Congenital kyphoscoliosis associated with thoracic hemivertebrae in a nine-month-old Racking filly

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Abstract

Thoracic hemivertebra is one of the rare congenital vertebral anomalies in horses resulting in vertebral column deformity with or without neurological signs. A nine-month-old Racking filly was presented with a distinct bump over the back region. This bump was apparent at birth and has been increasing in size. Clinical examination revealed a painless kyphoscoliosis over the thoracic vertebrae with mild ataxia and abnormal gait of hindlimbs. In plain lateral radiographs of thoracic vertebrae, hemivertebrae were seen in the 13th-15th thoracic vertebrae. At myelography performed under general anaesthesia, the ventral and dorsal aspects of the contrast medium column were narrowing at the level of the 13th-14th thoracic vertebrae, and no sign of contrast medium after the 14th thoracic vertebra that represents spinal cord compression. Little information about hemivertebra in horses exists and a better understanding of hemivertebra etiology is required.

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Thoracic Hemivertebrae In A Racking Filly

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Summary

Thoracic hemivertebra is one of the rare congenital vertebral anomalies in horses resulting in vertebral column deformity with or without neurological signs. A nine-month-old Racking filly was presented with a distinct bump over the back region. This bump was apparent at birth and has been increasing in size. Clinical examination revealed a painless kyphoscoliosis over the thoracic vertebrae with mild ataxia and abnormal gait of hindlimbs. In plain lateral radiographs of thoracic vertebrae, hemivertebrae were seen in the 13th-15th thoracic vertebrae. At myelography performed under general anaesthesia, the ventral and dorsal aspects of the contrast medium column were narrowing at the level of the 13th-14th thoracic vertebrae, and no sign of contrast medium after the 14th thoracic vertebra that represents spinal cord compression. Little information about hemivertebra in horses exists and a better understanding of hemivertebra etiology is required.

KEYWORDS

Horse, filly, hemivertebra, kyphoscoliosis, thoracic

Key points

Developmental abnormalities of the thoracic vertebrae are rare in horses.

Hemivertebra is a rare congenital spinal anomaly resulting from failure of formation on one side of the vertebral body.

Hemivertebrae can lead to deformation of spine due to unbalanced spinal growth, resulting in kyphosis, lordosis, or scoliosis with or without neurologic gait deficits.

INTRODUCTION

The vertebral column, as a central axis of the skeleton, is composed of alternating series of vertebrae (Ward et al., 2018). Several developmental vertebral defects are occurred in horses, that cervical vertebral stenotic myelopathy (CVSM) and occipitoatlantoaxial malformation (OAAM) are relatively common developmental abnormalities (Auer & Stick, 2018; Crochik et al., 2009; Unt & Piercy, 2009). However, other congenital anomalies of the vertebral column including atlantoaxial instability, block vertebrae, butterfly vertebrae, hemivertebrae, and spina bifida, occur rarely in horses (Auer & Stick, 2018). These conditions occur as a result of a failure of normal formation, segmentation, differentiation, or union of structures during embryologic development stages (Auer & Stick, 2018; Castriota-Scanderbeg & Dallapiccola, 2006). Hemivertebra is a condition in which asymmetric vertebral body formation occurs so, a part of the vertebral body is deficient (Bertram et al., 2019; White & Goldberg, 2022). Depending on hemivertebra location and severity, it may be associated with moderate to severe angulation of the normal spine alignment with or without neurologic gait deficits (Auer & Stick, 2018; Haussler). In horses, hemivertebra is a rare congenital vertebral malformation, limited to reports of individual cases (de Heer & Nout, 2011; Kirkberger & Gottschalk, 1989; Wong et al., 2005). This case report describes the clinical and radiographic findings of congenital thoracic hemivertebrae in a nine-month-old Racking filly.

CASE HISTORY AND CLINICAL FINDINGS

A nine-month-old 153 kg Racking filly with a distinct bump over the back region was referred to the Surgery Section of the Veterinary Teaching Hospital of the Ferdowsi University of Mashhad. According to the owner, this bump was present since her birth and increased in size over time. There was no history of trauma. On physical examination the filly was alert and her body condition was normal. The respiratory rate, heart rate, and temperature were within normal limits. Examination of the filly back revealed abnormal curvature of the spine along the sagittal plane (kyphosis) with mild scoliosis (kyphoscoliosis) over the thoracic vertebrae (Figure 1). No pain was detected on palpation of this region. Any forelimbs deficits or cranial never abnormalities were not observed. The cervical range motion was normal. Very mild ataxia and abnormal gait in hindlimbs were observed on examination (grade 1/5). No urinary or fecal incontinence was reported by her owner.

The blood sample was obtained and hematology and serum biochemistry profiles were evaluated. Hematology parameters were within the normal range. Biochemistry analyses showed a raised in aspartate aminotransferase (AST: 393; reference range 226–366 U/L) and Creatine kinase (CK: 1083; reference range 108–430 U/L). Plain and contrast radiography was carried out. A myelogram was performed under general anesthesia. For this purpose, the filly was sedated with 1.1 mg/kg of xylazine (20 mg/ml; Alfasan; Holland) intravenously, followed by 0.1 mg/kg of diazepam (5 mg/ml; Zepadic®; Caspian tamin Pharmaceutical Co.; Iran). Then anaesthesia was induced by 2.2 mg/kg ketamine (100 mg/ml; Alfasan; Holland) intravenously and was intubated orotracheally. Anaesthesia was maintained with isoflurane (AErrane®; Baxter Healthcare Corporation; USA). Myelographic evaluation was performed with the cisternamagna technique. The area was aseptically prepared. The head was maintained in a flexed position and an 18G spinal needle was placed in the atlantooccipital space. After 50 ml cerebrospinal fluid (CSF) was withdrawn, intrathecally injection of a similar volume of iodixanol 270 mg/ml (270 mg/ml; Visipaque^{TM}; GE Healthcar Inc.; USA) was done for 3 minutes. Before imaging, the head was maintained in an elevated position. In plain lateral radiographs of thoracic vertebrae, multiple hemivertebrae were seen in the 13th-15th thoracic vertebrae with several abnor-

mal ribs (Figures 2 and 3). At myelography, no sign of significant abnormality in contrast medium column was seen in lateral and flexed radiographs of cervical myelography. In the thoracic region, the ventral and dorsal aspects of the contrast medium column were narrowing at the T13- T14, and the contrast medium was not passed that revealing extradural compression of the spinal cord in mentioned thoracic regions (Figure 4). Collected CSF was evaluated. Various aspects of this disorder including its progression, treatment options, outcomes, prognosis, and animal welfare were fully explained to the owner, but the owner refused its management.

DISCUSSION

In horses, developmental vertebral disorders, commonly cause malformations of the cervical vertebrae (de Heer & Nout, 2011; Dorner et al., 2022; Rendle et al., 2008; Unt & Piercy, 2009; Wong et al., 2005) while thoracic and lumbar vertebral malformations are infrequent (de Heer & Nout, 2011; Wong et al., 2005). Vertebral malformations can be congenital (present at birth) or acquired (later in life) (Wong et al., 2005). Hemivertebra is one of the congenital vertebral abnormalities. Failure of chondrification centers to develop during the formation or the segmentation during somitogenesis stages and failure of ossification of the vertebra due to improper formation of the intersegmental arteries of the vertebral column and therefore alternation in vascular supply at the developmental stage has been proposed as the pathogenesis of hemivertebra (Besalti et al., 2005; Goldstein et al., 2005; Johal et al., 2016; Varras & Akrivis, 2010). Hemivertebrae can result in deviations of the vertebral column in the sagittal plane dorsally (kyphosis), ventrally (lordosis), or medially (scoliosis) (Auer & Stick, 2018; Denoix, 2005).

In humans, hemivertebra is a rare congenital anomaly and its incidence is estimated at 0.05%-0.1% and is more common in females (Shah et al., 2020; Varras & Akrivis, 2010; Xu et al., 2020). In animals, hemivertebra is the most common congenital malformation in dogs (De Rycke & Saunders, 2017; Ryan et al., 2017; Schlensker & Distl, 2016; Wyatt et al., 2018). The breed-related incidence of hemivertebra in dogs has been investigated and reported that hemivertebra is very common in screw-tailed brachycephalic breeds with a prevalence range from 78% to 94% (Davitkov et al., 2020; Wyatt et al., 2018).

In a study, vertebral lesions have been reported in 202/443 horses (38.6%), with the main causes being crowding and overriding of the dorsal spinous processes in the caudal thoracic and cranial lumbar regions. A total of 15 horses (2.9%) vertebral malformations including scoliosis, lordosis, and kyphosis were identified (Jeffcott, 1980).

To date, only a few cases of hemivertebra in horses were reported previously. According to these and the presented case report, the horses affected by hemivertebra are of various breeds, including the Quarter (Wong et al., 2005), Friesian (de Heer & Nout, 2011), and American saddle horse (Kirkberger & Gottschalk, 1989). To our knowledge, this is the first reported case of thoracic hemivertebra in the Racking horse. In the previous case reports of hemivertebra, this condition was described in male horses including geldings and colts (de Heer & Nout, 2011; Kirkberger & Gottschalk, 1989; Wong et al., 2005), but in the present case, hemivertebra reported in female foal (a filly). However, due to a few published case reports, it is difficult to find the association between sex, breed, and incidence of hemivertebra in the horse.

In agreement with the case reported by Wong (Wong et al., 2005), rib anomalies were observed in our case. Ribs irregularity and structural change were observed at radiography in this case. This finding detects as an incidental finding on radiography or post-mortem specimens and does not contribute to neurological signs (Denoix, 2005; Wong et al., 2005). It has been shown that hemivertebra may be associated with other congenital musculoskeletal anomalies including spine, limbs, and ribs malformations, neural tube defects, and extramusculoskeletal anomalies including the cardiac, genitourinary, central nervous system, and gastrointestinal tract anomalies, probably due to anatomically unbalanced growth between hemivertebra and structural differentiation of surrounding spine (Bohiltea et al., 2022; Chaturvedi et al., 2018; Moser, 2005; Varras & Akrivis, 2010; Yang et al., 2020). Because the structure of the ribs is formed in close association with vertebrae during gestation, the same developmental failures that lead to hemivertebra are attributed to rib abnormalities (Fischer & Degenhardt, 2008).

In the present case, kyphoscoliosis was observed. It has been demonstrated that Hemivertebra is one of the important causes of deviation from the normal curvature of the spine because the growth potential of the affected vertebrae is like the normal vertebrae (Yang et al., 2020). This condition can result in different structural deformities of the spinal axis including scoliosis, lordosis, and/or kyphosis (Xu et al., 2020). Structural abnormalities may affect the spinal cord so, a neurological examination should be considered to determine the neurological signs (Kaplan, Spivak, & Bendo, 2005).

Based on the published data about hemivertebra and spine curvature disorders in horses, this condition may be associated with neurological deficits or not (Denoix, 2005; Wong et al., 2005). Even, though it has been noted that the horses with deviations of thoracolumbar without any neurological deficit can be undergoing normal use, these horses are predisposed to intervertebral osteoarthrosis (Denoix, 2005). Unlike the thoracolumbar region, the hemivertebra and deviations of the cervical spine are usually accompanied by neurological deficits (Denoix, 2005). Kirberger and Gottschalk reported a foal affected by Kyphoscoliosis of the thoracolumbar spine (T7-L4) and hemivertebra at T15 but, no neurological deficits were detected (Kirkberger & Gottschalk, 1989). Wong et al described a 9-month-old Quarter gelding with severe hindlimb ataxia and paresis were associated with multiple thoracic malformations of T4-T8 including hemivertebrae, kyphosis, and fused dorsal spinous processes (Wong et al., 2005). In another report by de Heer and Nout, a 5-day-old Friesian colt was presented for kyphosis at the level of T18–L4 that was affected by neurological deficits including pelvic limb paraparesis and severe ataxia (de Heer & Nout, 2011). In the case report published by Kothstein et al, acquired kyphosis a due to compression fractures of T14 to T18 in association with mild hindlimb ataxia was described in a 20-month-old horse (Kothstein et al., 2000). Rendle et al reported a kyphosis of the thoracolumbar spine with thoracic neurenteric cyst and butterfly vertebrae at T6 and T7 in a seven-month-old colt that suffered from very severe neurologic deficits (Dorner et al., 2022). In the presented filly here, a mild neurological deficit was observed in hindlimbs that may be due to compression and narrowing of the spinal cord at the T13. It has been described that several factors such as the severity of kyphosis, hemivertebra subtype, breeds, and presence of other pathologic conditions may be associated with the presence or absence of neurological signs in dogs with hemivertebra (De Decker et al., 2019). However, the development of clinical signs related to hemivertebra in horses is not completely known.

Increased AST and CK were observed in our case and these changes may be related to kyphoscoliosis. Spinal misalignment can alter body biomechanics and muscles loading so, kyphosis may be attributed to several muscle impairments (Briggs et al., 2007; Fasser et al., 2021). Although an elevation in serum CK along with AST is an indication of muscle damage, these enzymes may be affected by other different factors (Harris et al., 1990; Satué et al., 2022).

The etiological factors causing vertebral malformations have not been fully explained in horses, but genetic and heredity factors, dietary imbalances, toxic factors, hormonal changes, environmental cusses, trauma, and in-utero insult may be responsible (Auer & Stick, 2018; Dorner et al., 2022; Kirkberger & Gottschalk, 1989; Kothstein et al., 2000; Unt & Piercy, 2009; Wong et al., 2005). It has been mentioned that alteration in genes expression, especially members of Hox family, may contribute to malformations of the axial skeleton and vertebrae (Unt & Piercy, 2009). However, the role of genetics, heredity and other factors of hemivertebra in horses are generally considered unknown, probably because of the rarity of the condition. The etiology of the present case here remains undetermined as well as other previous hemivertebra case reports (de Heer & Nout, 2011; Wong et al., 2005). Although there were no similar congenital anomalies in filly parents the owner had no knowledge about previous generations. Also, there was no known exposure to toxic substances or trauma. However, since this condition was present at birth, genetics may probably be explaining the anomaly in this case.

Although in humans, early diagnosis and appropriate surgical management are recommended before the development of neurological signs or severe deformity and disability (Han et al., 2011; Oksanen et al., 2021; Ruf et al., 2006; Yang et al., 2020)but, there is no treatment for this condition in horses (Auer & Stick, 2018). In dogs, surgical management of cases with clinical signs associated with hemivertebra with vertebral column malangulation can be challenging, and there are only a few reports of such cases in the veterinary

literature (Charalambous et al., 2014).

Congenital thoracic hemivertebra in horses is a rare anomaly that may be with a deformity resulting in congenital scoliosis and kyphosis. This condition may also cause spinal cord compression and neurological signs. Rib anomalies may occur in association with hemivertebra. The actual incidence and prevalence of thoracic hemivertebra in horses are little known and future reports are required to better understand the different aspects of such an anomaly.

AUTHOR CONTRIBUTIONS

All the authors contributed to the clinical examinations, interpretation of the clinical data, and preparation of the manuscript. All authors approved the final version of the manuscript.

CONFLICT OF INTEREST

No conflicts of interest have been declared.

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ETHICS STATEMENT

This case reports details the management of a clinical case that was a part of the clinical caseload. High veterinary care has been performed with the consent of the animal owner. All identifying information has been removed.

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Figure legends

Figure 1. Clinical appearance of thoracic kyphoscoliosis in the presented filly

Figure 2. Schematic representation of the abnormality in vertebrae noted. Grey shadow areas show abnormal shape of the 12th-16th thoracic vertebrae and wedge shape in body of the 13th-15th thoracic vertebrae.

Figure 3. Laterolateral radiograph of the thoracic vertebrae in the mid to caudal thoracic region. Kyphosis shows in the caudal-thoracic region of the spine caused by hemivertebrae in the 13th-15th thoracic vertebrae.

Figure 4. Laterolateral radiograph of the caudal thoracic vertebrae shows an abnormal myelogram. Note that narrowing of the ventral contrast medium column at the level of the 12th thoracic vertebrae. The ventral and dorsal contrast medium columns are narrower at the level of the 13th-14th thoracic vertebrae and no sign of contrast medium after 14th thoracic vertebra that represent spinal cord compression.