Case report
Bifid rib with fused vertebrae - A rare abnormality of the skeletal system: A case report

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ABSTRACT

Introduction: Bifid rib as pathoanatomical findings on chest X-ray is a skeletal disorder. It is usually unilateral and commonly found in males. Bifid ribs commonly happen in absence of structural defects of the vertebrae.

Presentation of case: The case of this report is a 65-year-old with severe infection of Covid 19. He was admitted to the ICU. Imaging findings indicate existence of a bifid rib with vertebral fusion.

Discussion: Bifurcated ribs usually have been described without vertebral defects. In our case, there is a bifurcated rib with fused vertebrae. These defects are also defined in the criteria of Gorlin syndrome.

Conclusion: Unlike other rib abnormalities, the bifid rib occurs mainly in the absence of a vertebral defect, but this study reports a bifid rib with vertebral defects.

1. Introduction

1.1. Congenital skeletal abnormalities

The ribs as a part of the axial skeleton originate from the somites during embryonic development. Somites are located at sides of the neural tube [1]. Numerical and structural anomalies of the ribs are observed in approximately 2% of people [2,3]. The occurrence of congenital rib abnormalities are mainly associated with structural changes. Cervical and bifid ribs are considered in the group of structural abnormalities [1,2,4]. The prevalence of bifid ribs is between 0.15 and 3.4% of people [5]. Unlike other rib abnormalities, the bifid rib occurs mainly in the absence of a vertebral defect [3]. However, S. Zeeshan, et al. reported the bifid rib with vertebral defect [6].

1.2. Gorlin syndrome

Gorlin-Goltz Syndrome is known as Basal Nevus Carcinoma Syndrome (NBCCS). It is a rare multisystem disease with autosomal dominant inheritance pattern [7]. The main clinical manifestations include several basal cell carcinomas (BCC), Odontogenic maxillary keratocysts, palmar and plantar hyperkeratosis, skeletal abnormalities, abnormal intracranial calcification and facial deformity (macrocephaly, cleft lip, palate and severe eye abnormalities) [8]. Existing skeletal abnormalities include protrusion of the frontal or parietal area and dilated nasal root, hypertelorism, lateral displacement of the inner corner of the eye and mandibular protrusion [9,10]. Other skeletal abnormalities include bifid ribs (more than one rib unilaterally or bilaterally), rib synostosis, kyphoscoliosis, vertebral fusion, cervical rib, spina bifida, shortened fourth metacarpal bone (Albright mark) and palmar and plantar pits [11,8]. Kimonis et al. explain some criteria for detecting of Gorlin syndrome that include at least 2 major criteria or 1 major criteria and 2 minor criteria [12].

2. Case report

A 65-year-old man, married, childless with symptoms of cough, shortness of breath and severe pneumonia Covid 19 had referred to Shahid Sattari Hospital in Qarchak (Tehran). His past medical history (PMH) refers to diabetes and high blood pressure. Various tests are performed for blood factors associated with Covid 19. PCR test result was positive. The patient also underwent computed tomography (CT). Then he admitted to the intensive care unit due to severe involvement of both lungs in pneumonia Covid 19 and the severity of symptoms

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By three-dimensional reconstruction of CT scan, the right fourth rib was identified as a bifid rib (Figs. 1a, 2a-c). Anatomically, the bifurcation site begins in the middle of the rib’s shaft. Bifid rib had connected to the sternum through two rib cartilages. The fusion of the thoracic vertebral bodies (4 and 5th) was another skeletal deformity of this case. In the reconstruction of the thoracic vertebra, we observed the fusion of the vertebrae spinal processes (4 and 5th) too (Fig. 2a-c). Unfortunately, the patient died due to severe lung involvement.

3. Discussion

Similar to most previous studies, our reported case shows bifid rib on the right side. Bifid ribs are more common on the right side [13]. Our case had a completely fused T4 and T5 vertebral bodies. A similar study in 2020 shows bifid rib with partially fused T2 and T3 vertebral bodies [6]. In addition, in our report, bifid rib is along with fused T4 and T5 spinal processes. The presence of a bifid rib in imaging is usually asymptomatic and does not require special intervention, although it may appear as a mass in the chest wall [5]. However, identifying of this rib uses for differential diagnosis such as to find out Gorlin syndrome. Despite recent advances in the molecular diagnosis of congenital anomalies, due to the high cost of genetic and molecular tests, the diagnosis of these anomalies often relies on clinical and radiological criteria [14]. Our case had a bifid rib and fused vertebrae as a major diagnostic criteria for Probability of Gorlin syndrome diagnosis, but because of his death, no definitive diagnosis can be made. Of course according to the patient’s statements related to history of his skin disorders, the patient did not have multiple basal cell carcinoma, so our study is just a case report of a rare rib anomaly with vertebrae deformity. These deformities cannot be easily ruled out, because in some cases, the symptoms of the syndrome are hidden. Earlier detection in younger ages can improve the prognosis of patients with Gorlin syndrome and it is possible to prevent the inherited distribution of this disease with an autosomal dominant inheritance pattern in society. This work has been reported in line with the SCARE 2020 criteria [15].

Source of funding

None.

Ethical approval

This report is based on CT scan images of the patient and he agreed to report the case.

![Fig. 1. Coronal reconstruction from chest CT scan. A: Both lungs are affected by coronavirus. Note the location of the rib's bifurcation, B: fusion of spinal processes (arrow) C: fusion of vertebral body (circle).](image)
Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

**Consent**

**Author contribution**

1. Soheil mirzaei - Conception and design, Acquisition of data, Writing the paper, Final approval of the version to be published.
2. Dr. Sare moslemi – Imaging report, Critical revision of the article, Final approval of the version to be published.
3. Dr. Faride shafeghat - Acquisition of data, Critical revision of the article, Final approval of the version to be published.
4. Dr. Sana karimi - Writing the paper, Final approval of the version to be published.
5. Dr. Mina eftekharzadeh - Conception and design, Writing the paper, Final approval of the version to be published.

**Registration of research studies**

Not applicable.

**Guarantor**

Not applicable.

**Declaration of competing interest**

None.

Fig. 2. Chest CT scan - Three-dimensional reconstruction of a bone A: lateral view of bifurcated rib (circle) – fused vertebral prosses (arrow), B: anterior view of bifurcated rib, C: fused vertebral body.
References

[1] M. Mallo, T. Vinagre, M. Carapuço, The road to the vertebral formula, Int. J. Dev. Biol. 53 (8–10) (2009) 1469–1481.
[2] A. Castriota-Scanderbeg, B. Dallapiccola, in: Abnormal Skeletal Phenotypes: From Simple Signs to Complex Diagnoses, Springer, New York, 2005, pp. 124–144.
[3] D. Wattanasirichaigoon, C. Prasad, G. Schneider, et al., Rib defects in patterns of multiple malformations: a retrospective review and phenotypic analysis of 47 cases, Am. J. Med. Genet. A 122A (1) (2003) 63–69.
[4] T.R. Yochum, L.J. Rowe, in: Essentials of Skeletal Radiology, third edition 291-293, Lippincott/Williams & Wilkins, Philadelphia, 2005, pp. 319–324.
[5] H. Kaneko, H. Kitt, A. Mahuchi, et al., Isolated bifid rib: clinical and radiological findings in children, Pediatr. Int. 54 (6) (2012) 820–823.
[6] S. Zeeshan, S.N.F. Hussain, Z. Mughal, S.S.M. Anwar, S.N. Naem, Bifurcated rib with vertebral defects - a rare anatomical variant: case report with literature review, Int. J. Surg. Case Rep. 67 (2020) 203–206.
[7] A.R. Casaroto, D.C. Loures, E. Moreshchi, V.C. Velrini, C.L. Trento, V.D. Gottardo, V.S. Lara, Early diagnosis of Gorlin-Goltz syndrome, case report, Head Face Med 7 (2011) 2. Jan 25.
[8] Muzio L. Lo, Nevoid basal cell carcinoma syndrome (Gorlin syndrome), Orphanet J. Rare Dis. 3 (2008) 22.
[9] T. Pindborg, Multiple basal cell nevi, odontogenic keratocyst and skeletal anomalies. In Syndrome of Head and Neck, (1st ed.), 400-407.
[10] B.W. Neville, D.D. Damm, C.M. Allen, J.E. Bouquot, in: Oral and Maxillofacial Pathology, 1st ed., W.B. SaundersCompany, 1995, pp. 501–503.
[11] K.S. Kartsiga, B.Sivapatha Sundraraj, R. Manikandan, Neviod bascal cell carcinoma syndrome, Indian J. Dent. Res. 17 (1) (2006) 50–53. Jan-Mar.
[12] V.E. Kimonis, A. Goldstein, B. Pastakia, et al., Clinical manifestations in 105 persons with nevoid basal cell carcinoma syndrome, Am. J. Med. Genet. 69 (3) (1997) 299–308.
[13] A. Tirpude, M. Gaikwad, A. Haldar, Occurrence of right third bifid rib in a 60-year-old male cadaver: serendipic or syndromic? J. Anat. Soc. India (2018).
[14] F.M. Vanhoenacker, W. Van Hul, J. Gielen, AM.De Schepper, Congenital skeletal abnormalities, an introduction to the radiological semiology, Eur. J. Radiol. 40 (3) (2001) 168–183. Dec.
[15] for the SCARE Group, R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, The SCARE 2020 guideline: updating consensus Surgical Case Report (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.