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Analysis of imaging features and clinical manifestations in children with congenital rib deformities: a retrospective study

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Abstract

Background Congenital rib deformity is a common thoracic deformity that has a potentially far-reaching impact on the growth and lung function development of young children. Early diagnosis and accurate assessment of congenital rib deformity is of great importance. This study was to retrospectively analyze the number, location, and types of deformities, imaging features as well as clinical symptoms of children with congenital rib deformities.

Methods Children who were diagnosed with congenital rib deformities between October 2019 and October 2021 in our hospital were included in this study. The rib deformities were analyzed according to the imaging results of chest X-ray and 3D volume rendering multidetector computed tomography (MDCT). The data were analyzed using SPSS 22.0.

Results A total of 472 male and 186 female children with rib deformities were detected in this study, with a male to female ratio of approximately 2.54:1. Of the deformed ribs, 417 (63.4%) were located on the right side, usually single and unilateral. The most common type of the detected rib deformity were bifid ribs (95.14%). Rib deformity was most common in the fourth rib (46.62%). The majority (76.16%, $n=428$) of rib deformities were incidental findings and asymptomatic.

Conclusions Congenital rib deformities in pediatric patients included in our hospital were more frequently observed in males than females, more frequently detected on the right than on the left side. The most common type of rib deformity is the bifid rib. MDCT examination are of great value in the diagnosis of rib deformity and can help guide clinical treatment.

Keywords Congenital rib deformity, Imaging features, Clinical, MDCT

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Background

During normal development, infants are born with 12 pairs of ribs [1], consistent between males and females. The ribs protect and provide space for vital organs including the heart, lungs, and others in the chest and abdomen. However, some infants are born with congenital malformations of the ribs [2]. These can lead to minor variations in normal anatomy to severe and life-threatening lung constriction conditions [3]. The different forms of rib deformities include extra (redundant) ribs [4], missing ribs (hypoplastic ribs) [5], abnormally short ribs [6], abnormally shaped ribs [7], and fused ribs [8].

Congenital rib deformity is a common thoracic deformity that can affect health and the quality of life of patients [9]. Abnormalities in rib quantity and structure are found in approximately 2% of the general population, and their prevalence may vary between racial categories and age groups [10]. Rib deformities may cause misalignment of rib counts during physical examination or surgeries, which may lead to accidental injuries during procedures, or even cause fatalities. Therefore, comprehensive understanding of the imaging features and clinical manifestations of rib deformities is essential for accurate diagnosis and avoidance of unnecessary interventions.

Although pediatric congenital rib malformations are usually asymptomatic and most are isolated, they may sometimes be combined with other congenital malformations or hereditary disorders, such as nevoid basal cell carcinoma syndrome, congenital spinal deformity, and thoracic outlet syndrome [11, 12]. Therefore, an in-depth analysis of epidemiologic data such as the overall incidence, gender differences, and site distribution of congenital rib deformities can function to assist in the early screening and diagnosis of congenital rib deformities. For example, respiratory function is mainly completed by the intercostal muscles and diaphragm [13]. For children with rib deformity, their intercostal muscles are different from uninfected population [14], which leads to limited thoracic mobility and insufficient motivation to complete respiratory function [15].

Therefore, it is of great significance to understand the mechanisms, clinical diagnosis and treatment of rib deformity. The information of the incidence rate, gender difference, location distribution, deformity type and clinical symptoms of rib deformity would be of great clinical significance. However, current research especially domestic investigation on rib deformities is scarce. On the other hand, from diagnosis aspect, since rib deformity needs to be differentiated from chest wall tumor and rib fracture, CT is more advantageous than X-ray in displaying rib abnormality [16]. Axial CT examination can show the relationship between bifurcated ribs and the thorax [10], but lacks in displaying the three-dimensional morphology. One technology we used in this study, 3D-CT reconstruction, has advantage of showing the structure of the bifurcated ribs as well as the adjacent area more clearly [17].

In the present study, by collecting the clinical data of 658 children with congenital rib malformations diagnosed in our hospital from October 2019 to October 2021, we performed retrospective analysis to extract the basic information, and to investigate number of rib deformities, occurrence sites, deformity categories, clinical symptoms and treatments of the included children, aiming to provide a valuable clinical reference. Our findings may help clinicians better understand the characteristics of congenital rib malformations and provide guidance for their management and treatment.

Materials and methods

Patient demographics

In this retrospective, single-center, cross-sectional descriptive study, we reviewed the hospital medical records of all children aged 0 to 14 years diagnosed with congenital rib deformities by chest radiography admitted to our hospital between October 2019 and September 2021. Patients who could not be evaluated optimally because of multiple bone fractures after trauma, history of thoracic surgery, primary or metastatic tumors, spinal (vertebral) deformity, or a movement artifact were excluded from the study. The following variables were collected for each patient: sex, age, the findings of chest X-ray and/or CT examination, the location of the rib deformities, and the presenting symptoms and signs. All data were presented as percentages or mean ± standard deviation (SD).

Analysis method

In this study, we retrospectively analyzed the basic information, the number of rib deformities, the site of occurrence, the type of deformity, clinical symptoms and treatment of the affected children. The classification criteria for the categories of rib deformity were defined as: 1) results of homeotic transformation: including supernumerary ribs (cervical) and aplastic rib; 2) segmentation errors: including costal fusion and costal bridges; and 3) anomalies of resegmentation: bifid ribs.

Imaging methods

Chest X-ray method: General Medical Merate S.P.A. digital DR was used. Children under 5 years of age were projected in the supine position, while children over 5 years of age were projected in the standing position. A filter grid was used for both positions. The exposure conditions were selected according to the age and size of the child, typically ranging from 60 to 70 kVP, and 6.0 to 8.0 mAs, with exposure mode automatic. Younger children were exposed during the intervals between cries, and older children were trained to acquire post-air closure to minimize the exposure field, to boost image clarity and reduce scattered rays. The digitized DR images were initially post-processed using the appropriate software [18]. The images were then post-processed using the imaging workstation, including narrowing the display field to improve contrast, adjusting the x-ray window width and level, and contrasting the bilateral lung fields to improve contrast.

Chest CT examination method: GE LightSpeed VCT and GE Revolution CT scanners with low-dose scanning mode were used for acquisition, with a scan layer thickness of 0.625 mm, a reconstruction layer thickness of 5 mm, and a pitch of 0.984:1. CT scan data were acquired on a GE AW 4.6 workstation for coronal, sagittal multi planar reformation (MPR), Maximum intensity projection (MIP), Min-IP, and/or volume rendering (VR) image post-processing. For children who were uncooperative, the CT scan was performed post-sedation and hypnosis, and the medication was 10% chloral hydrate 50 mg/kg.bw orally, or transanal enema if it was difficult to take orally (if necessary, phenobarbital sodium 5 mg/kg.bw was added, intramuscularly).

The CT images were read by two experienced diagnostic imaging physicians who look at the ribs and costal cartilages in the following order: rib and cartilage morphology, number, and alignment; sternal structure and morphology; and soft tissues of the chest wall. 3D reconstruction technology provided a clear and three-dimensional display of the thoracic tissues and their anatomical relationships to each other. Techniques including MIP, VR, and MPR had different effects on the overall display of ribs and costal cartilage, and VR had the best effect on the morphology, alignment, and contour of ribs and costal cartilage, which provided a close anatomical structure for the image effect.

Statistical analysis

SPSS 22.0 [19] was used for statistical analysis of the data. The data were described in the form of numbers (percentages), the difference between the groups was tested by chi-square. The Fisher exact experience was used when the expected frequency was less than 1. The Bonferroni method was used for pairwise comparison between groups. A p value of less than 0.05 (two-tailed) was considered statistically significant.

Results

Demographic and clinical characteristics

A total of 658 pediatric patients who had been diagnosed with congenital rib deformities during the study period were included in the study. The age of the patients ranged from 3 months to 14 years old, and the median age was 4.1 years old. Approximately three-fourths (71.73%, $n=472$) of the patients were boys, leading to male to female ratio of patients around 2.54:1. Among 658 patients, 61.55% ($n=405$) had chest X-ray findings only, 24.92% ($n=164$) had chest CT findings only, and 13.53% ($n=89$) had both chest X-ray and chest CT findings. Among them, 562 had a single rib deformity and 96 had multiple rib deformities.

Table 1 The side of rib deformity of the study population ($n=658$)

Side	Boys ($n=472$) (%)	Girls ($n=186$) (%)	Total ($n=658$) (%)
Right	297 (62.92)	120 (64.52)	417 (63.37)
Left	144 (30.51)	57 (30.65)	201 (30.55)
Bilateral	31 (6.57)	9 (4.84)	40 (6.08)

Table 2 The location of the single rib deformity of the study population ($n=562$)

Location	No. (%)
3rd	114 (20.28)
4th	262 (46.62)
5th	123 (21.89)
6th	38 (6.76)
Other ribs	25 (4.45)

Distribution of rib deformities

Among 658 patients, 562 (85.41%) had a single rib deformity and 96 (14.59%) had multiple rib deformities. Table 1 summarized the site distribution of deformed ribs. Of all the patients, two-third of deformed ribs were located on the right side. This distribution pattern was similar across genders. The location of rib deformities was presented in Table 2. Among single rib deformities, nearly half were located at the fourth rib.

Types and frequencies of rib deformities

There were a total 778 deformed ribs detected in 658 patients, in which 77 patients had 2 deformed ribs, 14 children had 3 deformed ribs, 5 children had 4 deformed ribs. The most recent proposal for developmental classification of rib anomalies divides deformed ribs into three categories: (I) results of homeotic transformation, referring to numerical aberrations; (II) segmentation errors, including costal fusion and bridging; (III) anomalies of resegmentation, resulting in bifid ribs [9]. Table 3 shows the types of rib deformities and their frequency of occurrence. Of all the patients, in 626 cases (95.14%) the types were bifid ribs (Fig. 1A-C), 22 (3.34%) costal fusion (Fig. 1D), 15 (2.29%) costal bridges (Fig. 1E), 5 (0.76%) aplastic ribs (Fig. 1F), and 2 (0.30%) cervical ribs.

Clinical symptoms of single rib deformity

Table 4 presents the clinical symptoms of the single rib deformity of the study population. The majority (76.16%, $n=428$) of rib deformities were incidental findings and asymptomatic. The most common presenting symptoms

Table 3 Type of rib deformities among 658 pediatric patients

Type	Mechanism	Frequency(%)
I	Results of homeotic transformation:	
	a. Supernumerary ribs (cervical)	2 (0.30%)
	b. Aplastic rib	5 (0.76%)
II	Segmentation errors:	
	a. Costal fusion	22 (3.43%)
	b. Costal bridges	15 (2.29%)
III	Anomalies of resegmentation:	
	Bifid ribs	626 (95.14%)

More than one of these types were present in some patients

were local protrusion of the chest (20.46%, $n=115$), followed by asymmetry chest wall (2.31%, $n=13$) and collapse of the chest wall (1.07%, $n=6$). The relationship between different types of rib deformities and clinical symptoms were also analysed. Among patients with bifid ribs, 22.5% presented with local chest protrusion, compared to 18.7% in other deformity types ($p<0.05$).

Discussion

Congenital rib deformity is a common thoracic deformity that can affect health and the quality of life of patients [20], and has a potentially far-reaching impact on the growth and lung function development of young children [21]. Early diagnosis and accurate assessment of congenital rib deformity is of great importance [22]. This study retrospectively analyzed the imaging and clinical data of 658 children with congenital rib deformities diagnosed by imaging followed by clinical confirmation in our hospital between October 2019 and October 2021. Consistent with reported studies in different populations, the results showed that congenital rib deformities in children were more frequent in males than females (with a male to female ratio of approximately 2.54:1), and more frequently observed on the right than on the left side. Some of previous studies have reported different prevalence between genders [23, 24], while this difference may result from the ethnicity and region variety. Moreover, in the present study, the incidence of rib deformity was ranked from highest to lowest as fourth rib > fifth rib > third rib > sixth rib > other ribs, which was generally consistent

Table 4 The clinical symptoms of the single rib deformity of the study population ($n=562$)

Symptoms	No. (%)
Asymptomatic	428 (76.16%)
Local protrusion of the chest	115 (20.46%)
Asymmetry chest wall	13 (2.31%)
Collapse of the chest wall	6 (1.07%)

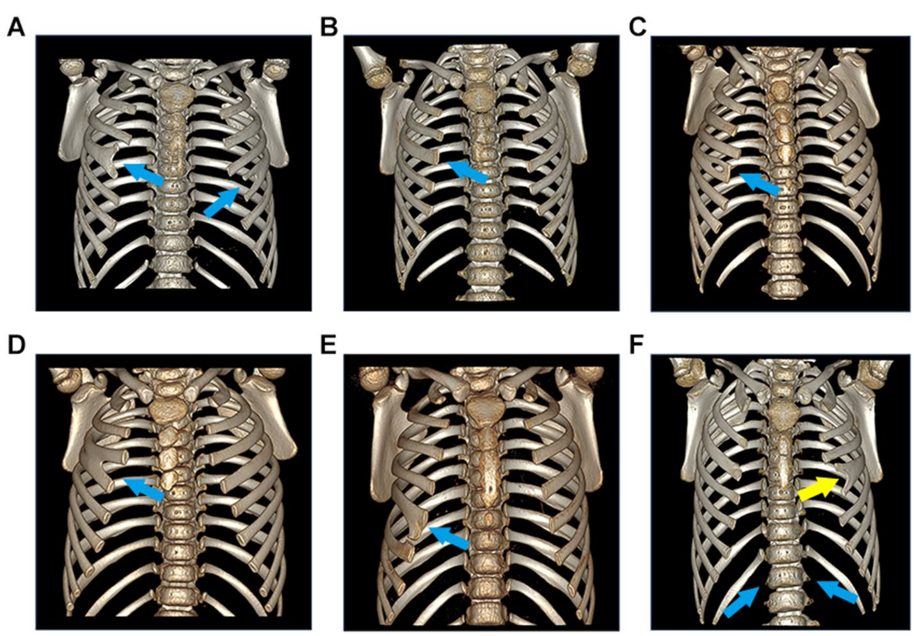

Fig. 1 3D volume rendering multidetector computed tomography (MDCT) images showing the rib deformity. **A**-**C** top panels from left to right; **D**-**F** bottom panels from left to right. **A**-**C** Three different cases of bifid rib type congenital abnormality; **A** Fork type bifid ribs (arrows); **B** Enlargement in the rib (widened type, arrows); **C** Hole type (arrows); **D** Costal Fusion between the anterior parts of the right 3st and 4th ribs; **E** Costal bridges (blue arrows) in the anterior parts of the right 6th and 7th ribs, (blue arrows); **F** Aplastic ribs (11 pairs of ribs) and fork type bifid rib in the left 4th rib (blue arrows)

with previous reports in the literature [25, 26]. In this and previous studies, the highest incidence of solitary rib deformity was found in the fourth rib [27], which may be related to certain biomechanical factors during rib development [26].

In a study conducted by Davran et al., congenital rib anomalies were investigated in 650 individuals (231 female, 419 male) with multidetector computed tomography (MDCT), and 54 bifid rib anomaly was found in 44 (6.76%). 82% of bifid ribs were in men, and in unilateral ones, both sides were equal [28]. In another study by Oner Z et al., they conducted with a higher number of cases in the Turkish population, a lower rate of bifid rib variation (1.34%) was found [29]. In that study, more bifid rib was found in women and on the right side (6.5x). The most common location of bifid rib variation was 3-5th ribs, similar to previous studies [25, 26, 28]. Our study's findings are in consistent with these reports. In our study, the most common type of rib deformity detected was the bifid rib. An average of 2% of people have anatomical abnormalities in their rib structure [30, 31]. Of these, bifid rib, a condition where the ribs split into two parts, accounts for 28% of the whole population with rib abnormalities, making it the most common form of rib deformities [17, 30, 32]. While most cases of multiple bifurcations are found on one side of the body, there have been instances of bilateral occurrences [25, 32, 33]. The third or fourth ribs are most commonly affected by bifurcation, with a tendency to appear more frequently on the right side compared to the left side [32]. Additionally, research consistently shows that bifid rib is more prevalent in males than in females [32].

The origin of the most common congenital rib deformity bifid rib, was deduced by previous reports. Regarding the developmental origins of bifid rib condition, Oostra and Maas [34] made a significant proposition by theorizing that this abnormality arises early in the developmental process, possibly during the re-segmentation phase of sclerotomes, which is critical for the formation of vertebrae. Distinct from other rib deformities, bifid ribs typically do not occur alongside vertebral anomalies [30]. This observation strongly supports the idea that the defect likely affects only the fusion between the layers of the sclerotome that give rise to the ribs, rather than the entire vertebral segment [34]. Despite the available evidences, scientific research has yet to unravel whether bifid is a consequence of developmental processes during the embryonic stage or if it manifests post-natal. This enigmatic feature of bifid ribs remains a topic of debate [35].

Despite the reported literatures, the underlying genetic factors of bifid ribs are still complex and yet to be solved [26]. Some reports considered bifid rib as a pathogenic anomaly [36, 37], while others have suggested that the presence of bifid ribs is closely related to genetic abnormalities [26, 38, 39]. The frequent co-occurrence of bifid ribs within various genetic syndromes has fueled theories that this structural rib variation is indeed orchestrated by genetic influences [26]. Despite these considerations, the rarity of bifid ribs among the broader population casts doubt on the condition's genetic basis [26]. This dichotomy adds a layer of complexity to our understanding of bifid ribs which show as the most detected deformity of ribs in an array of 658 Chinese pediatric patients, challenging the simplistic notions of its genesis.

In our study, among the 562 cases of solitary rib deformity, 134 children (23.84%) had associated clinical symptoms or signs, while the rest were unintentionally detected during chest X-ray or CT examination for other medical needs. The clinical symptoms or signs were mainly localized bulging or collapse of the chest wall, and none of the children showed impaired pulmonary function or growth restriction. Although the majority of cases involving a bifid rib are reported to be asymptomatic, a growing body of medical literature has documented an array of symptoms in patients with this condition, including chest wall irregularities, chest pain, and dyspnea [40]. Diagnosing rib deformities including a bifid rib, particularly in young children who may present with multiple abnormalities, poses a significant diagnostic challenge and, as a result, can often be overlooked by healthcare professionals [30]. In contrast to other rib deformities, a bifid rib typically presents independently, without accompanying spinal anomalies. This unique characteristic further underscores the complexity and the subtlety involved in recognizing and accurately diagnosing this rare rib deformity [30].

In the past, only clinical palpation and traditional X-ray examination were performed. Traditional X-ray examination typically provides overlapping images, since most of the ribs cannot be adjacent to the film during imaging. The overlapping of multiple organs in front and behind such as mediastinum, diaphragm, and abdominal organs also makes it difficult to detect certain deformities of the ribs and costal cartilages. This can result in misdiagnosis and omission of the diagnosis. Physical examination typically reveals limited elevation or depression of the chest wall on the affected side, with no tenderness [41]. Following the initial discovery of an anomaly, MRI and CT scans are frequently employed as essential diagnostic tools [25]. These advanced imaging techniques have proven invaluable in isolating and defining more detailed clinical manifestations [25, 42, 43].

While the use of CT scans in asymptomatic children is debatable, it was crucial in our study for accurate diagnosis and classification of rib deformities. To minimize radiation risks, we implemented the following measures:1. We used a low-dose scanning protocol specifically designed for pediatric patients.2. During the scanning process, we applied strict radiation protection for areas outside the chest. This included using lead caps, lead collars, and lead aprons to protect the head, thyroid, and abdominal areas. These measures ensured that we obtained high-quality diagnostic images while minimizing the radiation dose received by the patients. Future research will continue to explore more advanced low-dose CT technologies or other imaging techniques to further optimize the safety and effectiveness of pediatric imaging examinations.

A previous study demonstrated that a patient with a flared rib on a radiograph showed bifurcation of the costal cartilage on 3D-CT, and that patients with upper rib abnormalities on radiography had downward extension of the cervical or first rib articulating with the upper branch of the bifid first or second rib. This indicated that reconstruction 3D-CT can demonstrate complicated thoracic abnormalities in patients with atypical appearance of the rib on plain radiographs [24]. It has also been reported that multi-slice spiral CT (MSCT) is more advantageous than plain X-ray in revealing rib deformity [44, 45]. MSCT can demonstrate the relationship between the bifurcated ribs and the thorax. Furthermore, three-dimensional restructuring can reveal the relationship between the bifurcated ribs and the rib cartilage joints, as well as the relationship between adjacent tissues, more clearly, which enables the assessment of the patient's deformity status [46]. Furthermore, CT scanning assists to confirm rib abnormalities found on radiographs, and CT axial images can reveal the relationship of the ribs to the thoracic cavity, but may be limited in demonstrating three-dimensional morphologic abnormalities of the ribs [28]. In contrast, 3D CT permits multiplanar reorganization and a variety of post-processing, which can help to further identify sternal variations, rib variations, or deformities [47]. In the present study, 3D volume rendering multidetector computed tomography (MDCT) were used to show the rib deformity and different types of rib deformities including bifid ribs, costal fusion, costal bridges, aplastic ribs and cervical ribs were detected, indicating that MDCT is important and is essential for a definitive diagnosis. The use of three-dimensional CT reconstruction to make a definitive diagnosis and to rule out other diagnoses (including rib exostoses, calcified pleural plaques, foreign bodies, native or postsurgical heterotrophic calcification, and tubes and lines) is important to avoid unnecessary further testing or intervention [48].

Our study indicated that the imaging features of congenital rib deformity have potentially multiple manifestations and thus a complete imaging examination is of great importance for the diagnosis of congenital rib deformity.

There are some limitations to this study. This is a retrospective study and is limited by the completeness and accuracy of the medical record data. Some of the pediatric patients received only chest X-rays, which may have resulted in less accurate assessment of certain rib deformity types. The retrospective study design is prone to selection bias. In addition, although our study had a sufficient sample size, it was conducted on a specific population from a single center and may not be sufficiently representative of the broader population. More comprehensive and multi-center studies are needed to validate the results of this current study.

Conclusions

In conclusion, the results of this study suggest that children with congenital rib deformities in our hospital, China are more likely to be male than female, more likely to be right-sided than left-sided, and the most common type of rib deformity is the bifid ribs. The clinical symptoms of rib dysplasia differ significantly from those of other types of rib deformities and may be used as one of the indicators for early screening. Chest X-ray and CT examinations are of great value in the diagnosis of rib deformities and can help guide future clinical treatment. This study depicts the overall picture of congenital rib deformities in young children in Shanghai China by including large clinical samples, providing a theoretical basis and data support for further clinical studies and guiding clinical practices.

Abbreviation

MDCT Multidetector computed tomography

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Authors' contributions

Shujun Ke and Jing Liu designed the study, performed data analysis, and contributed to the manuscript writing. Huiyong Hu was responsible for data collection, collation, and statistical analysis. Xiuhua Duan conducted patient follow-up. Haifa Hong participated in the study design and manuscript revision. Li Shen was responsible for obtaining funding. All authors reviewed and approved the final version of the manuscript.

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Data availability

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
The research protocol for this study was approved by the Medical Ethics Committee of Shanghai Children's Hospital, Shanghai Jiao Tong University School of Medicine and the need for informed consent was waived by the Institutional Review Board due to the retrospective nature of the study.

Consent for publication
Not applicable.

Competing interests
The authors declare no competing interests.

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