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Case Report

Sacral rib: an uncommon congenital anomaly

Naohisa Miyakoshi, MD, PhD^{a,*}, Akira Kobayashi, MD, PhD^b, Michio Hongo, MD, PhD^a, Yoichi Shimada, MD, PhD^a

^aDepartment of Orthopedic Surgery, Akita University Graduate School of Medicine, 1-1-1 Hondo, Akita 010-8543, Japan

^bHonobono-en Long-Term Care Health Facility, 92-1 Kaidoshita, Showa-Okubo, Katagami 018-1401, Japan

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Abstract

BACKGROUND CONTEXT: Sacral rib represents an uncommon pathology in which rib-like structures arise from the sacrum. Supernumerary ribs may occur at any level of the spine, but supernumerary ribs in the sacrococcygeal area are extremely rare.

PURPOSE: To present the case of a patient with sacral rib and to discuss this entity with reference to the literature.

STUDY DESIGN: A case report and literature review.

METHODS: A 17-year-old girl presented with low back pain and discomfort in bilateral gluteal regions. Radiographs and computed tomography (CT) of the pelvis showed a smooth-surfaced, rod-like bony structure attaching to the sacrum on the left side. The appearance was consistent with sacral rib. The sacrum was hypoplastic and deviated to the right. Magnetic resonance imaging (MRI) showed insertion of the gluteus maximus (GM) onto the coccyx only on the right side. The sacral rib existed beneath the left GM muscle and received a partial insertion from the left GM muscle. No ligamentous continuation between the sacral rib and coccyx was observed.

RESULTS: Conservative treatment relieved symptoms, so no surgical intervention was performed.

CONCLUSIONS: Sacral rib is a rare congenital anomaly for which surgical intervention is usually unnecessary. However, appropriate workups with CT and/or MRI should be considered for women, because sacral rib may cause complications during childbirth. In the literature, sacral/coccygeal rib is sometimes called “pelvic rib.” However, sacral/coccygeal rib should be distinguished from pelvic rib, because pelvic rib originating from the ilium is considered to represent a different entity. © 2013 Elsevier Inc. All rights reserved.

Keywords:

Congenital anomaly; Pelvic rib; Sacral rib; Supernumerary rib

Introduction

Supernumerary ribs may occur at any level of the spine, but are most commonly observed in the cervical and lumbar areas [1]. Among them, cervical rib is clinically important because of the potential clinical manifestations, including thoracic outlet syndrome.

Supernumerary ribs in the sacrococcygeal area are extremely rare. To the best of our knowledge, only nine cases of sacral rib and two cases of coccygeal rib have been described in a total of nine reports in the English literature

[1–9]. We present here an additional case of sacral rib and discuss this rare entity with reference to the literature. The present report shows for the first time in the English literature three-dimensional (3D) computed tomography (CT) of a sacral rib. Magnetic resonance imaging (MRI) also demonstrated the anatomical location of the sacral rib in the present case.

Case report

A 17-year-old girl with a history of brace therapy for adolescent idiopathic scoliosis presented with a 3-month history of low back pain and discomfort in bilateral gluteal regions. She did not recall any specific trauma. She noticed that symptoms exacerbated with long periods of sitting. Neurological examination showed no abnormalities with regard to deep tendon reflexes, both motor and sensory.

FDA device/drug status: Not applicable.

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* Corresponding author. Department of Orthopedic Surgery, Akita University Graduate School of Medicine, 1-1-1 Hondo, Akita 010-8543, Japan. Tel.: (81) 18-884-6148; fax: (81) 18-836-2617.

E-mail address: miyakosh@doc.med.akita-u.ac.jp (N. Miyakoshi).



Fig. 1. Anteroposterior radiograph of the pelvis showing a smooth, rod-like bony structure attached to the middle of the sacrum on the left side.

Ranges of motion for the spine and hip were normal and no pain was induced with motion. Routine radiographs of the spine revealed a right thoracic curve of 11° from T5 to L1 and a left lumbar curve from L1 to L5 measuring 6°. A radiograph of the pelvis revealed a smooth, rod-like bony structure attached to the middle of the sacrum on the left side (Fig. 1).

Confirmation of an additional bone on the left side of the sacrum was seen on 3D-CT of the pelvis (Fig. 2). This bony structure, creating a small joint with the sacrum, arose from the level of the 4th sacral vertebra and extended obliquely toward the left ischial spine, measuring approximately 55×25×15 mm in its largest dimensions. Given the results of 3D-CT, the bony structure was diagnosed as a sacral rib. In addition, 3D-CT revealed that the inferior half of the sacrum was hypoplastic and deviated laterally to the right side. MRI showed that the sacral rib existed beneath the left gluteus maximus (GM) muscle, showing a normal cortical lining and homogeneous signal intensity identical to the bone marrow of adjacent bones. Axial MRI at coccyx level showed insertion of the GM onto the coccyx only on the right side (Fig. 3). On the left side, the coccyx received no insertion from the GM, but the left side of the sacral rib received an insertion from part of the left GM. No ligamentous continuation was observed between the coccyx and sacral rib.

Because symptoms for this patient subsided with administration of a nonsteroidal anti-inflammatory drug, no surgical intervention was performed. At follow-up 2 years later, she remained symptom-free.

Discussion

In human and mammals, the ribs and vertebrae develop from a common primordial mass that passes through

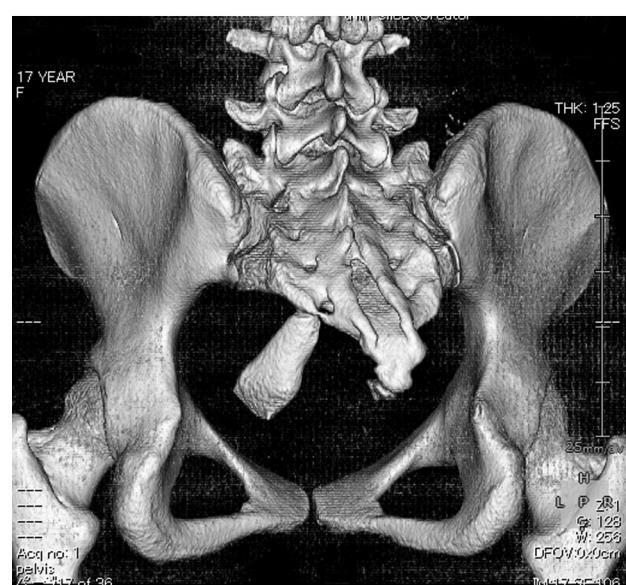
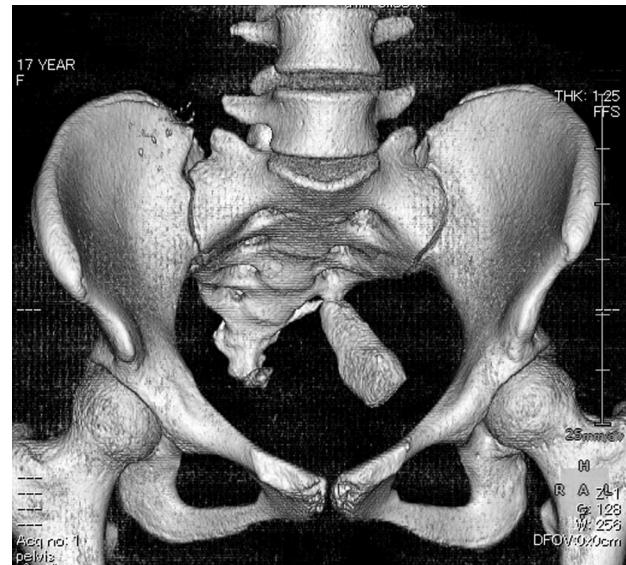


Fig. 2. Three-dimensional computed tomography of the pelvis. The sacral rib is attached to the sacrum on the left side with a small articulation, and the inferior half of the sacrum is hypoplastic and deviated to the right side. (Top) Anterior view; (Bottom) posterior view.

successive stages of membranous, chondrogenous, and osteogenous development [1,3,5]. During the earliest membranous stage, mesenchymal cells migrate toward the notochord to form the vertebral column anlage [1]. Chondrifications in parts of these blastemal masses are destined to become costal processes, neural arches, and ribs, but in all except the thoracic vertebrae, these structures ordinarily become fused with their respective vertebral centers by the seventh fetal week [1,5,8]. Any defect in fusion may cause the rib to keep growing and form a supernumerary rib [3]. Sacral ribs develop as a consequence of failure of the rib primordium to fuse with the vertebral centers of the sacrum [1,2,7].

Two-thirds of reported cases (8 of 12) with sacral/coccygeal ribs, including the present case, involved females, and



Fig. 3. Axial T1-weighted magnetic resonance imaging at the level of the coccyx. Gluteus maximus (GM) muscle (*) inserts onto the coccyx (arrowhead) only on the right side. The sacral rib (arrow) is located beneath the left GM muscle, which shows partial insertion onto the left side edge of the sacral rib. No ligamentous continuation between the coccyx and sacral rib is observed.

the average age at diagnosis was 22.2 years (range, 3–55 years) [1–9]. Associated anomalies were present in some cases, including congenital bowing of the femurs [3], mild hip dysplasia [4], talipes calcaneovalgus [4], caudal regression syndrome characterized by incomplete development of terminal spinal segments [7], and scoliosis [2]. As Mares et al. [4] speculated, although the sacral/coccygeal ribs seem to have no direct association with these anomalies, mild skeletal insult in the fetal period may play some etiological role.

Supernumerary ribs in the sacrococcygeal area are usually diagnosed incidentally on radiographs taken for different reasons [4,7]. Because patients typically appear largely asymptomatic, surgery is not usually indicated. However, in the case report by Heligman et al. [2], a 14-year-old girl underwent partial excision of the sacral rib to improve the range of motion of the right hip because the elongated sacral rib reached to the greater trochanter of the hip and restricted the range of motion. Sullivan and Cornwell [8] also resected a sacral rib from a 15-year-old girl to prevent possible obstetric problems in the future. The histology confirmed a normal rib-like structure with intact periosteum, well-formed cortex, cancellous bone in a medullary cavity filled with bone marrow tissue, and cartilaginous tissue at the distal portion [8].

Sacrococcygeal ribs may actually cause complications during delivery [3,8], potentially compromising the birth canal in females [8]. Indeed, Kaushal et al. [3] reported the case of a 34-year-old woman with a sacral rib who underwent two cesarean sections for fetopelvic disproportion. Appropriate workups with CT and/or MRI thus seem very important for women to consider potential surgery relating

to delivery. In the present case, 3D-CT showed the sacral rib clearly located within the pelvic ring, so future delivery might be affected. However, preventive resection of the sacral rib in the present case would have carried a risk of gluteal muscle weakness, because MRI showed the part of the GM inserting onto the sacral rib. If any complications during delivery are anticipated, cesarean section seems likely to represent a better option for such cases.

Differential diagnoses for sacral/coccygeal rib include calcified pelvic ligaments [1,7], posttraumatic mineralization (myositis ossificans) [1,7], sacral and chronic avulsion fractures [7], and osteochondroma [6,10]. However, compared with these lesions, sacral/coccygeal ribs are usually asymptomatic with no history of trauma. In addition, imaging features of calcified ligaments, myositis ossificans, fractures, and osteochondroma differ from the imaging feature of sacral/coccygeal rib, so diagnosis seems unlikely to be difficult. However, knowledge of this asymptomatic variant is important for identification.

In the literature, several authors have reported sacrococcygeal ribs as “pelvic ribs,” probably because the sacrum is a component of the pelvic ring [8,9]. However, we suggest that the term sacral/coccygeal rib be clearly distinguished from pelvic rib, because the term pelvic rib is also used to describe a bony protuberance from the ilium [11]. We agree with Shah et al. [7] that most pelvic ribs originating from the ilium are probably not true congenital ribs and probably represent myositis ossificans. Lame [12] reported a case with a finger-shaped bony structure arising from the iliac crest, terming this anomaly an “iliac rib.” This term appears suitable for distinguishing a bony protuberance from the ilium from a sacral/coccygeal rib.

Conclusions

This case report describes a rare case of a supernumerary rib in the sacrococcygeal area. The present report includes the first description in the English literature of 3D-CT of a sacral rib. MRI also demonstrated the anatomical location of the sacral rib in the present case. This rare congenital anomaly has several differential diagnoses, so knowledge to identify this asymptomatic variant is important. The terms “sacral/coccygeal rib” and “pelvic rib” should be differentiated, because the origins and natures of these entities are thought to differ.

References

- [1] Dunaway CL, Williams JP, Brogdon BG. Case report 222. Sacral and coccygeal supernumerary ribs (pelvic ribs). *Skeletal Radiol* 1983;9: 212–4.
- [2] Heligman D, Sullivan RC, Millar EA. Sacral ribs. A case report. *Orthopedics* 1987;10:1439–42.
- [3] Kaushal SP. Sacral ribs. *Int Surg* 1977;62:37–8.
- [4] Mares AJ, Bar-Ziv J. Sacral (pelvic) “rib” or coccygeal “duplication”? *Z Kinderchir* 1981;34:72–6.

- [5] Pais J, Levine A, Pais SO. Coccygeal ribs: development and appearance in two cases. *AJR Am J Roentgenol* 1978;131:164–6.
- [6] Rashid M, Khalid M, Malik N. Sacral rib: a rare congenital anomaly. *Acta Orthop Belg* 2008;74:429–31.
- [7] Shah JR, Sainani N, Patkar DP. Caudal regression syndrome with sacral rib: MRI features. *Acta Radiol* 2006;47:862–4.
- [8] Sullivan D, Cornwell WS. Pelvic rib. Report of a case. *Radiology* 1974;110:355–7.
- [9] Van Derslice R, Gembala R, Zekavat PP. Case report. Pelvic rib/digit. *Spine* 1992;17:1264–6.
- [10] Chin KR, Kim JM. A rare anterior sacral osteochondroma presenting as sciatica in an adult: a case report and review of the literature. *Spine J* 2010;10:e1–4.
- [11] Maegele M. Pelvic digit as a rare cause of chronic hip pain and functional impairment: a case report and review of the literature. *J Med Case Rep* 2009;3:139.
- [12] Lame EL. Case report 32. *Skeletal Radiol* 1977;2:47–8.