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Case report: Imaging findings in a "butterfly" vertebra

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Abstract

A "butterfly" vertebra is a rare congenital anomaly, presenting as a sagittal cleft in the vertebral body. In the literature it has been described as an isolated finding, but it can also be associated with various syndromes, such as Alagille, Jarcho-Levin, Crouzon and Pfeiffer syndrome. We present a case of a 35-year old man with chronic low back pain. The diagnosis of a butterfly vertebra of S1 with disc space narrowing of L5-S1 was made. Usually the abnormality occurs at the thoracolumbar spine and we are not aware of a description of a sacral butterfly vertebra. Although a butterfly vertebra is usually asymptomatic, it could lead to discal herniation or chronic back pain due to alteration in spinal biomechanics.

Introduction

First described by Rokitansky in 1844 (1), a sagittal cleft vertebra is quite uncommon (2-5). Other congenital vertebral anomalies such as block vertebra, transitional vertebrae or hemivertebra are more common.

A sagittal cleft vertebra consists of a symmetric fusion defect, leading to a funnel-like cleft in the vertebral body. This defect is considered to occur between the third and sixth week of gestation. The vertebral bodies are formed out of two lateral chondral centers that would normally fuse. If one fails to develop completely, it results in a hemivertebra. If the fusion fails, it generally results in a sagittal cleft or the so called "butterfly" vertebra, hence the shape of the two hemi-vertebral bodies representing the wings of the butterfly as seen on an anteroposterior radiograph (2).

Case report

A 35-year-old man was referred to our orthopedic department for evaluation of chronic low back pain since 1.5 years, irradiating to his sacro-iliac joints and upper legs. He had a sedentary occupation. The pain became worse in the evening.

Fig. 1. — Anteroposterior lumbar radiograph. Funnel-like cleft is seen at S1 (white arrow). Spinous process of L5 and spina sacralis of S1 are intact, excluding a spina bifida occulta (black arrows). Note the minor dextroconvex scoliosis.

On clinical examination the patient had no obvious scoliosis. No sensory or motor deficit was observed and reflexes were normal and symmetric. He consulted an urologist previously for frequent micturition and a burning sensation in his lower abdomen, but no renal or bladder pathology was found. Blood analysis was normal.

Anteroposterior lumbosacral and pelvic radiographs revealed a minor dextro-convex scoliosis due to an asymmetry in the corpus of L5 and the sacrum

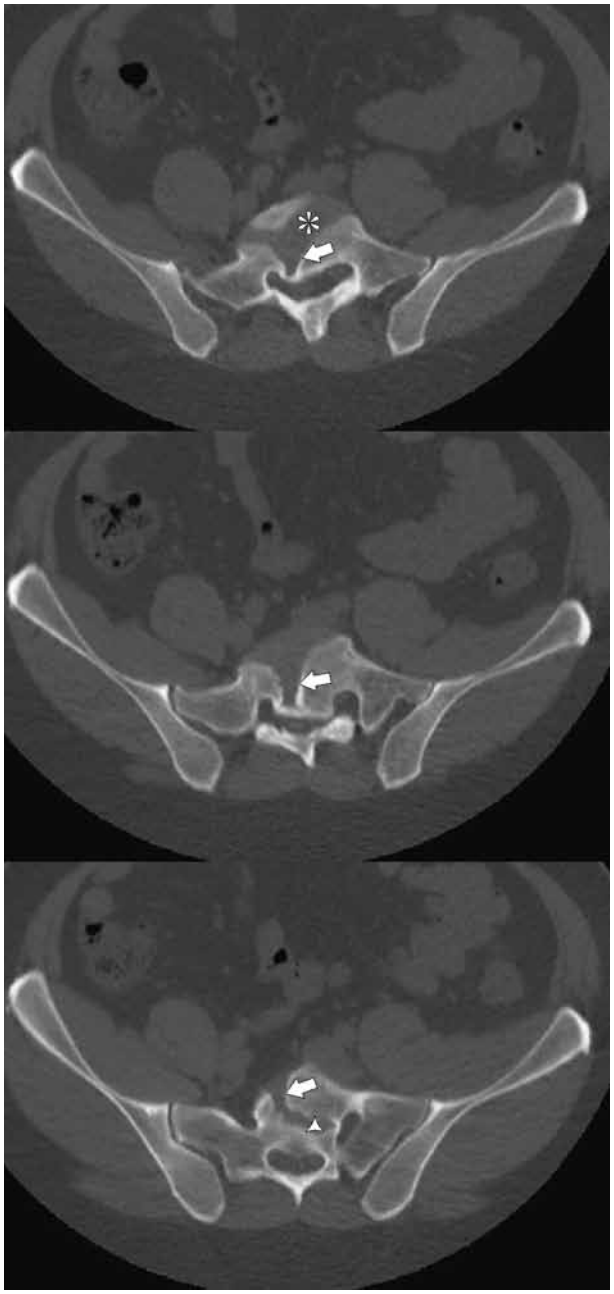

FIG. 2. — Axial CT reconstruction in bone windows (consecutive images). Sagittal cleft in the S1 vertebral body (white arrow), connecting the L5-S1 disc (asterisk) with S1-S2 (arrowhead).

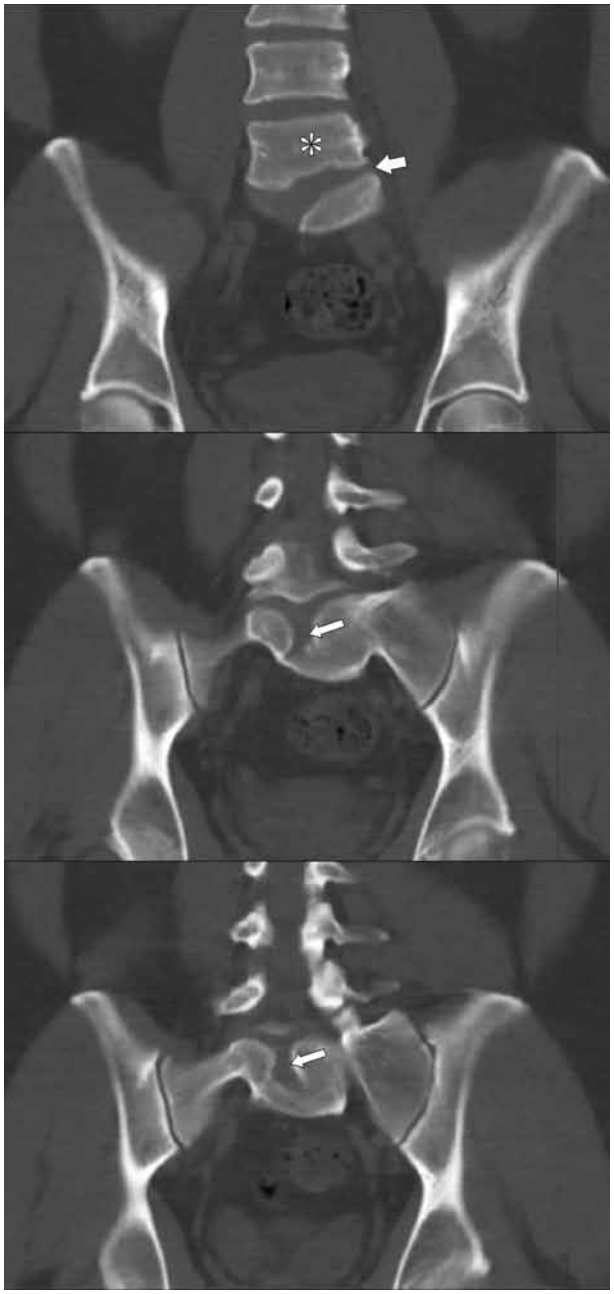

FIG. 3. — Coronal CT reconstruction in bone windows (consecutive images). Wedging of the vertebral body of L5 (asterisk) with underlying partial narrowing of L5-S1 disc (short arrow) on the left side. The sagittal cleft is clearly seen (long arrow).

and a slightly narrowed disc space at the L5-S1 level. It also showed a sagittally oriented lucency in the promontorium and S1 with sclerotic borders (Fig. 1). The processus spinosus of L5 appeared to be intact, without evidence for spina bifida. CT of the abdomen, performed during the examinations for his urological symptoms, was retrospectivally reviewed. It showed a sagittal cleft in the vertebral body of S1,

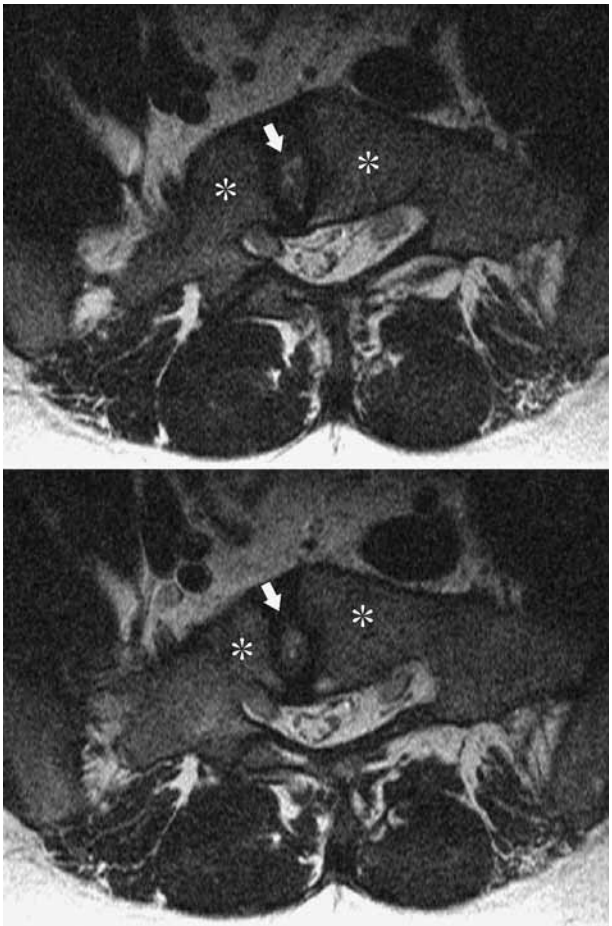

FIG. 4. — Axial T2-weighted images at the S1 level (consecutive images). Sagittal cleft with internal disc-like material (arrow). Notice the asymmetric hemi-vertebrae of S1 (asterisks).

connecting the discus of L5-S1 and S1-S2 (Fig. 2). The left hemivertebra of S1 was slightly larger than the right, resulting in a minor scoliosis and secondary enlargement of this side of L5 (Fig. 3). No discal bulging, nor intra-or extraforaminal radicular conflict was evident.

Because of the clinically evident irradiating pain, magnetic resonance of the lumbar spine was performed. It showed a funnel-like sagittal cleft in the body of S1, and an hypoplasia of the right side (Fig. 4). Inside the cleft discal material was seen, connecting the L5-S1 and S1-S2 disc spaces (Fig. 5). Discal dehydratation and narrowing of L5-S1 were seen, as well as a laterolateral asymmetry of L5, causing the dextroconvex scoliosis. No radiculopathy was seen.

The patient's pain was likely caused by the static alteration of the lumbosacral spine, resulting in muscle spasm and low back pain.

Discussion

A butterfly vertebra is believed to be a result of the persistence of the notochord during gestation (5). The cleft can be narrow or wide or even be bridged by bony strands. The hemi-vertebral bodies can be symmetrical or asymmetrical. Anterior wedging can be seen and it can be associated with kypho-scoliosis (2). It can also be associated with other vertebral anomalies, such as block vertebrae, supernumerary lumbar vertebrae or even spina bifida (6). Diastematomyelia is often associated with a butterfly vertebra (7). Some syndromes can also be associated, like Alagille Syndrome (arterio-hepatic dysplasia), Jarcho-Levins syndrome (spondylocostal dysostosis), Crouzon Syndrome (branchial arch syndrome) and Pfeiffer's syndrome (craniosynostosis). Patients with 22q11.2 deletions and monosomy 20p are also known to present with butterfly vertebra (3). Our patient did not have any of the above syndromes. Usually the butterfly vertebra occurs at the thoracic or lumbar level, and a sacral localization has not yet been described.

A butterfly vertebra is considered to be asymptomatic, but due to alterations in spine biomechanics associated with disc changes, asymmetry in vertebral bodies and scoliosis, it can give rise to muscle spasm resulting in chronic low back pain. In more pronounced cases, disc and facet joint degeneration could lead to a narrowed spinal canal and neuroforamina with more severe complications (9).

Radiographs of the spine may show minor scoliosis and slight asymmetry of the vertebral bone. The funnel like defect in the vertebral body can be visualized. It could be mistakenly interpreted as spina bifida, but the lumbar and sacral spinous processes are preserved in a butterfly vertebra. In dorsal and lumbar butterfly vertebrae significant antero-posterior wedging is demonstrated (4-7). This was less so in our patient.

CT is an excellent modality to depict the funnel shaped defect and possible wedging of the vertebral body as a result of asymmetry associated with butterfly vertebra. The facet joints are usually normal.

Magnetic resonance shows disc like material in the funnel shaped defect connecting the adjacent disc spaces (Fig. 5). Butterfly vertebra may give rise to adjacent disc protrusions (8). Our patient had no disc protrusion or hernia, but presented with disc narrowing at the L5-S1 level (Fig. 6).

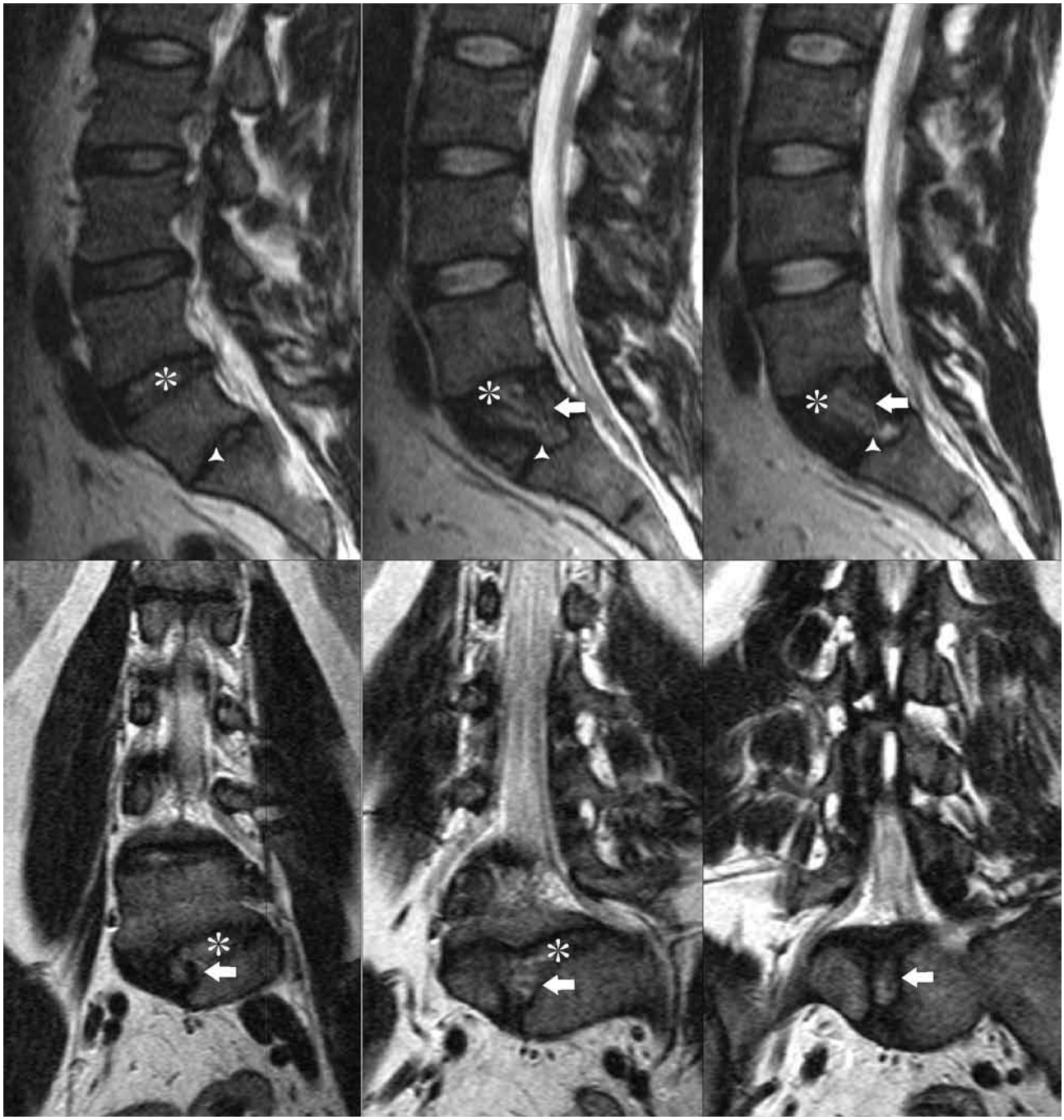

FIG. 5. — Sagittal and coronal T2-weighted images (consecutive images). Disc-like material inside the butterfly vertebra (arrow), connecting the L5-S1 disc (asterisks) with S1-S2 (arrowheads).

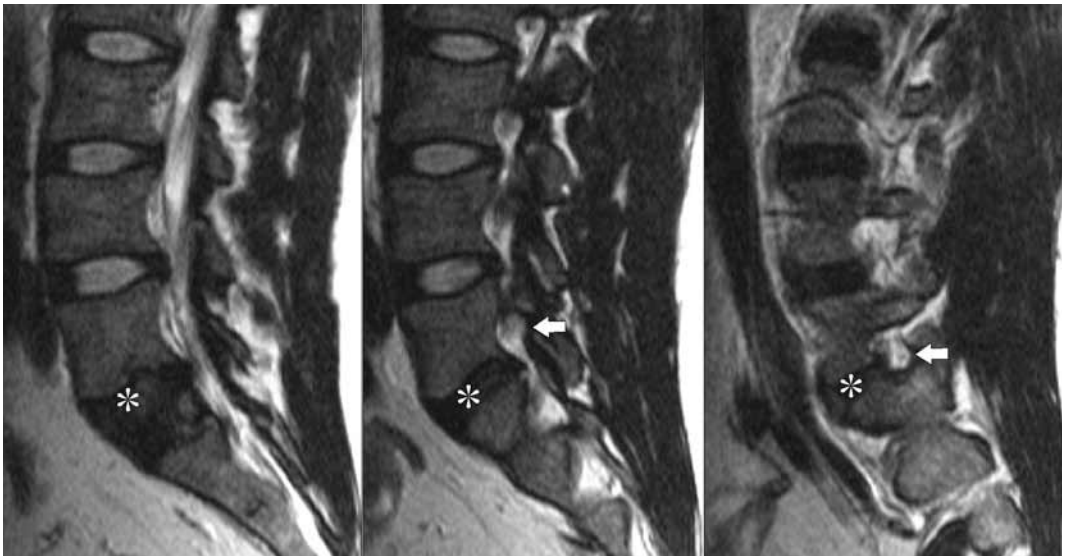

FIG. 6. — Sagittal T2-weighted images (consecutive images). Left sided disc dehydration (asterisks), but no adjacent neuroforaminal narrowing is observed (arrow).

In conclusion, butterfly vertebrae which are generally considered asymptomatic may increase the incidence of disc problems, especially when they occur at the lumbo-sacral level. They may also cause scoliosis which may cause muscle spasm with secondary low back pain.

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