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Clinical relevance of the bifid ribs in humans

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Abstract: Bifid ribs are a congenital anomaly that in the majority of cases does not demonstrate clinical symptoms. Nevertheless, their presence may be linked to several genetic disorders that affect different systems of the human body. Symptomatic conditions of bifid ribs can appear as follows: chest deformity and pain, breathing difficulties due to the structural changes in the thoracic wall, and neurological complaints caused by compression of the intercostal nerves.
Hereby, we presented a concise review of clinical reports documenting symptomatic cases of bifid ribs and their potential associations with systemic disorders. This article also examines cases of bifid ribs encountered in the clinical practice of plastic surgeons, pediatric surgeons, and cardiothoracic surgeons.

Keywords: rib abnormalities, bifurcated ribs, thoracic cage, Gorlin-Goltz syndrome.

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Introduction

A bifid rib is a skeletal anomaly characterized by the division of the sternal end of the rib into two distinct bony parts. The ribs usually bifurcate at an angle of about 60°, most commonly at a distance of 3 to 4 cm from their sternal end [1]. This anatomical feature distinguishes the bifid ribs from normal ribs, as demonstrated in Fig. 1. The division of a rib into two distinct branches typically results from disturbances during fetal development, which lead to abnormal rib formation [2]. Thus, their way of articulation with the sternum depends on the configuration of the sternal end. The bifid ribs can be joined to the sternum separately by their own costal cartilages or jointly via a single cartilage. This anomaly has been recorded in human skeletons from different historical periods, including the Neolithic era, and is still observed in present-day populations [3–5].

Typically, the prevalence of bifid ribs ranges from 0.15% to 3.4%; however, in geographically isolated populations, the frequency of bifid ribs can be higher (up to 8.4%), as observed among inhabitants of the Samoa Islands [6]. Bifid ribs are generally more prevalent in males compared to

females, although studies have shown inconsistent results regarding sex-related dependencies [1]. According to Oner *et al.*, there is a greater occurrence of bifid ribs in females, especially on the right side [7].

The bifid ribs account for approximately 20% of all congenital rib anomalies, demonstrating various anatomical configurations that can cause clinical symptoms or remain asymptomatic [8]. For this reason, the bifid ribs have drawn the attention of researchers attempting to associate this anomaly with various clinical conditions. Nevertheless, the existing literature on symptomatic bifid ribs remains limited. Therefore, the aim of this study was to analyze scientific reports on the clinical implications of bifid ribs and their association with skeletal abnormalities and other diseases.

Fig. 1. A bifid rib alongside a normal rib seen in superior projection. The specimens derive from the museum collection of the Department of Anatomy, Jagiellonian University Medical College.

Anatomical patterns of the bifid ribs

As it was previously mentioned, the bifid rib is characterized by the division of the anterior end, which articulates with the sternum. The two separate branches of the bifurcated rib are usually connected to the sternum via their individual costal cartilage. Occasionally, the costal cartilages can fuse, leading to the formation of a singular segment that is attached to the sternum (Fig. 2).

The existence of a single bifid rib or multiple bifid ribs is classified as a congenital skeletal anomaly. Such anomalous ribs can take on various forms, including bifurcated, fused, hypoplastic, forked, or bridging configurations [9]. As a result, bifid ribs may exhibit different appearances due to surface irregularities or widening, along with varying placements in the thoracic region. Furthermore, costal anomalies may involve intrathoracic ribs that can also be bifid, although they occur relatively rarely [8, 10].

The pattern of bipartition of the sternal end of the rib can exhibit various configurations, depending on the symmetry of the branches (similar or different in size), bifurcation shape (U-shaped or V-shaped), and depth of notch between branches of the bifid rib (flat, shallow, deep). This classification of bifid ribs has been proposed by Mietlińska-Sauter *et al.* [11].

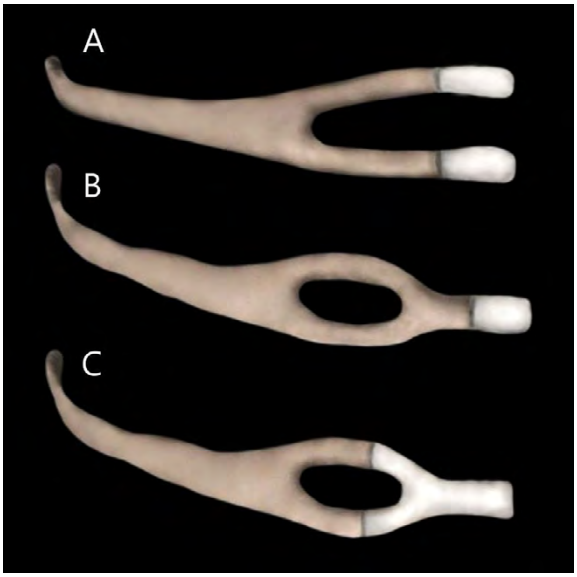

Fig. 2. Schematic representation of various sternal ends of bifid ribs, depending on the configuration of the bifurcation. A: forked type — both divisions of the rib have their own costal cartilage; B: hole type — both divisions jointly form a bony segment terminating with a single costal cartilage; C: a variant of the hole type in which the costal cartilages from both divisions converge to create a single cartilage.

Uncommon formation of the additional intercostal spaces can result from rib bifurcation.

Kumar *et al.* identified a case involving the left 3rd rib and its associated costal cartilage, which exhibited bifurcation. This bifurcation resulted in the formation of upper and lower divisions that enclosed a small circular intercostal space located between the 2nd and 3rd intercostal spaces [12].

A similar case was described by Bagoji *et al.*, who found that the bifurcated ends of the right 3rd rib and its costal cartilage were joined together to form an oval additional intercostal space [13]. Song *et al.* reported a case where two cartilaginous segments of the bifurcated 4th rib rejoined and attached to the sternum [9]. Thus, the upper intercostal space situated between the upper division and the 3rd rib exhibited a narrowing. Also, Nayak identified in an adult male a bifurcation of the right fourth rib and its costal cartilage, which ends joined with one another to surround an additional intercostal space about 8 cm long [14]. In turn, Osawa *et al.* noted that the costal cartilage of the left 5th rib divides into upper and lower segments, which subsequently reunite to create common segment [15].

The bifid ribs usually occur unilaterally, being frequently an asymptomatic lesion, whereas multiple bifid ribs existing unilaterally or bilaterally have been reported as extremely rare cases. The incidence of bifid ribs is observed in the following order: 3rd, 4th, 5th, 6th, and 2nd.

In a study involving 40,000 chest radiographs, Etter found that most of the bifurcated ribs existed on the right side of the thorax, with the predominance of the 4th rib, which bifurcated in 60 cases, whereas on the left side it was in 33 cases. The lowest numbers (5 and 8 cases) of the bifurcation demonstrated the 2nd and 7th ribs on both sides of the thorax. Bilaterally bifid ribs (3rd and 4th) were observed only in 7 cases versus 250 cases of unilateral bifurcation [16].

Further research rarely documented bilateral occurrence of bifid ribs. Patil *et al.* identified on the chest radiograph taken of a 21-year-old female bilaterally bifid ribs: right 6th and left 7th ribs [17]. Dhana *et al.* identified bifidity in the 2nd rib on the right side, the 4th rib on the right side, and the 3rd rib on the left side [18].

Clinical relevance of the bifid rib

Bifid ribs may occur in isolation or in conjunction with genetic syndromes. They are commonly associated with the Gorlin-Goltz syndrome (nevoid basal cell carcinoma syndrome) affecting multiple organs, including bones, due to the fact that approximately 60–70% of patients manifest such an anomaly [19, 20].

Occasionally, bifid ribs are encountered as a part of Jobs syndrome (Hyper-IgE syndrome — high level of immunoglobulin E and recurrent infections) and Kindler syndrome (a rare congenital disease of the skin; a subtype of inherited epidermolysis bullosa being a rare type of genodermatosis) [21]. In turn, multiple bifid ribs were identified in a patient with a possible diagnosis of Teebi syndrome (a rare genetic disorder characterized by hypertelorism and facial features that are similar to those observed in craniofrontonasal dysplasia) [8].

As mentioned, bifid ribs occur in congenital syndromes, which include a spectrum of skeletal malformations; hence, the bifid ribs can be associated with vertebral defects [22].

Koutnik *et al.* reported a case of several bifid ribs accompanied by vertebral anomalies and other abnormal conditions such as bridging of the sella turcica and calcification of the falx cerebri [23].

The presence of bifid ribs can be a sign of predisposition to the formation of malignancies. Previous research indicated that rib abnormalities have been linked to an increased risk of cancers occurring in children [24, 25]. Schumacher *et al.* found that children affected by neuroblastoma have a higher incidence of bifid ribs (4.5%) compared to the general population [26].

Other clinical implications of bifid ribs may involve the local deformities of the chest wall and their potential to contribute to thoracic trauma. The presence of a bifid rib may cause local pain or discomfort in the region where there is palpable alteration on the external surface of the thorax; however, this symptom does not always occur [27, 28].

De Olivera Alvarenga and Duarte identified radiographically a bifid rib of the 7th costal arch, which deformed the left side of the costal arch of the thorax [29]. In turn, Kok and Janssen reported the case of a chest deformity caused by a bifid rib observed in a 15-year-old boy with a swelling located on the right side of his thorax [30], and Wong *et al.* described asymptomatic focal chest wall bulging caused by the presence of bifid ribs examined at a group of children with a median age of 5 years [31]. Donnelly *et al.* reported a singular case of a bifid rib that was responsible for the lesion found on the child's chest wall [32].

The division of the distal shaft of the bifid rib incorporated in the thorax may considerably affect the size of the intercostal spaces; hence, the upper intercostal space can be narrowed, whereas the lower intercostal space will be widened. Such alteration of intercostal spaces can restrict movement of the ribs and cause a traumatic effect on the neurovascular structures running within the intercostal space, either affecting the function of the intercostal musculature. Nieuwstraten and Huijstee reported cases of symptomatic bifid ribs, which caused compression of the intercostal nerves, presenting as chronic chest pain [33].

A specific bilateral fusion between the cervical ribs and the first ribs formed bifid ribs, causing right-sided thoracic outlet syndrome in a 13-year-old girl, as described by Cağli *et al.* [34].

During a radiographic examination of two children, Kaneko *et al.* identified downward extension of the cervical rib or first rib, which articulated with the upper branch of the bifid 1st or 2nd rib [35]. Noteworthy, cervical ribs, similarly to bifid ribs are developmental skeletal anomalies related to the sonic hedgehog signaling pathway controlling proper organ development and patterning during embryogenesis [36, 37].

Particularly dangerous are cases of intrathoracic bifid ribs, which are characterized by a bony protrusion of a rib extending into the thoracic cavity. Their presence can be a reason for serious clinical disorders. Such ribs can demonstrate different anatomical variants; therefore, only a few configurations can cause clinical symptoms [38]. The traumatic effect of intrathoracic ribs depends on their size and position in the thoracic cavity. Kamano *et al.* reported a case of a bifid intrathoracic rib arising from the anterior-lateral portion of a depressed 4th rib found on chest radiography of a 21-year-old woman. In spite of this costal anomaly, the lung was intact [10]. In contrast, Ovaere *et al.* reported a case of a 36-year-old male in whom a bifid rib caused perforation of the lung after lifting a heavy object [39].

Another clinical aspect associated with bifid ribs was emphasized by Molina *et al.* [40]. These researchers indicated that the bifid ribs encountered during breast reconstruction operations may create complications in this procedure. The bifurcation primarily affects the 3rd and 4th ribs, which are situated in the region potentially designated for breast reconstruction [41]. Possible narrowing of the intercostal space can restrict easy access to the internal thoracic (internal mammary) vessels used for anastomoses. Thus, surgery in this region may result in chronic intercostal neuralgia [42]. Additionally, bifid ribs may be supplied by perforating blood vessels that may arise from the internal thoracic artery (internal mammary artery). Thus, the presence of bid ribs may be challenging for plastic surgeons not only to attain the proper shape and position of the reconstructed breast, but also to prevent injury to the recipient blood vessels, particularly the perforators of the internal thoracic vessels, which are utilized for free flap anastomosis during autologous breast reconstruction after mastectomy [43]. To conclude, the presence of bifid ribs can complicate breast reconstruction procedures; nonetheless, appropriate preoperative preparation and adjustment of the surgical technique allow for successful surgery.

Furthermore, rib malformations can simulate pathologic conditions that may manifest in the thoracic skeleton as the result of various diseases. The bifid ribs detected through palpation may be erroneously interpreted as a malignancy or a fracture of the costal cartilage. Guttentag and Salwen discussed a diverse range of pathologies that can influence the appearance of ribs in imaging diagnostics [44]. Notably, in the case of bifid ribs, chondroid lesions are of particular importance due to their tendency to occur near the anterior end of the rib.

We would like to pay special attention to cases of bifid ribs in the practice of cardiothoracic surgeons and children surgeons. Bifid ribs can pose an additional risk in some surgical procedures, e.g., surgical closure of a patent ductus arteriosus via left thoracotomy, which has been a standard treatment for this congenital heart defect [45]. The fact that thoracotomy is performed in an area where there is a higher likelihood of encountering bifid ribs (4th–5th intercostal spaces) means this procedure may be associated with complications, such as an increased risk of rib fractures and an increased risk of damage to intercostal vessels. There may also be a problem with the insertion of the rib spreader, which may result in lack of access to the operated area.

A similar problem can be encountered in pediatric surgery in the case of esophageal atresia surgery. The most performed incision during the operation is a right posterolateral thoracotomy. It is generally recommended to enter the chest cavity through the 4th intercostal space [46]. Thus, there is an even greater risk of encountering a bifid rib during the operation.

In some cases, anomalies of the 4th and 5th ribs have been successfully identified via USG, demonstrating the technique's reliability in delineating the costal cartilage bifurcation. Kryger *et al.* described that ultrasonography (USG) has proven to be a useful diagnostic tool for identifying bifid

ribs. According to these authors, such a diagnostic method is especially beneficial for children, as it eliminates the radiation exposure associated with X-ray imaging [47].

The costal cartilage serves commonly as a graft material used in the reconstruction of the ear, particularly for conditions like microtia [48, 49], as well as in the correction of post-traumatic deformities of the auricle, nasal deformities, and laryngotracheal defects. Due to widespread usage of costal cartilage in surgical reconstructions, we would like to put under consideration whether cartilage harvested from a bifid rib would be useful as a material used in plastic surgery. If so, patients having a bifid rib and other anatomical defects necessitating surgical reconstruction based on the cartilage could have two defects corrected at the same time. For example, coexistence of the bifid rib and undeveloped auricle (microtia) can be found in patients with the oculo-auriculo-vertebral spectrum (Goldenhamer syndrome). Khera *et al.* described the case of a patient with oculo-auriculo-vertebral spectrum who had bilateral microtia and bifid rib [50]. Additionally, Wu *et al.* highlighted the possibility of coexistence of the thoracic deformities, including rib bifurcation with microtia. In such cases, the cartilage from autografts is used to restore the proper appearance of the malformed auricle. Both Wu and Zhang described that microtia with associated thoracic deformities may represent a new, previously undescribed syndrome or be another extension of the broad spectrum of microtia [51, 52].

Although the coexistence of bifid ribs with body deformities and anatomical defects is rare, a simultaneous approach for their correction could appear an effective solution for patients subjected to plastic surgery. In this way, the cartilage of the bifid rib would be a new donor-site utilized in autografts. The surgical creation of a three-dimensional framework of the ear using costal cartilage has been regarded as the most challenging part of ear reconstruction [53–55].

Conclusions

Most cases of bifid ribs are asymptomatic and do not need medical interventions after diagnosis. However, their presence may be related to genetic disorders or isolated anomalies of the developing thoracic skeleton. Bifid ribs are generally a benign anomaly; their potential association with serious diseases needs monitoring, especially in children. Accidental encounter of the bifid rib during certain surgeries may be a challenge for surgeons. Therefore, future research and discussions on bifid ribs are needed, especially for physicians involved in pediatric surgery and interested in the variation of thoracic anatomy.

Conflict of interest

The authors declare no conflict of interest, nor any financial interest associated with the current study.

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