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Case Report

A rare incidental finding of an isolated S1 butterfly vertebra: A case report ☆☆☆

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ABSTRACT

Butterfly vertebra is an uncommon type of vertebral anomaly (sometimes referred to as a sagittal cleft vertebra or an anterior rachischisis) caused by persistent notochordal tissue. Butterfly vertebrae of S1, which is rarer anomaly compared to thoraco-lumbar region, may be associated with syndromic causes and usually asymptomatic with a funnel shaped defect seen in imaging which can later give rise to disk problems, facet joint degeneration and chronic low back pain. We here share a case of 35-year female presented with intermittent low back pain diagnosed with S1 butterfly vertebrae as an incidental finding in radiograph and magnetic resonance imaging. Radiologist and orthopedicians should be vigilant about this rare entity as a differential of low back pain and its association with other syndromes.

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Introduction

Vertebral anomalies have an incidence of 0.5-1/1000 live births which can be considered relatively frequent; however, a butterfly vertebra is an uncommon occurrence [1]. Most of them are seen at the lumbar spine when present, followed by the thoracic region with involvement of the sacral spine very rarely encountered and reported [1]. Butterfly vertebra, sometimes referred to as a sagittal cleft vertebra or an anterior rachischisis is caused by persistent notochordal tissue that

prevents the lateral halves of the vertebral body from fusing together [2]. The name is based on the way the 2 hemivertebrae on X-rays appear to emerge as butterfly wings from the central gap [3].

Butterfly vertebra is a congenital anomaly, diagnosed as an isolated finding or as part of syndromic diseases affecting other skeletal regions and/or other systems; particularly the gastrointestinal, genito-urinary, and central nervous systems. It typically appears in the lumbar spine [4]. Low back pain is the most common presenting symptom for both syndromic and non-syndromic cases [5]. Even though butterfly vertebrae

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Fig. 1 – (A, B) Anteroposterior (AP) views of X-ray pelvis (suboptimal quality) shows subtle vertical linear radiolucent area (pointed by white arrow and outlined by white dotted line along the left sided margin of the cleft in B) in midportion of S1 vertebral body consistent with butterfly cleft (this was missed by radiologist at first reading due to its subtlety). No similar cleft seen in other visualized lumbar vertebrae.

are typically thought to be asymptomatic, they may increase the likelihood of disk problems, particularly when they develop at the lumbo-sacral level [6].

We here have a case of a 35-year female presented with intermittent low back pain diagnosed with S1 butterfly vertebrae as an incidental finding.

Case presentation

A 35-year-old female presented to the out-patient department of orthopedic unit at Bir Hospital with complaints of intermittent low back pain for the last 5-6 years, sometimes radiating to the right leg along with concurrent numbness and tingling sensation in the radiated limb. The patient visited multiple clinics and hospitals for her problem in the past without significant improvement in her symptoms. Physiotherapy for her back and medical management prescribed each time by the doctors seemed to be not working satisfactorily for the patient. On physical examination, the patient did not have tender point(s) on palpation in the lower back; power was normal in both the lower limbs; sensory functions were intact; and the straight leg raise test was negative; the patient did not have any issues with her bladder and bowel habits and other relevant urine and blood lab works were normal as well. The patient also underwent X-ray and

then MRI examination where we found S1 butterfly vertebrae as an incidental finding. Many other associations for the butterfly vertebrae were pointed but none of them matched our finding.

X-ray pelvis in AP view showed subtle vertical curvilinear radiolucency at midline of S1 vertebral body (Fig. 1) which was initially missed by an orthopedician and a radiologist. MRI of lumbosacral spine was done after the 'normal' reported X-ray pelvis which demonstrated presence of a low signal vertical cleft at slight right paramedian region of S1 vertebral body in keeping with butterfly vertebral cleft (Fig. 2); normal intervertebral disk height was noted at adjacent L5-S1 intervertebral level (Fig. 3). X ray of cervical spine and chest X-ray showed no other associated vertebral anomalies.

The patient was given NSAIDs for low back pain and counseled about the finding. She is under regular follow-up with physio exercises and is doing well as per writing this case report.

Discussion

Butterfly vertebrae are aberration from the vertebral body during embryological development. The notochord typically forms at day 7 of gastrulation, during the first stage of the spine's development, known as neurulation. The formation of

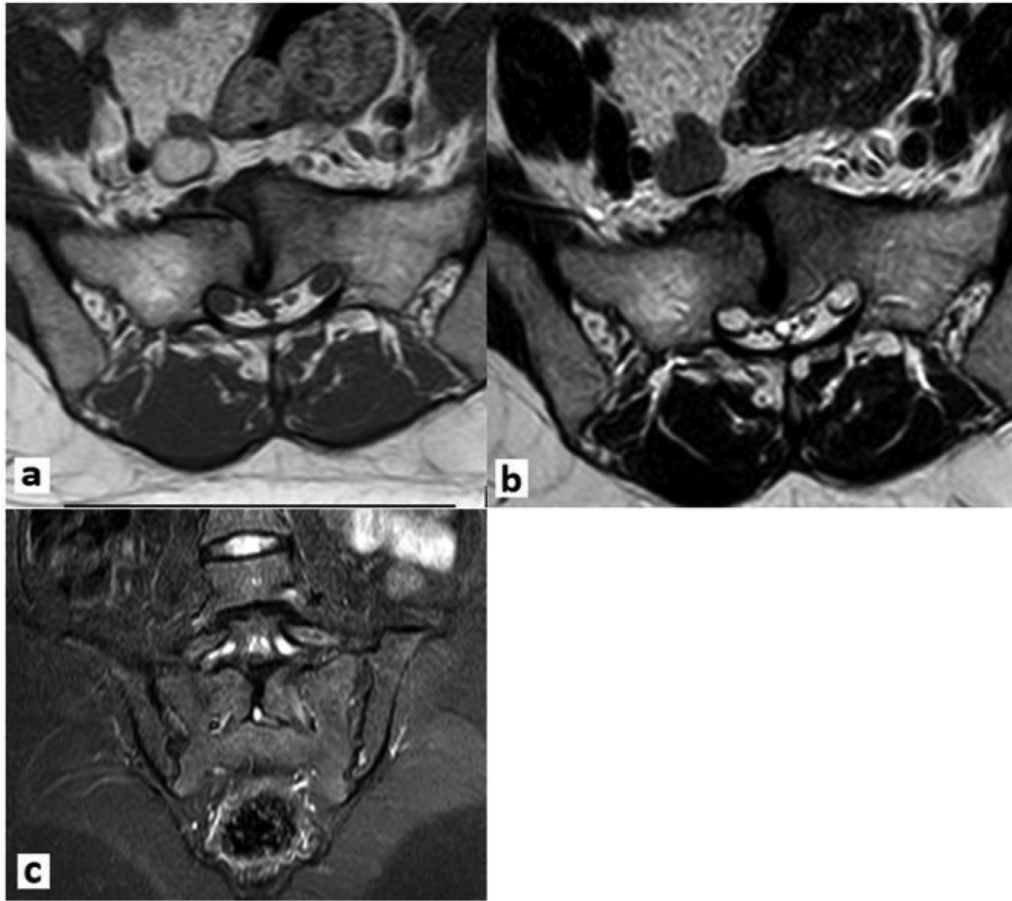


Fig. 2 – MRI axial T1WI (A), axial T2WI (B), and coronal short tau inversion recovery (STIR) image (C) show butterfly defect in slight right paramedian region of S1 vertebra with the cleft filled with material which shows iso signal intensity in T1WI and low-signal intensity in T2WI and STIR sequence. No similar cleft seen in other visualized vertebrae. Other findings: X-ray of C spine and AP chest X-ray show no other associated vertebral anomalies (images not provided).

cardiac, pulmonary, and renal tissue is also a function of the lateral mesoderm, which explains the strong correlation between spinal malformations and problems with other important organ systems [7]. In a systematic review of case reports, the related anomalies in the genitourinary, cardiac, and neurological systems were 30%, 27%, and 23% respectively. Similarly, 43 % of patients with a butterfly vertebra had another musculoskeletal problem [5]. Our patient had no abnormalities in other systems.

In a study, there was a strong correlation between a syndromic presentation and the existence of several butterfly vertebrae. In 89% of cases with more than one butterfly vertebra, a syndrome was also present. But a considerable proportion of cases having a single butterfly vertebra also showed up as a condition (32% of cases) [5]. Many associations are seen with butterfly vertebrae. These includes: anterior spina bifida +/- anterior meningocele, Alagille syndrome, Crouzon syndrome, VACTERL association, Pfeiffer syndrome, or Jarcho-Levin syndrome [1,2]. But our case had no other associated syndrome(s).

The knowledge on radiographic characteristics of butterfly vertebrae is important for differentiating them from other clinical processes such as fracture, metastatic infection, sy-

ringomyelia, and Charcot's disease [3]. Magnetic resonance images show disk-like material in the funnel shaped defect connecting the adjacent disk spaces. Butterfly vertebra may give rise to adjacent disk protrusions [4]. In a case report of an 80-year-old patient presenting with low back pain, in X-ray, there was a lucent defect with sclerotic edges in the central portion of S1 while MRI (sagittal T2) showed incidental S1 butterfly vertebra with a small paraspinous cleft and severe L2-L3 and L3-L4 spinal canal stenosis with proximal cauda equina redundancy [8]. In our case, butterfly defect in slight right paramedian region of S1 vertebra with the cleft filled with disk material was seen which showed iso signal intensity on T1-weighted images and low signal intensity on T2-weighted images and short tau inversion recovery sequence. Our patient had no disk narrowing or significant disk protrusion.

Although a butterfly vertebra is thought to be asymptomatic, due to changes in the spine's biomechanics brought on by disk degeneration, asymmetry in the vertebral bodies, and scoliosis, it can cause muscular spasms and chronic low back pain. A narrower spinal canal and more serious consequences from narrowing of neural foramina could result from more severe disk and facet joint degeneration [9]. Our patient



Fig. 3 – MRI mid-sagittal T1WI (A) and T2WI (B) show normal height of L5-S1 intervertebral disk space with signal intensity similar to the other lumbar intervertebral disks. Superior margin of S1 vertebral body is ill defined due to presence of vertical cleft in midportion as described in previous images.

had intermittent back pain as her symptoms without evidence of degenerative changes at and around S1 vertebral body on imaging.

Conclusion

Butterfly vertebrae of S1 is rare entity mainly associated with syndromic causes and asymptomatic with a funnel shaped defect in imaging which can later give rise to disk problems, facet joint degeneration and chronic low back pain. Thus, radiologist and orthopedicians should be vigilant about this rare entity and its association with other syndromes as a differential of low back pain.

Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Ethics approval and consent to participate

This case report did not require review by the Ethical committee for publication.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images assuring him about the confidentiality. A copy of the written consent is available for review by the Editor-in-Chief of this journal whenever required.

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