scientific reports

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Bifid rib in bioarchaeological material on the example of new cases from Poland with literature review and proposal of classification

Joanna Mietlińska-Sauter[1]✉, Elena Varotto[2,3], Elżbieta Żądzińska[1,4], Wiesław Lorkiewicz[1,5] & Francesco Maria Galassi[1,4,5]

The anomaly known as a bifid rib is difficult to quantify in bioarchaeological collections not only because of its rarity in the population itself (less than 1.5%), but also because of vulnerability to postmortem damage of this part of the skeleton as well as similarity to other developmental anomalies, which can lead to its misidentification. This work presents five cases of rib bifurcation (from four individuals) from four different populations living in Kujawy region in north-central Poland from the Neolithic until the Early Modern Era. Morphological analyses are matched by a paleoradiological investigation. Furthermore, this study endeavours to summarize all known cases of rib bifurcation in archaeological collections and discuss the correctness of the diagnoses. Additionally, criticism of the arguments allegedly proving the presence of naevoid basal cell carcinoma syndrome (NBCCS, Gorlin-Goltz syndrome) in the archaeological populations based on available photographic data of bifurcated ribs and associated anomalies is made. Finally, a new proposal for the classification of this anomaly in osteological material is presented.

Keywords Forked rib, Rib bifurcation, Congenital anomaly, Poland, Gorlin-Goltz syndrome, Naevoid basal-cell carcinoma syndrome

The frequency of congenital rib anomalies varies quite widely: 0.15–3.4% in populations of European and African ancestry[1], currently accounting for about 2% of all skeletal malformations[2]. Traditionally, they are divided into two types: numerical (mainly the presence of extra ribs, e.g. cervical and lumbar ones[3]) and structural, where changes in bone density as well as abnormal size and shape of ribs are included[4]. Bifid rib belongs to the latter group, in the literature known also as bifurcated rib, forked rib or Luschka's forked rib[5]. It consists in doubling the anterior costal and chondral part of rib (less often only chondral one[6]), in the shape of the fork, but the range of variation in this respect is quite wide – from only a slight widening of the sternal end of the bone rib with the duality of the costochondral junction, to the clear division of the bone itself into two parallel branches, which may constitute a significant part of the overall length of the rib (for contemporary examples of different shapes of bifid rib see e.g.: Song et al.[7] and Tsoucalas et al.[8]).

Contemporary population studies show a clearly more frequent occurrence of bifid rib in males (with a predominance in about 60%[9]), usually unilateral (right > left[9–11]), although cases of bilateral occurrence have also been recorded[12,13]. It affects second to sixth, but most often concerns third and fourth rib (prevalence: third > fourth > fifth > sixth > second[10]). Based on clinical research, bifid rib is a rare condition[14–17] and it accounts for approximately 28–60% of all diagnosed anomalies of ribs[3,11]. Its proper frequency, however, is difficult to determine as it is usually asymptomatic and occurs in an isolated manner[8]. In addition, population studies suggest that its frequency can vary widely between ethnic groups. In the modern Samoan population studied by Martin[9] the frequency of bifid rib was by far the highest amounting to 8.4% – it is possible, however,

[1]Department of Anthropology, Faculty of Biology and Environmental Protection, University of Lodz, Banacha Str. 12/16, Łódź 90-237, Poland. [2]Archaeology, College of Humanities, Arts and Social Sciences, Flinders University, Adelaide, Australia. [3]Department of Cultures and Societies, University of Palermo, Palermo, Italy. [4]School of Biomedicine, Faculty of Health and Medical Sciences, The University of Adelaide, Adelaide, Australia. [5]Shared senior authorship (co-last authors). ✉email: joanna.mietlinska@biol.uni.lodz.pl

that it was slightly overestimated, as it also included spurs on ribs as well as wide or enlarged ribs, treated by the author as a manifestation of the same anomaly as bifid rib. Davran et al.[18] found the presence of bifid rib in 6.8% of patients of Mustafa Kemal University Research Hospital, Hatay, Turkey. In other populations, this frequency is much lower: 2.5% Scottish and 1.8% "mixed Europeans"[19], 2.2% of the French[20], 1.2% of Koreans[11]. The lowest frequency was noted in Americans in their research by Steiner[21] – only 0.01% and Etter[10] – 0.64%.

The aetiology of bifid rib is not fully understood, but the reasons for its formation are currently being sought in embryological development disorders. As reported by Oostra and Maas[22], the direct cause of a bifid rib is most likely an anomaly of "process of resegmentation of the sclerotomes which leads to the formation of vertebrae", as both vertebrae and ribs have the same origin. However, unlike other structural malformations of ribs, bifid rib usually does not coexist with vertebral anomalies, indicating that the defect "only concerned the laminar and costal sclerotome derivatives"[22].

In the case of an isolated bifid rib, this is primarily an incidental finding, contextual with other examinations such as chest X-ray[23] or postmortem examinations[6,8]. Less commonly, it can manifest as a local chest protrusion, being detected by palpation on a physical examination of patients[16]. It may only occasionally contribute to anterior puncture swelling and chest pain[15,24], chest deformity (e.g. so called pigeon breast[25]), respiratory difficulty[26], neurological complaints[13], dyspnoea, and haemoptysis[27]. Mori et al.[28] speculate that bifid rib may have been the cause of desmoid-type fibromatosis due to mechanical stimulation in a 42-year-old female patient of their clinic. Less often bifid rib is found with other skeletal and soft tissues malformations (e.g. of cardiac muscle, kidney, skin), which results from the fact that these structures have the same mesodermal origin[29]. In some cases, bifid rib also occurs as one of many symptoms in more complex genetic syndromes, e.g. as one of the consequences of chromosomal anomalies[30], VACTERL association (containing vertebral anomalies, anorectal malformations, cardiovascular anomalies, tracheoesophageal fistula, esophageal atresia, renal and/or radial anomalies as well as limb defects)[3], multiple congenital anomalies[3], malignancy in childhood[31], Jobs syndrome[32], Kindler syndrome[33] or naevoid basal cell carcinoma syndrome (NBCCS), that is so called the Gorlin-Goltz syndrome[34], caused by a mutation in the tumour suppressor PTCH1 gene.

From the very beginning of research on NBCCS, bifid rib is mentioned, erroneously but surprisingly often, in the context of the pathognomonic features of this syndrome. Gorlin and Goltz[34] were the first to present a list of skin and bone changes accompanying NBCCS (already then containing rib bifurcation), which was significantly extended in later years and still undergoes some modifications[29]. Regardless of this, there are so-called major and minor criteria of this syndrome. The most frequently mentioned major criteria visible on bones are odontogenic keratocysts of the jaw, bilamellar calcification of the *falx cerebri* as well as bifid, fused or markedly splayed ribs. Minor criteria includes for example macrocephaly, cleft palate, frontal bossing, hypertelorism and others[35]. A positive diagnosis requires a minimum of one major and two minor or two major criteria, unless genetic testing is possible. Although NBCCS is a complex genetic alteration of wide phenotypic variability, some researchers continue to pay particular attention to the presence of bifid ribs, suggesting a direct link to NBCCS, and only a few consciously emphasize that it is insufficient[15,22]. This first approach has also regrettably been adopted in bioarchaeological studies, which has led to some abuse of the presence of bifid rib in osteological material as an almost certain indicator of the existence of NBCCS. Another problem with the documentation of this change is that other cases of rib anomalies (e.g. double-headed ribs) are misdiagnosed as bifid rib. The situation is even more difficult as bifid rib publications are rare in the bioarchaeological literature at all[36]. It is most likely related not only to the generally sporadic occurrence of this change, but also to the poor condition of the ribs in archaeological collections (especially the sternal ends are often damaged postmortem). Additionally, relatively few cases are ribs with a pronounced bifurcation of the sternum end, and cases of only slight widening and duality of the articular surfaces may be simply overlooked. Another problem is the misclassification of natural changes in skeleton with age as bifid rib (e.g. ossification of costal cartilages).

The aim of this study is a systematic review of previously published cases of rib anomalies diagnosed as bifid rib, along with an attempt to discuss the correctness of the diagnoses and criticism of the arguments allegedly proving the presence of NBCCS in the archaeological material on basis of occurrence of bifid rib. In addition, five novel cases (found in four individuals) of rib bifurcation are presented, coming from four different populations living from the Neolithic to early modern times in the Kujawy region in north-central Poland. According to our data, this is the first time that bifid rib has been subject of interest for larger scientific bioarchaeological studies. Literature customarily publishes bifid rib only as a marginal mention or anatomical curiosity, but without a broader context, analysis of the frequency or possible links with the individual's health condition and the results of detailed studies.

Materials and methods

Bioarchaeological sample

The individuals with bifid ribs analysed in the present study came from four sites (Fig. 1). Three of them are located within the Brześć Kujawski town, historical centre of Kujawy region, one of the longest and most intensively inhabited regions of Poland[37]. The first population, Neolithic one (4600–4000 BCE), comes from the site Brześć Kujawski 4 (BK4) and was a typically agricultural community representing the Brześć Kujawski Group of the Lengyel Culture (BKG), dealing with wheat cultivation as well as animal husbandry including cattle, sheep, goats, and pigs[38,39]. The second, discovered on the site Stary Brześć Kujawski 4 (SBK4) and dated back to the Middle Ages (12th–16th century CE), was mostly rural, its period of operation coincided with the strong demographic and economic development of this region[40,41]. The most recent population is dated to the 15th–19th century CE and represents a typically urban community with a relatively low standard of living, which is related to the economic decline of Brześć Kujawski at that time as a result of a series of wars for hegemony in the Baltic Sea basin, as well as the fact that the cemetery from which the analysed skeletons come, was at least partly under the management of the poorhouse of Holy Spirit Hospital (Polish: Święty Duch, abbr. SD),

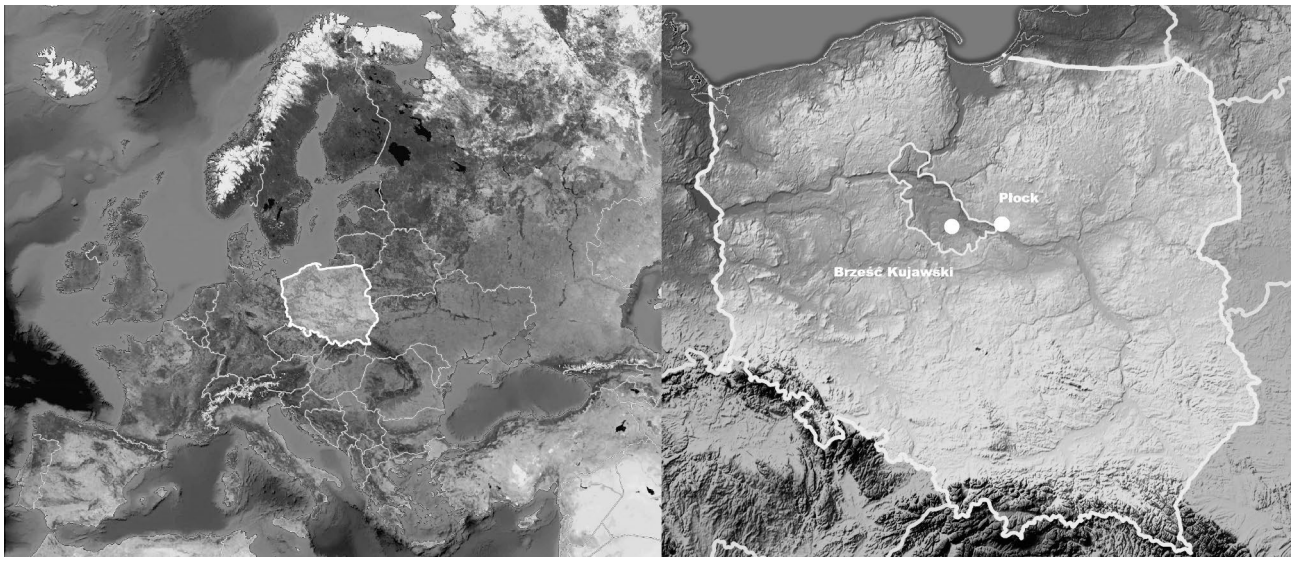

Fig. 1. Map of the location of the Kujawy region as well as towns where bifid ribs were discovered: Płock (with one site) and Brześć Kujawski (with three sites within); image source: openstreetmap.org (CC BY-SA 2.0), original image edited by the first author.

operating in the town at the time[42,43]. The fourth site is found in Płock, a city which at the turn of the 11th and 12th century was the capital of Early Medieval Poland. From this site comes the fourth series which represents Early Medieval population (12th–13th century CE). This site was probably a church cemetery (archaeological research in progress).

The sex was determined only for adult individuals: it was made on the basis of pelvic and cranial morphology[44]. Age at death was estimated on the basis of long bones length (for younger subadults), fusion of secondary ossification centres (for adolescents) as well as tooth wear, morphology of the pubic symphysis and of the auricular surface of the ilium for adults[45–50].

A more detailed morphological study was performed using radiological techniques, in particular X-ray and CT scan imaging with 3D virtual reconstructions. X-ray imaging parameters (Apollo EZ): 45 kV, 20.0 mAs. CT scan imaging parameters (Somatom Sensation 16 S): kV 120, 200 mAs. The acquired radiological images were processed and studied utilizing the OsiriX MD (Pixmeo Sarl) software version 14.0.

All analysed skeletal series came from the collections of the Department of Anthropology, University of Lodz, the Museum of Archaeology and Ethnography in Łódź and Mazovian Museum in Płock. The possession and scientific analysis of the samples were in accordance with the legal status of archaeological human remains in Poland[51].

Anomaly under investigation

A bifurcated rib is defined in this work as a rib that has "a single head and body that divides or forks at the sternal end"[52] connecting with two independent cartilaginous ribs and should not to be confused with the bicipital rib which is a true fusion of two independent ribs or with other congenital rib anomaly, e.g. spurs on ribs, flaring rib or merged rib[53]. Signs of rib bifurcation were searched macroscopically by visual inspection. Out of almost 2,600 individuals making up the analysed collection, only those with at least one rib preserved (calculated on the basis of the number of preserved spinal ends[54]) were included in the study. Emphasis was put on true ribs (1st–7th), as to the best of our knowledge, no case of bifurcation of false and floating ribs (8th–12th) has been found so far.

Analysis of literature data and verification of existing bifid rib cases

Information on known cases of bifurcated ribs in the bioarchaeological literature was collected using biomedical and academic databases: *Google Scholar*, *Pubmed*, *Scopus (Elsevier)*, *Web of Science*, *Wiley*, etc. as well as archaeological monographs and posters. The search was conducted in the following European languages: English, French, German, Italian, Polish, Spanish. Verification of known cases was made on the basis of available photos, illustrations and written morphological descriptions. Additionally, whenever possible, the authors of the papers in which these cases were published for the first time were directly contacted to obtain additional clarifying information regarding the anomaly under investigation.

Results

New cases of bifid rib from Poland

In the studied populations, five bifid ribs were found in four individuals (each one from the Neolithic (BK4), Early Medieval (Płock), Medieval (SBK4) and Early Modern (SD) population), which gives the prevalence of this anomaly in: 0.89 [0.85–2.64]% (n/N = 1/112), 2.04 [1.92–6.00]% (1/49), 0.26 [0.25–0.76]% (1/388) and 0.40 [0.38–1.17]% (1/252) of the population. Information about these cases is summarized in Table 1.

Chronology	Site, city/region and country	Inventory number of individual	Age at death (in years)	Sex	Location—side of the body (number of ribs, rib number)	Probable cause of occurrence according to authors	Other congenital anomalies
4600–4000 BCE	BK4, Brześć Kujawski, Poland	BK4_36	20–25	Male	Left (one rib, number 4)	Congenital anomaly	Spina bifida of S1
1100–1300 CE	1st of May 3/5 Street, Płock, Poland	Płock_20	13–14	Unknown	Right (one rib, number 4)	Congenital anomaly	Widening of left R3
1100–1600 CE	SBK4, Old Brześć Kujawski, Poland	SBK4_664	35–40	Unknown	Right (one rib, number 5)	Congenital anomaly	None
1400–1900 CE	Holy Spirit, Brześć Kujawski, Poland	SD_56	45–55	Male	Right (two ribs, number 3 and 4)	Congenital anomaly	None

Table 1. New cases of bifid rib from Poland.

Fig. 2. General view of bifid ribs: (**a**) fourth right rib of a male (45–55 years) from the SD site; (**b**) third right rib of a male (45–55 years) from the SD site; (**c**) fifth (?) right rib of individual of undetermined sex (30–35 years) from the SBK4 site; (**d**) fourth right rib of subadult (13–14 years) from the Płock site; (**e**) fourth left rib of a male (20–25 years) from the BK4 site.

Case 1 BK4_36 (4600–4000 BCE). The skeleton of a male died at the age of ca. 20–25 years (young adult) is almost completely preserved, the observed bipartition concerns the fourth left rib (Figs. 2e and 3). The bony part of the rib is clearly extended at the sternal end and shows two independent surfaces for the attachment of the cartilaginous ribs, but it has not undergone a full-blown dichotomy (Fig. 4e). Apart from the described anomaly, the individual had a supratrochlear foramen in the left humerus and spina bifida at the level of S1.

Case 2 Płock_20 (1100–1300 CE). Nearly completely preserved skeleton of ca. 13–14 years old subadult shows anomaly of fourth right rib. Strong extension of the arch begins about 3 cm from the sternal end, but there is no clear bifurcation, only two independent surfaces to connect with the cartilaginous rib are visible (Figs. 2d and 5). Although the upper surface was partially destroyed postmortem, it seems that they were of the same size (Fig. 4d). This individual shows also widening (without bifurcation) of third left rib with the extension starting at the level of the angle. The analogous rib on the right side is not preserved, so it is impossible to assess whether the anomaly was symmetrical.

Case 3 SBK4_664 (1100–1600 CE). The partially preserved skeleton belongs to an individual of undetermined sex, who died at the age of ca. 35–40 years (middle adult). On the probably fifth right rib, a strong extension of the sternal end was observed (Figs. 2c and 6) with one (lower) of the two surfaces preserved for connection with the cartilaginous rib (superior branch of the furcation was lost postmortem, Fig. 4c). No anomalies were observed within the preserved bones of the skeleton, apart from numerous minor degenerative changes (in the right sternoclavicular joint and sternocostal joints, and small osteophytes around the edge of the vertebral bodies), which are most probably related to the age of the individual at the time of death.

Case 4 SD_56 (1400–1900 CE). The partially preserved skeleton belongs to a male who died at the age of ca. 45–55 years (old adult). In his case, there was a clear duality of the bony part of the two right ribs (the third and the fourth ones, Figs. 2a,b and 7). In the case of the fourth rib, a strong asymmetry in the size of the attachment surface of the cartilaginous parts of the ribs is visible – the lower surface is almost three times larger than the upper one (in the case of the third rib, both surfaces are of similar size, Fig. 4a,b). In this individual, degenerative changes in the articular processes of the upper and lower vertebrae in the thoracic region of moderate expression, extensive attachments of the costoclavicular ligaments and inactive lesions on the inner surface of the manubrium were observed. In addition, linear enamel hypoplasia, slight porosity within the articular cavities of the shoulder blades, osteophytes around the labra of these bones, eburnation of the heads of both humeri and degenerative changes in the ulnar fossa of the left humerus were observed.

Fig. 3. BK4_36 individual: (**a**) conventional X-ray image in anteroposterior (A-P) projection of the bifid rib; (**b**) CT scan image – coronal view of the rib with its extremities (bifurcation of the left side for the viewer); (**c**) 3D virtual reconstruction of the rib through CT scans – superior and inferior view with a detail of the bifurcation area. Scale = cm (also for Figs. 5, 6 and 7).

Fig. 4. Close-up of the sternal ends of the bifid ribs: (**a**) fourth right rib of a male (45–55 years) from the SD site; (**b**) third right rib of a male (45–55 years) from the SD site; (**c**) fifth (?) right rib of an individual of undetermined sex (30–35 years) from the SBK4 site; (**d**) fourth right rib of a subadult (13–14 years) from the Płock site; (**e**) fourth left rib of a male (20–25 years) from the BK4 site.

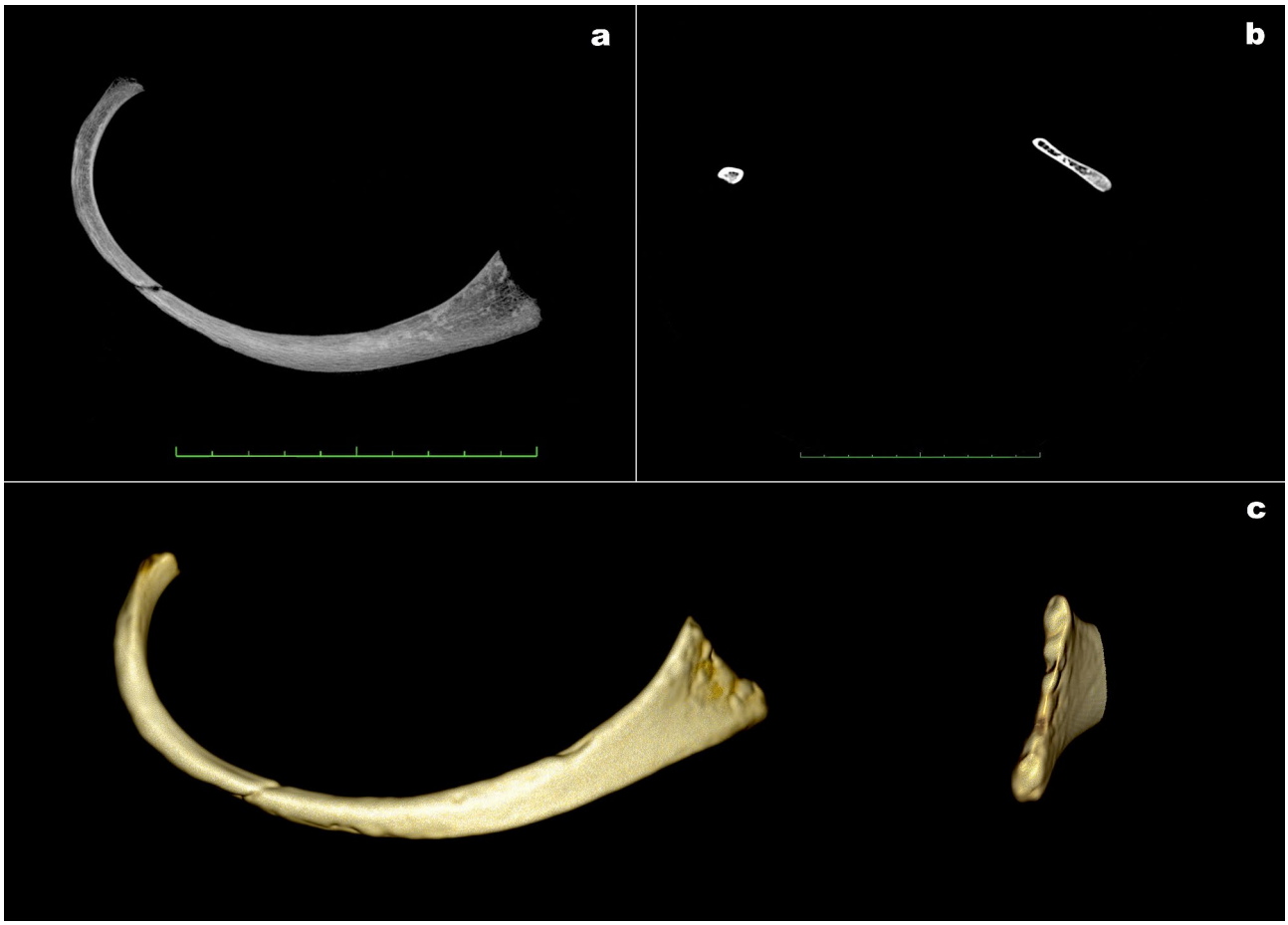

Fig. 5. Płock_20 individual: (**a**) conventional X-ray image in anteroposterior (A-P) projection of the bifid rib; (**b**) CT scan image – coronal view of the rib with its extremities (bifurcation of the right side for the viewer); (**c**) 3D virtual reconstruction of the rib through CT scans – superior view with a detail of the bifurcation area.

Due to the lack of deviations of the obtained results from those presented by other historical cases, samples from Brześć Kujawski were included in the general analyses of the frequency of bifid rib occurrence in the archaeological material.

The application of radiological techniques permits to highlight a flaring of the sternal end of the ribs evidenced by the characteristic rarefaction (lower density) of trabeculae in the central portion, while laterally, where the bifurcation occurs, bone densification can be appreciated. In addition, in the 3D models, in most cases, it is possible to observe an unevenness and thinning of the rib surface where radiologically the trabecular rarefaction was observed: this is even more evident with the 3D model than by macroscopic observation of the rib surface.

Bifid rib in skeletal remains under research

The collected literature data presenting bifid rib cases were divided into three groups: certain cases (with photographic or drawing documentation, Table 2), probable cases (not confirmed by this type of data, Table 3) and rejected cases (Table 4). In the group of accepted cases (Tables 1, 2 and 3; Fig. 8), the majority of bifid ribs were found in males (n/*N* = 13/19, 68.4% of individuals with the possibility of sex assessment vs. only 6/19, 31.6% of females in this group). The vast majority concerned the right side (16/22, 72.7% of individuals having information about the side of the anomaly occurrence vs. 5/22, 22.7% on the left side and 1/22, 4.6% bilaterally), for 10 individuals (31.3%) there was not such information. As many as 30 individuals (93.8%) had an anomaly within only one rib, one – two ribs (3.1%, on the same, right side), and one had numerous (five) bifid ribs (3.1%, in this number only one on the right side). The bifurcation concerned mainly the middle ribs of the thorax: the fourth rib (8/17, 47.1% of individuals for whom the exact information about the bifurcated rib number was given) as well as the fifth (3/17, 17.7%) and sixth rib (2/17, 11.8%). A case of clefts of the 2nd and 3rd ribs was found in one individual each time (5.9% each), moreover, multiple bifurcations were found on the 3rd and 4th ribs (one individual, 5.9%) and ribs 2–5 (one individual, 5.9%). In the remaining 15 individuals, adequate information was not provided.

It seems that most, if not all, of the cited cases were not associated with impaired respiratory function – in the thoracic skeleton no pathological changes of its kind were found to suggest such an altered physiology *intra vitam*. The only visible minor changes in the ribs and vertebrae (Schmorl's nodes, osteophytes) could have

Fig. 6. SBK4_664 individual: (**a**) conventional X-ray image in anteroposterior (A-P) projection of the bifid rib; (**b**) CT scan image – coronal view of the rib with its extremities (bifurcation of the right side for the viewer); (**c**) 3D virtual reconstruction of the rib through CT scans – superior and inferior view with a detail of the bifurcation area.

been a mere consequence of the advanced age or heavy load on the axial skeleton of some of the individuals. These changes were noted by Bencerić, Boston, Capasso, Mazzucchi et al., Miguel-Ibáñez et al., Šikanjić et al. and Vymazalová et al.[36,55–60], they also concerned individuals SBK4_664 and SD_56. The only exception is represented by skeleton 277 from Bridgwater[61], belonging to a subadult who died at the age of 15. However, considering that the observed degenerative changes concern the L1–S1 vertebrae, it does not seem that they could have any connection with limitations in thoracic mobility.

Since the bifid rib is a type of congenital anomaly, it was also checked whether in the examined individuals and in the cases known from the literature there are other abnormalities of the skeleton, especially its axial part. Information on this subject was obtained for 26 individuals out of 32 analysed, with other skeletal anomalies in 13 (cf. Tables 1, 2 and 3), and for the remaining 13 bifid ribs was the only abnormality observed. Among the noted pathologies, the most frequently mentioned were disturbances in the ossification process (failure of sclerotomes resegmentation, sutural and vertebral ossicles, spina bifida, delayed or extra ossification centres, foramina in sternum and segmented sternum) and deviations from the typical structure among other ribs (flared ribs, hyperplastic ribs, fusion of ribs), but it should be noted that flared rib may also be a consequence of metabolic disorders, including rickets[2]. In the case of fusion of foot phalanges[62], it is difficult to determine whether it was a developmental change or a posttraumatic state.

Rejected cases

Based on a thorough analysis of the available photos and descriptions of published cases, the authors propose to reject nine of them (Table 4):

(1) E 235 from Assyut, Egypt[63,64]. This is probably one of the two most famous and oldest examples of bifid rib in the available bioarchaeological literature. Unfortunately, the photos of this individual clearly indicate the ossification of the cartilaginous part of the ribs (in the form of bony spurs), which would not be strange considering the advanced age of the individual (50–70 years). This case is mainly known from works looking for a link between NBCCS and skeletal changes[65]. Of course, it cannot be ruled out that this individual actually suffered from Gorlin-Goltz syndrome, as he has a number of other bone changes, but bifid rib is not one of them.
(2) N/932 from Alkali Ridge site 13, Utah, USA[66]. This is one of the cases which the author himself does not describe as bifid rib (quote: "the left first and second ribs are fused at the necks"), while the incorrect terminology was introduced only by subsequent authors.
(3) N/922 from Alkali Ridge site 5, Utah, USA[66]. As in the case of individual N/932, the author does not provide information about the bifurcation (quote: "two ribs, a second and third, are fused at the posterior part of bodies"), while this as incorrectly introduced by subsequent authors.

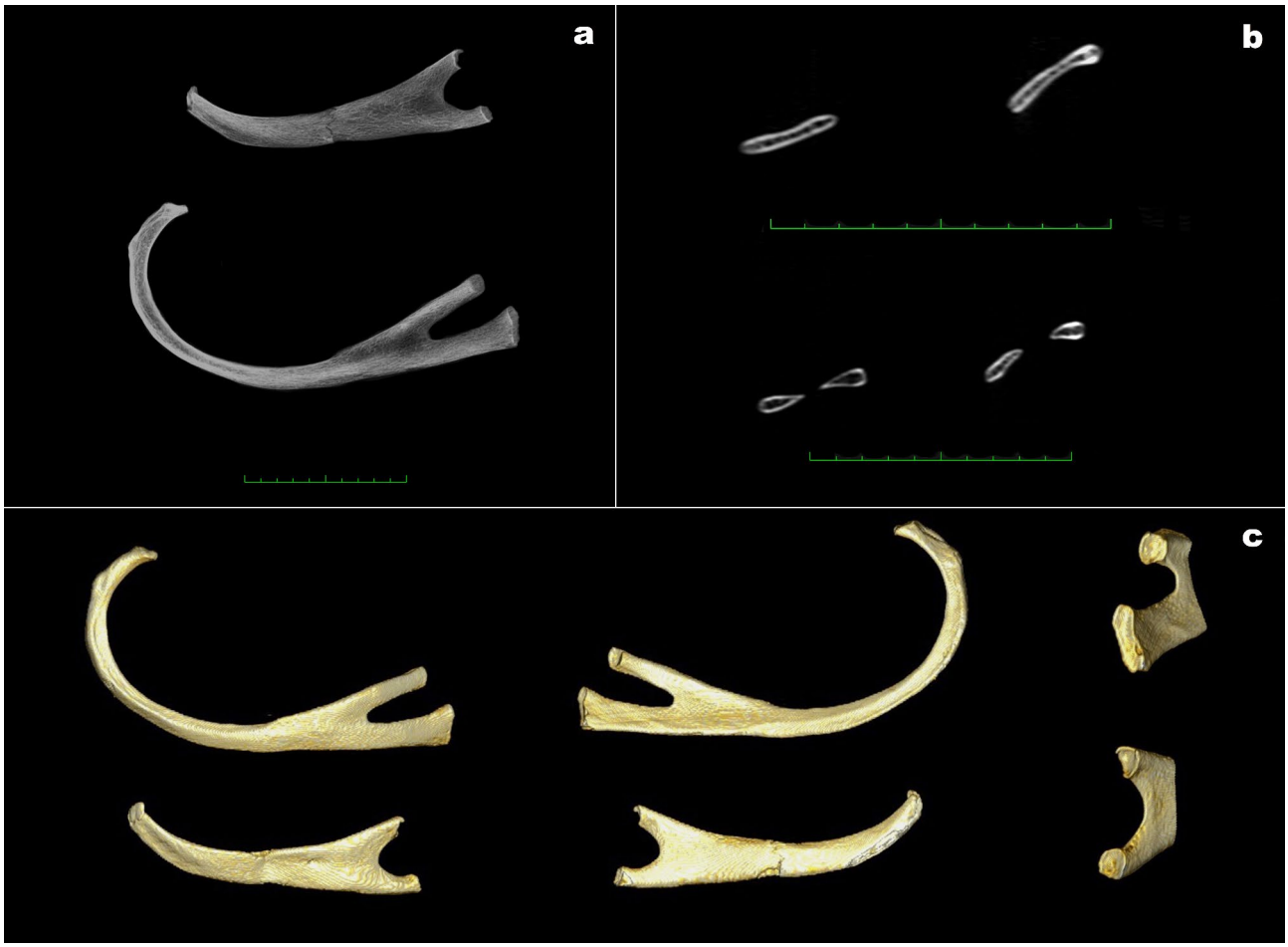

Fig. 7. SD_56 individual: (**a**) conventional X-ray image in anteroposterior (A-P) projection of the bifid ribs: third right one at the top and fourth right below; (**b**) CT scan image – coronal views of the same ribs with their extremities (bifurcations of the right side for the viewer); (**c**) 3D virtual reconstruction of the ribs (fourth right one at the top and third right below) through CT scans – superior and inferior views with a detail of the bifurcation areas.

(4) N/923 from Alkali Ridge site 5, Utah, USA[66]. The author's description states that "the right first rib shows fusion of an anomalous element which extends upward and inward from the middle part of the rib", which indicates rather a rib spur, and similarly to the case of other individuals from Alkali Ridge, it was overinterpreted as a bifid rib.
(5) #8 from Caldwell Village, Lapoint, Utah, USA[67]. Based on the published description and photo, it can be said with certainty that this case is partial bridging or articulation between first and second rib (according to the classification by Barnes[68]). Similarly, as in previous cases from the USA, the author himself does not mention a bifid rib when evaluating this pathology, hence the erroneous term was introduced later.
(6) B418 from cemetery of St. Oswald's Priory, Gloucester, England[69]. Based on the available photo, it can be concluded that it is a congenital fusion of the ribs, not rib bifurcation (there are two heads and two separate arches which connect to each other only at one point, forming actually bridging at vertebral end). The author herself uses both the terms "bifid rib" and "fusion of ribs", thus treating them equally.
(7) 118 from Grasshopper Pueblo, Arizona, USA[70]. Hinkes states that "the sternal ends of ribs one through eight are bilaterally expanded", but clearly states that there is new bone formation on them (perhaps due to periosteal stimulation) and combines the resulting changes with an unspecified disease entity. As bifid rib, this pathology was probably wrongly presented later in paleopathology textbooks.
(8) #419 from Mound 7 at Las Humanas, Gran Quivira, New Mexico, USA[71]. In the case of this individual, the variation is described as "broad protrusion, a large point, about halfway along its length". The erroneous definition of it as bifid rib by subsequent authors is most likely due to the fact that this change was discussed together with the cases of bifurcation known to the author, but it should be emphasized that he himself clearly indicates that this anomaly is rather closer to the one presented by the individual #8 from Caldwell Village (see above).
(9) Individual from Prewitt, New Mexico, USA[72]. This skeleton has, according to the author's description, anomaly analogous to individuals #8 from Caldwell Village and #419 from Mound 7 at Las Humanas. Thus, there is no reason to believe that it is a bifid rib, and the misclassification is secondary.

Reference	Chronology	Site, city/ region and country	Inventory number of individual	Age at death (in years)	Sex	Location—side of the body (number of ribs, rib number)	Probable cause of occurrence according to author	Other congenital anomalies
Šikanjić et al.[35]	5351–5214 BCE	Vinka factory, Vinkovci, Croatia	N/A (individual from grave 1)	35–45	Male	Right (one rib, number 4)	Unknown (Gorlin-Goltz syndrome, Job syndrome, Kindler syndrome and Menkes syndrome were excluded on the base of anatomical investigation)	Hiperplasia of R3, flared R8 at sternal end, ossicles in the lambdoid suture, parastyle on 27 tooth, supracondylar processes on both humeri
Fily et al.[78]	ca. 3400 BCE	Adaima, Egypt	N/A (upper individual from grave S34)	25–30	Male	Right (one rib, number 6)	Minor developmental anomalies arising from a similar genetic determinism with epigenic control of development	N/A
Boano et al.[79]	2450–1955 BCE	Gebelein, Egypt	GED44, E22, drawer 400	4	N/A	Right (one rib, number 4)	Unclear (Gorlin-Goltz syndrome?)	Delay in ossification of the posterior arch of atlas
Satinoff[64]; Satinoff and Wells[63]	2450 –1955 BCE	Assyut, Egypt	E225	20–25	Male	N/A (one rib, N/A)	Gorlin-Goltz syndrome	Spina bifida of S3–S5, relative shortening of 4th metacarpals
Kozieradzka-Ogunmakin[80]	332–30 BCE	Saqqara-West, Egypt	PP B.609	25–30	Male	Right (one rib, number 4)	N/A	N/A
Capasso[56]	700 BCE–79 CE	Herculaneum, Italy	E1	25–30	Male	Left (one rib, number 2)	N/A (according to Ponti et al. 2016 – NBCCS)	Asymmetry of occipital bone, one ossicle in the lambdoid suture, foramen in the xiphoid process
Bencerić[57]	0–400 CE	Ozad Arene, Pula, Croatia	G4	18–27	Male	Left (one rib, number 6)	Insignificant anatomic variations	None
Bencerić[57]	0–400 CE	Ozad Arene, Pula, Croatia	G15	30–40	Male	Left (one rib, number 4)	Insignificant anatomic variations	Two sternal foramina and fused elements of the sternum
Kirkpatrick[81]	0–700 CE	Fag el-Gamous, Faiyum Governorate, Egypt	NW 70 (grave 130/140 N 20/30 W)	15–20	Female?	Right (one rib, N/A but probably upper-thoracic part)	Congenital defect	None
Merbs[75]	1150–1450 CE	AZ U:9:42 (ASU) in Tempe, Arizona, USA*	N/A	N/A	N/A	N/A but probably left (one rib, N/A)	N/A	N/A
Anderson[76]	1280–1400 CE	Homoľovi III, Arizona, USA	N/A (individual from plaza, Feature 32)	9.5 Foetal month	N/A	Right (one rib, N/A but probably upper-thoracic part)	Klippel-Feil syndrome	Failure of separation of the right hemi-arches of the C2 and C3 and the right hemi-arches of two upper- to mid-thoracic vertebrae, bicipital R2 – probable fusion of left R2 and R3
Vymazalová et al.[60]	1805–1806 CE	Staňkova Street, Brno, Czech Republic	800/6	20–30	Male	Right (one rib, N/A)	Anatomical variation	None
Molleson[77], personal communication**	Modern Era	Tell Abu Hureyra, Syria	Tr.A72.338D	Old adult	Female	Left (one rib, N/A)	Developmental anomaly associated with the hypermobility of an individual	N/A
Bolk[88], Oostra and Maas[22]	before 1844 CE	Amsterdam, Netherlands	N/A (original Vrolik collection)	Neonate	N/A	Bilateral (2nd to 5th rib on the left side and 5th rib on the right side)	Unclear (possible developmental defect similar to that seen in open brain (opb) mouse mutant or Gorlin-Goltz syndrome)	Probable anatomical anomaly of hands, malformed laminae of Th1-7 and intercalated ossicles, ossification in the intervertebral disc space between Th11-12 – interpedicular fusion

Table 2. Confirmed cases of bifid rib known from archaeological literature. N/A – data not available; * – known locally as the Broadway and McClintock site; ** – the authenticity of the anomaly confirmed on the basis of a photo sent in private communication.

Reference	Chronology	Site, city/region and country	Inventory number of individual	Age at death (in years)	Sex	Location—side of the body (number of ribs, rib number)	Probable cause of occurrence according to author	Other congenital anomalies
Brothwell et al.[61]	1000–700 BCE	Cannington Park Quarry, Bridgwater, England	277	14–16	Unknown	N/A (one rib, N/A)	N/A	Enlarged antero-posterior length of neural arch of the atlas, segmented sternum, and a partial sacral cleft
Kozieradzka-Ogunmakin[80], personal communication	332–30 BCE	Saqqara-West, Egypt	PP B.439	35–45	Female	Right (one rib, number 5)	N/A	None
Mazzucchi et al.[58]	550–650 CE	Brescia, Lombardy, Italy	N/A (individual from grave T3 US 553)	Perinatal	N/A	N/A (one rib, N/A)	N/A	None
Mazzucchi et al.[58]	550–650 CE	Brescia, Lombardy, Italy	N/A (individual from grave T6 US 591)	30–40	Male	N/A (one rib, N/A)	N/A	None
Miguel-Ibáñez et al.[55]	650–750 CE	La Alcudia de Elche, Alicante, Spain	UE 15.4 (individual number 3 from fossa UE 15)	> 55	Male	N/A (one rib, N/A)	N/A	None
Reed[71], Peckham 1962	950–1100 CE	LA:6402, McCarty's, New Mexico, USA	#7	N/A	N/A	N/A (one rib, N/A)	N/A	N/A
Miles[84]	1100–1300 CE	1801, Wetherill Mesa, Mesa Verde National Park, Colorado, USA	41052/719	5	Unknown	N/A (one rib, N/A)	N/A	None
Matthews et al.[85]	1150–1450 CE	Salado River Valley near Tempe*, Arizona, USA	N/A (Hemenway Collection)	N/A	N/A	N/A (one rib, N/A)	N/A	N/A
Hart and Holbrook[83]	1200–1300 CE	Malmesbury Abbey, Wiltshire, England	B21	18–25	Female?	Right (one rib, number 3)	N/A	None
Milner and Smith[62]	ca. 1300 CE	Norris Farms 36, Illinois, USA	41 (catalogue number 819963)	30–35	Female	Right (one rib, number 5)	N/A	Fused medial and distal phalanges of right and left 5th toes
Milner and Smith[62]	ca. 1300 CE	Norris Farms 36, Illinois, USA	157 (catalogue number 821081)	0–2 moths	Unknown	Right (one rib, number 4)	N/A	None
Milner and Smith[62]	ca. 1300 CE	Norris Farms 36, Illinois, USA	178 (catalogue number 821114)	12–24 months	Unknown	Right (one rib, number 4)	N/A	None
Barnes[68]**	1350–1600 CE	Pueblo IV-V from Hawikuh, New Mexico, USA	NMNH 308632	Adult	Female	N/A (one rib, N/A)	N/A	Flared other rib
Boston[59]	1749–1856 CE	Greenwich Hospital, London, England	3229	> 50	Male	Right (one rib, mid-thoracic part)	N/A	Spina bifida occulta of S3–S5

Table 3. Unconfirmed but probable cases of bifid rib known from archaeological literature. N/A – data not available; * – the exact name of the site is not given, the remains must come from one of the following: Los Muertos, Las Acequias, Los Hornos or Los Guanacos (= Los Altos) representing Salado culture; ** – it should be noted that there are certainly more bifid ribs in the NMNH collection, as evidenced by numerous photos. However, due to the lack of descriptions of the illustrations, it is not possible to assign them to descriptions of individuals. The description given in the table is the only one that contains more detailed data, but it does not have a reference to the particular figure.

Discussion

Regularities in occurrence of bifid rib in populations from Poland

Research on the frequency of bifid rib in the human remains from Brześć Kujawski shows a certain universality of its occurrence: it was observed in populations from different archaeological periods (from the Neolithic to early modern times), inhabiting various types of environments (from the settlements of the first farmers to highly organized cities on the brink of the industrial revolution), characterized by different degrees and types of physical load due to the type of work undertaken (from hoe-farming to sedentary work operating handicraft machines) and, finally, contrasting general standard of living and health (including groups representing the lowest social strata, i.e. poorhouse wards). Moreover, the analysed series differ also in genetic terms, which is particularly visible in the comparison of historical and Neolithic populations (the second with Anatolian ancestry[73]). Despite these discrepancies, the detected cases of bifid rib are characterized by a number of analogies: in all series they were found with a similar, low frequency (0.26–2.04% of the population), they concerned the male sex (in cases where sex determination was possible), were located unilateral, mostly on the right side (in 3 out of 4 individuals), and concerned the ribs of the middle part of the rib cage (numbers 3–5). These results are consistent with literature data for contemporary and, as can be seen below, also for other archaeological populations. Similarly to most of them, they were not accompanied by macroscopically visible changes suggesting the occurrence of serious genetic disorders, but only minor developmental anomalies.

Reference	Chronology	Site, city/region and country	Inventory number of individual	Age at death (in years)	Sex	Location—side of the body (number of ribs, rib number)	Probable cause of occurrence according to author	Other congenital anomalies
Satinoff[64]; Satinoff and Wells[63]	2450–1955 BCE	Assyut, Egypt	E235	50–70	Male	Bilateral (three ribs each side, N/A)	Gorlin-Goltz syndrome	Asymmetry of occipital bone, shortening of 4th metacarpal bone, spina bifida (probably S2–S5), ossicles in the right lambdoid suture
Brues[66]	750–780 CE	Alkali Ridge site 13, Utah, USA	N/932 (Pit House D)	36–55	Male	Left (one rib, number 1)	A consequence of inbreeding	Cervical rib on C7, spondylolysis of L5 (no information if the arch was separated by trauma or if it was a developmental disorder)
Brues[66]	900–1100 CE	Alkali Ridge site 5, Utah, USA	N/922 (burial 2)	18	Male	N/A (one rib, number 2)	A consequence of inbreeding	None
Brues[66]	900–1100 CE	Alkali Ridge site 5, Utah, USA	N/923 (burial 3)	14–15	Unknown	Right (one rib, number 1)	A consequence of inbreeding	Partial bipartiteness of the left first cuneiform bone
Reed[67]	1050–1200 CE	Caldwell Village, Lapoint, Utah, USA	#8 (FS 268)	> 50	Male	Right (one rib, number 2)	N/A	None
Lewis[69]	1153–1857 CE	cemetery of St. Oswald's Priory, Gloucester, England	B418	Neonate (39 weeks)	N/A	Bilateral (one rib each side, N/A)	Congenital syndrome	Cleft vertebrae
Hinkes[70]	1275–1400 CE	Grasshopper Pueblo, Arizona, USA	118	Neonate	N/A	Bilateral (eight ribs each side, number 1–8)	N/A	None
Reed[71]	1550–1672 CE	Mound 7 at Las Humanas, Gran Quivira, New Mexico, USA	#419 (catalogue number Q4427/37)	25–27	Female	Right (one rib, number 2)	N/A	Acromion bipartite, dichotomy of the right occipital condyle
Reed[72]	N/A	Prewitt, New Mexico, USA	N/A	N/A	N/A	N/A (N/A but probably one rib, N/A)	N/A	N/A

Table 4. Rejected cases of bifid rib known from archaeological literature. N/A – data not available.

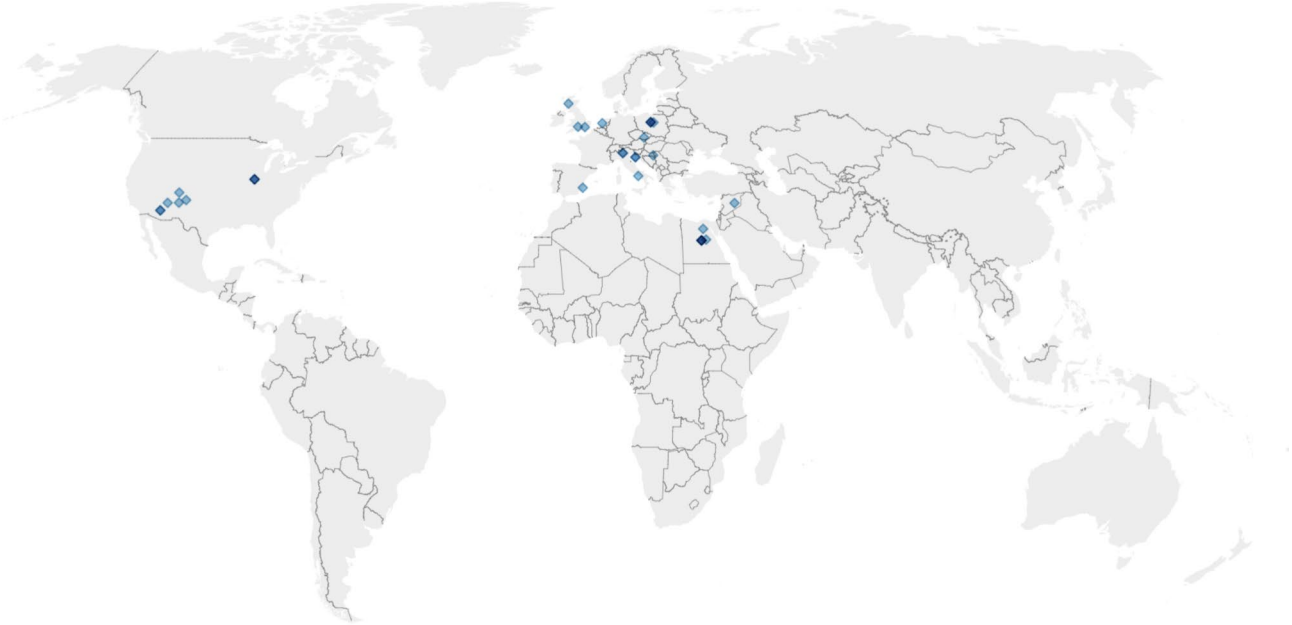

Fig. 8. Map of the sites of all known confirmed and probable cases of bifid ribs of bioarchaeological importance (the darker colour the more known cases from particular site).

Frequency of bifid rib in bioarchaeological populations against the background of contemporary data

The data collected in Tables 1, 2 and 3, although relatively scarce, confirm the regularities known from the clinical literature, regarding almost all examined features: predominance of male sex[9,10], occurrence on one, usually the right side of the body[9–11] and within only one rib[6,7,9,17,28]. Moreover, this defect usually does not correlate with the occurrence of other systemic developmental pathologies in either historical or contemporary[8] series, which suggests that in most cases it is a consequence of only minor disturbances of the embryogenesis process, e.g., incomplete fusion of cranial and caudal segments of single adjacent sclerotomes during their resegmentation, in

particular the costal sclerotome derivatives[22,74]. It is worth noting, however, that in the studied group of accepted cases, as many as 7/32 (21.9%) were individuals who died before the age of five. Although no other skeletal pathologies were detected in four of them, the occurrence of soft tissue anomalies in life cannot be ruled out. Such a high percentage of the youngest individuals in the studied group may suggest that in their case bifid rib was part of complex genetic syndromes leading to premature death. It would therefore be instructive to conduct appropriate studies in the future to test this hypothesis.

Osteological investigations also showed that bifid ribs were not associated with respiratory disfunctions in the examined individuals, which is consistent with the results of contemporary publications reporting that this anomaly is usually diagnosed accidentally during other examinations such as chest X-ray[23], and is not itself a reason for impaired chest movements[8]. The only deviation from the statistical data for contemporary populations is the relatively rare occurrence of the third rib bifidity (only in one individual, while modern literature states that this is the rib most often affected by this anomaly[10]). However, the often-unsatisfactory state of preservation of skeletal remains and the relatively small research sample that could have influenced the obtained result should be taken here into account.

Problems with creating a reliable inventory of known cases

Reliable review of the bifid rib in the bioarchaeological literature is a very complex problem. Most of the older original papers now function only locally as typescripts or extracts from inventory lists, and the newer publications that quote them, copy numerous errors and distortions in the interpretation of the studied anomaly. Therefore, in this work only direct reports with published photographic documentation or drawings of a given case were treated as unquestionably true. Only 13 such cases were collected[36,56,57,60,63,64,75–81], beyond four individuals published here for the first time. This group should be joined by another one from a museum collection of historical significance[22,82] (see Table 2; Fig. 8). A separate group consists of cases not confirmed by photographic documentation (14 individuals). Some of them are known only from short notes when describing the state of preservation of bones or existing skeletal pathologies of a given individual, e.g. in inventory lists[55,58,59,61,62,80,83,84] or from reviews[68,71,85]. Both of these groups were classified as "probable cases" (see Table 3; Fig. 8).

Despite a very thorough review of the available literature, there is also a risk of existing cases of bifid rib not listed in this work. This applies in particular to older papers (e.g[66,67,71,72]). , in which the authors describe cases known to them only in very general terms (e.g. as "coming from Egypt"), without giving specific citations or even specifying the name of the site, age and sex of the individual with the anomaly in question. This led to the risk of duplication of the presented cases, and currently it is impossible to verify the origin of some information about a given find, especially that over time some of the available descriptions have changed significantly (e.g. information about the sex of an individual) in subsequent works that quote each other. This was the case especially in anatomical collections, e.g. NMNH-H (from the National Museum of Natural History in Washington), published so far in many manuals – some of them include both photos and inventory numbers of individuals, some only photos and the general name of the collection they come from, while some provide only a general verbal description of the variation along with the name of the collection. This creates some confusion and the inability to confirm and organize the available data, which suggests that in the future there will be more reports of bifid rib cases not cited here.

Another obstacle to a reliable inventory of all known cases of bifid rib turned out to be a certain freedom in interpreting the definition of what this anomaly is. Some of the cases functioning in the literature as bifid rib, the authors of this work decided to reject, because they indicate other, currently known developmental pathologies of the skeleton. The problem here is also the multitude of synonyms describing Luschka's rib. Although the authors have tried to check all the terms used in the contemporary literature for forked rib (vd. *Introduction*), it is possible that some cases have not been identified by giving them rarely used local names.

The last one, the smallest but important group consisted of cases originally published as belonging to the more general group of developmental disorders of the ribs (without giving them a specific name), wrongly assigned by later authors to bifid rib, or those that were wrongly classified from the very beginning (cf. *Rejected cases*), which has not been verified by subsequent publications.

Bifid rib and Gorlin-Golt syndrome

It is important to note that the majority of currently published bioarchaeological papers link the presence of bifid rib with NBCCS, which is a definite abuse – as e.g. this research has shown, forked rib is an anomaly that occurs largely alone, and its presence should not be directly associated with such a serious genetic disease. Even though today bifid rib occurs in less than 3% of the population (cf. *Introduction*), while in patients with NBCCS – in as much as 26%[35,86], it should be strongly emphasized that the presence of bifid rib itself is not a pathognomonic feature of this condition. According to the data collected in this study, Gorlin-Goltz syndrome was found in four individuals with bifid rib, while the descriptions of coexisting anomalies allow to confirm it only in one (E225 by Satinoff[63,64]). Among those with rejected diagnosis are: (1) GED44, E22, drawer 400 by Boano et al.[79], (2) E1 by Capasso[56] (diagnosis made by other researchers) and (3) child from Vrolik collection By Oostra and Maas[22]. In the aforementioned works, the authors do not mention (Capasso) or mention only one (Boano et al., Oostra and Maas) minor criteria typical for NBCCS (according to Kimonis et al.[86]), i.e. "other vertebral anomalies", which is insufficient to make a reliable diagnosis and can at most indicate only general disorders of the ossification process of the axial skeleton. With the current advancement of science, genetic tests seem to be the only reasonable tool for the diagnosis of NBCCS.

Proposal of bifid rib classification in archaeological populations

Considering the above listed examples, it should be underlined that currently there are inconsistencies in the definition of what bifid rib is. Therefore, for the needs of bioarchaeological research, we propose the following

wording[87, changed]: *bifid rib is a rib that has a single head and body that divides or forks at the sternal end, connecting with two independent cartilaginous ribs*. Therefore, such a rib must have two independent surfaces to connect with the cartilaginous rib (in the bioarchaeological and clinical cases published so far, there is no information about the existence of bifurcations of false and floating ribs). This definition therefore excludes all other irregularities of ribs segmentations, e.g. holes in ribs, flared ribs, hyperplastic ribs, bicipital ribs, merged (fused) ribs, bridged ribs and all kind of spurs on ribs, although some authors treat some of them as the mildest form of bifurcation[68]. Separating the category of bifid rib understood in this way is correct as some of the mentioned anomalies (e.g. merged ribs and bridged ribs) arise as a result of the actual fusion of two initially independent ribs, while the bifid rib is a consequence of splitting the arch of a single rib[88]. The definitions assume the presence of spur ribs, flared ribs, and hyperplastic ribs are much closer to our definition of bifid ribs, since they are in fact a form of varying degrees of overgrowth of singular rib[9]. However, taking into account the descriptive nature of the existing definitions, we propose to separate bifid rib as an independent unit.

At least several bifid rib classifications have been proposed in the medical literature so far[7,9,18,89], but they also take into account the cartilaginous part of the ribs (cf. case presented in Mann and Hunt[87]), which makes it impossible to apply them to most osteological materials. Therefore, below we propose a classification dedicated to osteological materials, applicable in bioarchaeology. The following should be taken into account:

(1) Depth of notch between branches of the rib:

   a. No notch visible (only shaft extension and two articular surfaces are visible) – "F" (from flat).
   b. Notch visible but shallow (the length depth of the notch is less than its width) – "S" (shallow).
   c. Deep notch (the depth of the notch exceeds its width) – "D" (deep).

(2) Bifurcation shape:

   a. U-shaped – "U".
   b. V-shaped – "V".

(3) Symmetry of branches/articular surfaces:

   a. Symmetrical branches/surfaces (similar in size) – "S".
   b. Asymmetric branches/surfaces (visibly different in size) – "A".

According to the above classification, there is a hypothetical possibility of 12 combinations, but in practice the notch on a flat bifurcation type will always be a wide and shallow "U", so there will be 10 or less possible combinations (Fig. 9). Table 5 contains examples of the analysed ribs arranged according to the proposed classification. As can be seen, not all types of bifurcations have their representatives in the discussed osteological material. However, it is very likely that future publications will fill this gap.

Conclusions

The variant known as bifid rib is a very rare, usually asymptomatic developmental anomaly detected mostly accidentally during postmortem examinations or imaging tests used to diagnose other diseases. In the case of studies conducted on osteological human remains gained from archaeological excavations, estimating its actual frequency is even more difficult than in living populations, mainly due to general taphonomic processes and the greater sensitivity of the ribs to postmortem mechanical damage than other bones. An additional problem in building a coherent picture of the occurrence of bifid rib in bioarchaeological materials is the relatively large number of publications, which seem to significantly overestimate the frequency of this anomaly – as this research has shown, many of them do not meet the criterion of the correctness of the diagnosis. This study also proves that there is no basis to treat bifid rib as pathognomonic for NBCCS. Archaeological data most often show this anomaly as unrelated to other bone pathologies, which on the one hand confirms clinicians' assumptions about its source in laminar and costal sclerotome derivatives disorders, and on the other – should alert researchers to too hasty diagnosis of rare diseases in bioarchaeological populations.

The definition and classification of the anatomical variety known as bifid rib proposed in this article will not only help avoid the above-mentioned errors in future bioarchaeological analyses, but also organize the description of its variability. At the same time, it may constitute a reference for future research in the context of a set of features that should be given special attention when analysing developmental changes in ribs.

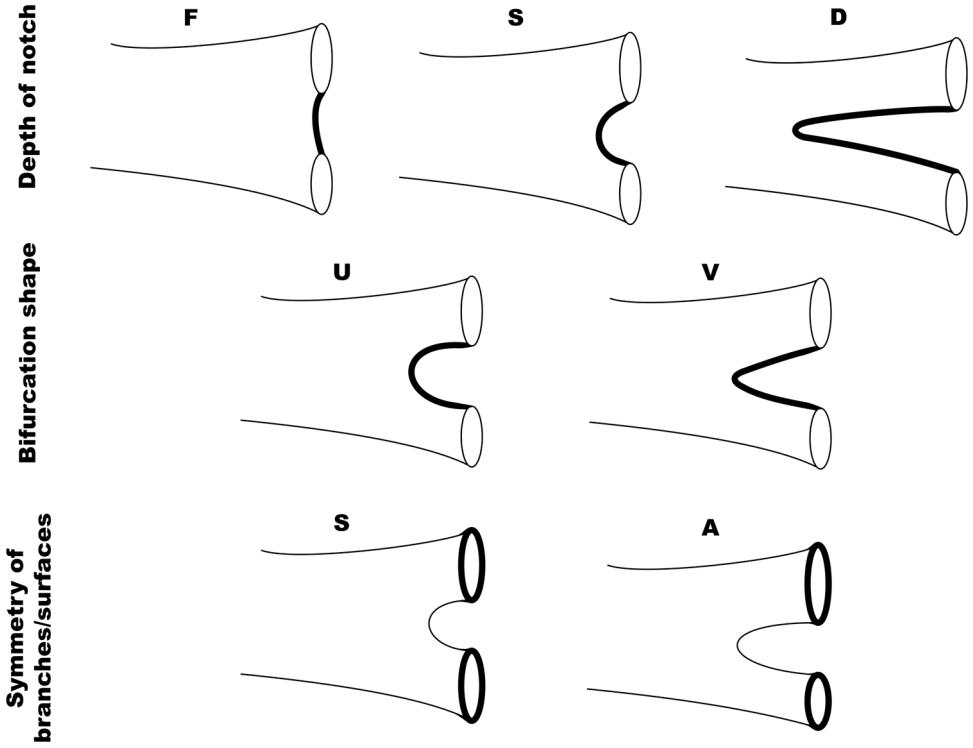

Fig. 9. Proposal of a classification of rib bifurcations in archaeological material (description provided in the text).

O.n.	Type of bifurcation	Description	Example from archaeological material
1	FUS	Flat, U-shaped notch with symmetrical branches	BK4_36 (this study), E225 (Satinoff[64]; Satinoff and Wells[63]), Płock_20 (this study), SBK4_664 (this study)
2	FUA	Flat, U-shaped notch with asymmetrical branches	Individual from grave 1 (Šikanjić et al.[35])*
3	FVS	Flat, V-shaped notch with symmetrical branches	N/A
4	FVA	Flat, V-shaped notch with asymmetrical branches	N/A
5	SUS	Shallow, U-shaped notch with symmetrical branches	E1 (Capasso[56]), 3rd rib of SD_56 (this study)
6	SUA	Shallow, U-shaped notch with asymmetrical branches	Upper individual from grave S34 (Fily et al.[78])
7	SVS	Shallow, V-shaped notch with symmetrical branches	N/A
8	SVA	Shallow, V-shaped notch with asymmetrical branches	Individual from plaza, Feature 32 (Anderson[76])*
9	DUS	Deep, U-shaped notch with symmetrical branches	G4 (Bencerić[57])*, both 5th ribs of individual from Vrolik collection (Bolk[88]; Oostra and Maas[22]), 800/6 (Vymazalová et al.[60])
10	DUA	Deep, U-shaped notch with asymmetrical branches	PP B.609 (Kozieradzka-Ogunmakin[80])*, NW 70 (Kirkpatrick[81]), 4th rib of SD_56 (this study)
11	DVS	Deep, V-shaped notch with symmetrical branches	Individual published by Merbs[75]*, GED44 (Boano et al.[79])
12	DVA	Deep, V-shaped notch with asymmetrical branches	N/A

Table 5. Proposal of a classification of rib bifurcations in archaeological material according to morphology of sternal end of the costal rib (description provided in the text). N/A – data not available; * – the diagnosis is uncertain due to post-mortem bone destruction or poor quality of photo representing the bifurcation.

Data availability

Upon a reasonable request, the corresponding author will provide the data supporting the findings of this study.

Received: 4 June 2024; Accepted: 13 November 2024
Published online: 03 December 2024

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Acknowledgements

We would like to thank Iwona Kozieradzka-Ogunmakin for sharing with us all the unpublished results of her research on the population from cemeteries at Saqqara-West, Egypt. Similarly, we would like to thank Theya Molleson for providing additional information about the individual Tr.A72.338D from Syria. We would also like to thank Meredith Vasta, Collections Steward from Peabody Museum Research Office (Peabody Museum of Archaeology and Ethnology, Harvard University), Bill Burkett, the editor of Series "The Arizona Archaeologist",

Ellie Large, the president of the Arizona Archaeological Society, Ryan Tubbs (Division of Human Anatomy at Michigan State University) and Casey L. Kirkpatrick (Max Planck Institute for Evolutionary Anthropology) for their invaluable help in collecting the necessary literature.We would also like to express our gratitude to Corrado Protasi and Michela Miceli (Laboratori Campisi, Avola, Sicily and, previously, Centro Polidiagnostico "San Luigi", Rosolini, Sicily) for their technical assistance with the radiological procedures.

A preliminary version of this study was presented at the 76th congress of the Italian Society of Anatomy and Histology (SIAI) held in Modena (Italy), 11–13 September 2023.

Author contributions

J.M.S.: conceptualization, investigation, methodology, visualization, writing – original and revised draft; E.V.: imaging, investigation, visualization, writing – revised draft; E.Ż.: critical revision of the first and revised draft, investigation; W.L.: critical revision of the revised draft, investigation, supervision, writing – original draft; F.M.G.: investigation, supervision, writing – original and revised draft.

Declarations

Competing interests

The authors declare no competing interests.

Ethical standards

For this study the authors followed the Polish national regulations and laws for the analysis of archaeological human remains. Permission to study the remains was issued by the directorate of the Department of Anthropology and by the Faculty of Biology and Environmental Protection of the University of Lodz (Poland).

Informed consent

The present study does not contain information or images that could lead to identification of a study participant. The investigated osteological material is of an archaeological nature, therefore patient consent is not applicable.

Additional information

Correspondence and requests for materials should be addressed to J.M.-S.