Bifid Rib in a male cadaver: Serendipic or Syndromic

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Abstract

Bifid rib presents usually as an isolated and asymptomatic finding on x-rays and cadaveric dissections. This may be due to atypical process of segmentation of developing somites. Sometimes it may occur with other malformations. Aim of the case report presented here was to give a description of the characteristics of bifid rib and to look for any associated abnormalities. During routine dissection, bifid rib was found in thoraco-abdominal region of a male cadaver. The dimensions of the bifid rib were measured with digital vernier calliper of resolution 0.01 mm and photographed using a digital camera. Survey of other regions of the cadaver was done by dissection. Bifurcation of anterior part of right third rib enclosing an additional intercostal space was found. Follow-up dissection in other regions of the cadaver revealed characteristics of Crohn's disease of jejunum, ileum, and transverse colon and signs of dysmorphism of both the kidneys. The knowledge of such variations will be of use to the clinicians, radiologist and anatomist to investigate any incidental bifid rib finding.

Key words: bifid rib, lobulations, strictures, pax genes

Introduction

Ribs aid in interpretation of radiological image and are used as an essential landmark during physical examination. Knowledge of underlying muscles, and neurovascular anatomy is applicable for planning surgical and percutaneous procedures. Ribs develop as an elongation of costal processes of thoracic vertebra that are derived from somites through the process of endochondral ossification. Anatomical variations of ribs can be structural or numerical. This includes developmental deformities, cervical rib, short rib, hypoplasia, fibrosed costal cartilage with adjacent rib, complete fusion, bridging, intrathoracic rib and bifurcation of rib. Bifid rib or forked rib also called as sternum bifidum is a cleavage of either cartilaginous or bony or both parts of rib encircling a small inter costal space. It is usually an accidental, sporadic, asymptomatic and unilateral cadaveric or radiological finding. The overall prevalence of bifid rib is 0.15% to 3.4% and it accounts for up to 20% of all congenital anomalies.

Bifid ribs are more common on the right side. It is more common in males than females and are located most frequently along 3rd and 4th rib rather than second, fifth and sixth ribs respectively. Bifid rib may be associated with several genetic disorders such as basal cell nevus syndrome, Jobs syndrome and Kindler syndrome. It may be associated with abnormalities of vertebral segmentation, congenital anomalies or part of malformation such as VACTERL [vertebral, anal, cardiovascular, trachea-oesophageal, renal and limb defects without other specific diagnosis], MURCS [Mullerian, renal and cervico-thoracic somitic abnormalities] and contiguous polytopic developmental defects such as acrogenal, cardiomeglic and gastromelic. Because ribs are mesodermal in origin, it is not surprising that costal defects are associated with malformations in other organs which are derived from mesoderm such as heart and kidney. A case of unilateral bifid rib in a 60-year-old male cadaver with a detailed examination is presented here to create awareness among clinicians regarding this condition and associated anomalies.

Case Report

During routine dissection of sixty-year old male cadaver allotted to first year medical students a bifid rib
was noticed in the anterior aspect of the third rib on the right side. Using a digital vernier calliper with a reading sensitivity of 0.01 mm, the dimensions of bifid rib and area enclosed were measured.

Observations

Both chondral, osseous component and costochondral joints were duplicated leading to the creation of an additional elliptical intercostal space [ICS] [Fig. 1]. The reunion of bifurcation was at 43.5 mm from the sternal end. The breadth of upper and lower division was 12.2 mm and 12.3 mm respectively. The bifurcation encroached and narrowed the anterior second ICS whereas anterior third ICS was slightly widened. A thin fibrous layer representing external intercostal lamina covered this additional ICS. Beneath the fibrous layer, muscle fibres running down and laterally representing internal intercostal muscle were visible. Posteriorly fibres of sternocostalis muscle were extending over this region. The elliptical space was supplied by second anterior intercostal artery branching from the internal thoracic artery. The venous distribution was corresponding to that of the arterial supply. The third intercostal nerve ran along the costal groove of the lower division, but no collateral branch ran along the upper division. A small twig from third intercostal nerve was supplying the elliptical ICS. Remaining part of 3rd rib was normal.

It was found that the greater length of jejunum, ileum and colon was involved in discontinuous transmural shortening [skip areas] indicating features of Crohn's disease [Figs. 2]. Terminal part of ileum extended from right lower quadrant to right upper quadrant to form ileocolic junction. Caecum was found conical, shrunken, and in sub-hepatic region. Ascending colon was very short. The root of the mesentery which normally runs from duodeno-jejuno-flexure to right lower quadrant extended to subhepatic caecum. Among the retroperitoneal structures it was noticed that the kidneys were placed normally but had features of lobar dysmorphism. Sharply defined linear remnants of interlobar groove and multiple indentations were visible on external surfaces of kidneys [Fig. 3]. A prominent hump on lateral border of left kidney was seen [Fig. 3]. The vertical section of right kidney revealed features of persistent [hypertrophied] column of Bertini. The column of renal cortical tissue was thick, hypertrophic and protruding into the renal sinus [Fig. 4].

Discussion

Similar to earlier reports in literature bifurcation was observed on the third rib of right side. The distance from lower division to the second rib was similar to width of the lower intercostal spaces. Therefore, we are of the opinion that upper division is abnormal. The intercostal muscle was present in the elliptical space of the bifid rib, and might not be present if the upper division divides from one rib into two divisions rather than growing out of the lower division. The source of blood supply to bifid space [second anterior intercostal vessels] and pattern of innervation indicates that the muscle in bifid space originated from muscles of the second intercostal space, and separated from each other by the upper division. It was difficult to determine whether the upper division arose from the lateral side to medial side or medial to lateral side.

Though the incidence of costal abnormalities evident on X-rays' range from 0.15 % to 2.8 %, the ethnic prevalence of bifid rib have not been documented satisfactorily and reports from the Indian subcontinent are sporadic.

Various morphological patterns of bifid ribs are documented in literature. The anomalous bifid rib pattern reported here has been identified as 'fork or slender' and 'type II' as per Kamano et al and Wu Chul Song et al respectively. Not only the proximal and distal part of a rib and their junctions develop from lateral plate mesoderm, surface ectoderm and their interdependent complex interactions but are controlled by Pax 1 and Pax 3 genes respectively. The down regulation of Pax 3 genes along with mutation in ptch genes may affect the mesodermal and ectodermal interaction necessary for normal development of distal rib. This may result in abnormal development of the dermomyotome of concerned rib which may provide a rationale to the altered morphology of bifid rib.
Authors are of the opinion that the renal cortical dysmorphic features represent persistent fetal lobulations, dromedary hump and hypertrophic column of Bertin that are normal anatomical variations in this case. Lobulation are found in approximately 5% of adults submitted to imaging and may simulate renal mass lesion or renal scar on cross sectional imaging in otherwise healthy parenchyma.\textsuperscript{17,18} During first and second trimester, fetal kidney presents with well-defined cortical sulci on the outer surface. It becomes smooth and interlobar grooves become invisible in the third trimester because of growth and increase in number of nephrons.\textsuperscript{19} Some authors have suggested the grooves appear because of partial fusion of embryonic parenchymatosus masses called ranunculi.\textsuperscript{20} The dromedary hump kidney is characterized by a change in the shape and contour of the posterolateral aspect of the left kidney, because of a focal prominence of the renal parenchyma, probably due to impression of the spleen during fetal life.\textsuperscript{17,19} Crohn disease [CD] is an idiopathic, chronic regional enteritis that most commonly affects the terminal ileum but has the potential to affect any part of the gastrointestinal tract [of endodermal origin] from mouth to anus. Authors found multiple strictures in small intestine and large intestine. Migration of caecum to sub-hepatic region may be due to contraction of mesocolon.\textsuperscript{21} It is arduous to link Crohn's disease to bifid rib and renal morphological variants [both mesodermal in origin]. But it can be explained, based on the concept of developmental field. Developmental field is defined as "a dysmorphogenetically reactive unit, i.e., a set of embryonic primordia that reacted identically to different dysmorphogenetic causes".\textsuperscript{22} Thus, a developmental field is a part of the embryo that reacts as a temporally and spatially coordinated unit to localized forces of the organization and differentiation.\textsuperscript{23} Disturbance of the developmental field complex by whatever cause can lead to multiple, usually contiguous, but sometimes distantly located anomalies. More recently, the term polytopic developmental field defect has been used to describe patterns of malformation where two or more progenitor fields are involved, such as acrorenal, cardiomeclic, gastromeclic, etc.\textsuperscript{23} Rib anomalies would fit
well into a variety of polytopic developmental defects and indeed have been found in relatively high frequency with acrorenal field defects and with aberrant bronchi and cardiovascular defects. As features of coinciding syndromes were absent and authors preclude any syndromic involvement of bifid rib in the present case.

**Conclusion**

The authors assess this serendipic cadaveric dissection discovery of right third bifid rib and dysmorphic renal silhouette as an event of audited *Pax* gene expression tendering altered tissue interaction association in the orientation of rib development with an unexplained gastromelic component. Bifid rib occurrence on imaging warrants a further prophylactic clinical examination and differential diagnostic evaluation and to exclude other mesodermal and polytopic anomalies. To the authors' knowledge, this exhibit is an unreported finding but enlightening for a morphologist and an anatomist.

**References**


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