebra presenting with a sacral fracture and pelvic ring injury.

Introduction

Butterfly vertebrae are congenital sagittal defects in the vertebral body caused by failure of fusion of the two chondrification centers during embryonic development [1]. Although butterfly vertebrae occur as part of several syndromes such as Alagille syndrome, Crouzon syndrome and Pfeiffer syndrome, incidental butterfly vertebrae are rare with only scattered reports throughout the literature [2]. There have only been eight previous reports of a butterfly vertebra occurring at S1 most of which occurred as part of the syndrome known as spondylolocostal dysostosis [3–6]. Our case describes an incidental finding of a scaral butterfly vertebra in a patient with a Denis zone 1 sacral fracture with superior and inferior rami fractures (AO/OTA 61-C1.3). This is the first described case of a butterfly vertebra presenting as an incidental finding with a sacral fracture and pelvic ring injury.

Case Report

History

This is a 34 YOM roofer who fell 20 ft off a roof landing on his back and was subsequently unable to ambulate secondary to pain. He was brought to our facility by ambulance where a level 2 trauma was activated. He had a previous history of multiple trauma following a fall 2 years earlier during which he suffered from a closed pilon fracture. This was treated with external fixation and delayed open reduction and internal fixation at the time. His history was also significant for the removal of a cystic hygroma from his chin but was otherwise healthy.
Examination

On exam the patient was found to be alert, hemodynamically stable, and neurologically intact but complaining of low back pain on palpation of his sacrum. Computer tomography (CT) scan of his chest and abdomen revealed healed previous rib fractures from past injury as well as a Kommerell diverticulum with an aberrant left subclavian artery (Fig. 1) however no other rib abnormalities were noted. AP radiographs (Fig. 2) and computer tomography scanning (Figs. 3 and 4) of the pelvis showed the presence of a complete left Denis zone 1 sacral fracture with superior and inferior rami fractures (AO/OTA 61-C1.3). The patient was found to have a congenital sagittal cleft of the S1 vertebral body—or butterfly vertebrae which was an incidental finding. He was also noted to have sacralization of his left L5 vertebrae, with pseudoarticulation of the transverse process with the sacral ala through which he had also fractured. CT examination of the cervical, thoracic and lumbar spine did not reveal any other congenital malformation, however the patient was noted to have a traumatic compression fracture of L2.

Operation

The patient was taken to the operating theater and found to have an unstable pelvis under fluoroscopic stress examination with external rotation of the pelvis. A closed reduction maneuver was performed and the S2 transosseous screw corridor was utilized for a percutaneously placed fully threaded 7.3 mm cannulated screw. This path was chosen as the S1 corridor was abnormal. A retrograde anterior column partially threaded 7.3 cannulate screw was used to get additional fixation and stability of the pelvic ring (Fig. 3).

Post-operative course

Post operatively the patient was kept non-weight bearing in his left lower extremity but was able to mobilize with assistive devices and was discharged uneventfully several days later. Two months status post instrumentation the patient was walking...
unassisted despite our non-weight bearing restrictions with minimal pain. On examination at this time he had some weakness in his abductors but was neurologically intact distally. His radiographs showed interval healing without displacement of his hardware (Fig. 5). He was subsequently lost to follow up despite multiple attempts to contact him by phone.

Discussion

Butterfly vertebrae are rare congenital abnormalities of the spine where there is persistence of the notochord leading to a symmetric fusion defect which results in a sagittal cleft in the vertebral body. While congenital abnormalities of the spine are common, there are only a handful of case reports in the literature of a butterfly vertebra in the lumbar and thoracic spine [7]. Typically the lumbar spine has been reported as the most common location for a butterfly vertebra [1]. Moreover, there are only 8 reported cases of the abnormality occurring in the sacrum, most of which were part of a systemic genetic illness known as spondylocostal dysostosis. This is the first reported case of a butterfly vertebra occurring as part of a sacral fracture and pelvic ring injury.
Table 1
Cases of S1 butterfly vertebrae.

| Author       | Year | Age | Sex | Single or multiple | Location | Syndrome                        |
|--------------|------|-----|-----|--------------------|----------|----------------------------------|
| Brasili      | 2002 | 36  | F   | Multiple           | L5, S1   | None                            |
| Takikawa     | 2006 | 4   | F   | Multiple           | L5, S1   | Spondylocostal dysostosis       |
| Takikawa     | 2006 | 3   | M   | Multiple           | L5, S1–S3| Spondylocostal dysostosis       |
| Takikawa     | 2006 | 5.5 | M   | Multiple           | C6, C7, T1–T3, L4, L5, S1, S2 | Spondylocostal dysostosis       |
| Takikawa     | 2006 | 0.75| M   | Multiple           | C6, C7, L2, S1, S2 | Spondylocostal dysostosis       |
| Takikawa     | 2006 | 5   | M   | Multiple           | C5, C6, L2, C7, S1, S2 | Spondylocostal dysostosis       |
| Boulet       | 2011 | 35  | M   | Single             | S1       | None                            |
| Kapetanakis  | 2016 | 40  | F   | Single             | S1       | None                            |

While this patient did have other abnormalities of his sacrum and lumbar spine, he did not exhibit any signs of spondylocostal dysostosis which typically presents with multiple vertebral abnormalities (≥2) short stature and rib fusions [4].

The first reported case of an S1 butterfly vertebra was discovered by Brasili in the Frassetto osteological collection and belonged to an otherwise healthy 36 year old female [3]. The second was reported by Kapetanakis in a 40 year old female with sciatica resulting from stenosis caused by the butterfly vertebrae [5]. The third case occurred in a 35 year old male with low back pain and was treated conservatively [6]. Cases 4–8 all occurred in patients with spondylocostal dysostosis [4]. See Table 1 for a complete list of sacral butterfly cases.

Our patient was otherwise healthy and had no significant history of a genetic syndrome. He did have a history of cystic hygroma as well as an incidental Kommerell diverticulum of his aortic arch with an aberrant left subclavian artery discovered on CT. These specific abnormalities have not been known to occur as part of a constellation of problems with butterfly vertebrae. However, multiple abnormalities of the cardiovascular system and aortic arch have been reported to occur with butterfly vertebrae, specifically coarctations of the aorta, dextrocardia and teratology of Fallot [4,8,9]. While certainly part of the pelvis, the butterfly configuration of the S1 may not have played a role in the fracture as no defect in the sacral ala was noted on imaging.

The characteristic anatomy of butterfly vertebrae is that of two hemivertebrae which have failed to fuse. They may create focal kyphosis which is generally compensated for with anterior endplate overgrowth of the adjacent vertebrae [2]. Histological reports have shown that the butterfly cleft to be composed of chondrocytes in a myxoid hyaline stroma similar to nucleus pulposus tissue [10].

We present this case to highlight an interesting morphological rarity of the sacrum as an incidental finding in a complex pelvic ring injury requiring surgical fixation. It is important to recognize butterfly vertebrae on imaging as generally benign and not part of other pathological processes such as trauma or neoplasm.

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Conflicts of interest/disclosures

None.

References

[1] F. Müller, R. O’Rahilly, D.R. Benson, The early origin of vertebral anomalies, as illustrated by a “butterfly vertebra”, J. Anat. 149 (1986) 157–169.
[2] P. Cave, Butterfly vertebra, Br. J. Radiol. 31 (1958) 503–506, https://doi.org/10.1259/0007-1285-31-369-503.
[3] P. Brasili, B. Bondiglioli, A.R. Ventrella, A case of “butterfly” vertebrae from Sardinia, Int. J. Osteoarchaeol. 12 (2002) 415–419, https://doi.org/10.1002/oa.643.
[4] K. Takikawa, N. Haga, T. Maruyama, A. Nakatomi, T. Kondoh, Y. Makita, A. Hata, H. Kawabata, S. Ikegawa, Spine and rib abnormalities and stature in spondylocostal dysostosis, Spine 31 (2006) E192–E197.
[5] S. Kapetanakis, E. Giovannopoulos, E. Nastoulis, T. Demetriou, Butterfly vertebra. A case report and a short review of the literature, Polia Morphol. (Warsz) 75 (2016) 117–121, https://doi.org/10.5603/FM.a2015.0066.
[6] C. Boulet, A. Schiettecatte, J. De Mey, M. De Maeseneer, Case report: imaging findings in a “butterfly” vertebra, Acta Neurol. Belg. 111 (2011) 344.
[7] G. Patinharayil, C.W. Han, A. Marthya, K.C. Meethall, G.H. Rudrappa, Butterfly vertebra: an uncommon congenital spinal anomaly, Spine 33 (2008), https://doi.org/10.1097/BRS.0b013e31818ad3e1.
[8] D. García-Cruz, H. Rivera, L.O. Barajas, M. Jiménez-Sáinz, Z. Nazará, J. Sánchez-Corona, H. Durón-Huerta, C. García-Ochoa, J.M. Cantú, Monosomy 20p due to a de novo del(20)(p12.2). Clinical and radiological delineation of the syndrome, Ann. Genet. 28 (1985) 231–234.
[9] C.P. Ahn, R.S. Lachman, V.A. Cox, B. Blumberg, O.D. Klein, Brachydactylic multiple delta phalanges plus syndrome, Am. J. Med. Genet. A 138A (2005) 41–44, https://doi.org/10.1002/ajmg.a.30873.
[10] H.-L. Cho, J.-S. Kim, S.S. Paeng, S.-H. Lee, Butterfly vertebra with lumbar intervertebral disc herniation: case report, J. Neurosurg. Spine 15 (2011) 567–570.