CASE REPORT
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A case of cervical rib and neurovascular compression in Roman period

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SUMMARY

The possibility to study axial anomalies directly on a skeletal individual is not very frequent. One well preserved skeletal individual from an Italian site dating to the late antique period (5th -4th centuries CE) was studied. This individual shows some interesting skeletal changes in the vertebrae and ribs. A supernumerary rib was found. It is a cervical rib connected to the 1st thoracic rib, presumably with a fibrous bundle. The presence of cervical ribs can produce neurovascular compression of the brachial plexus and subclavian vessels. Because of this, it is often a cause of thoracic outlet syndrome (TOS). In our case the presence of a cervical rib articulated with the first thoracic rib through a probable fibrous band could have restricted the space where the brachial plexus and subclavian vessels pass through, creating a state of neurovascular compression, similar to what happens in costo-clavicular TOS.

Key words: Supernumerary ribs – Neurovascular involvement – Thoracic outlet syndrome – Roman period

INTRODUCTION

Between the axial congenital anomalies of the mammalians there are the supernumerary (or accessory) ribs (SNR) that can be either lumbar ribs (LR) or cervical ribs (CR). The biological significance of SNR is very problematic and debated. For example, in mouse models the spontaneous incidence of SNR is related to their species and ranges from <1% to >30%. Among humans, CRs have a low frequency, about 0.2-1% (Guttentag et al., 1999; Chang et al., 2013) and show variation in prevalence between different ethnic populations (Brewin et al., 2009). The first to describe CR in humans was Gruber, a German physician who proposed a classification system (the Gruber scale) that is still utilized (Gruber, 1869; Chang et al., 2013): 1) CR extending just beyond the transverse process; 2) CR extending beyond the transverse process with a free tip almost touching the 1st thoracic rib; 3) CR extending beyond the transverse process with fibrous bands or cartilage attaching to the 1th thoracic rib; 4) CR completely fused to the first rib. The most common location for CR is on the 7th cervical vertebra (C7), that becomes morphologically similar to a thoracic vertebra due to homeotic transformation in which the Hox genes are involved (Mallo et al., 2009). In mouse models both inactivation and ectopic expression of Hox genes can produce transformations of skeletal phenotypes. According to these experimental observations the Hox genes located at the 3’ end of the clusters (termed “anterior” Hox genes on the basis of their expres-

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sion domains) are involved in the control of anterior structures, while those located towards the 5' end (termed "posterior" Hox genes) are responsible for the control of posterior vertebral morphology (Mallo et al., 2009). Furthermore, in mouse models it has been shown that the largest sizes of cervical ribs are present in mice mutants homozygous for a Hoxa-5 mutation compared to heterozygotes Hoxb-8 (Charité et al., 1995; Bots et al., 2011). Another study on mice demonstrated that nutritional, pharmacological, and environmental stress during the pregnancy produce disregulation in the Hox genes involving Hoxa-5, Hoxb-8, Hoxa-9 and Hoxc-10 with presence of supernumerary ribs (Kim et al., 2015). In humans, CRs occur in various dimensions such that homeotic transformations are often incomplete (Varela-Lasheras et al., 2011). Until a few years ago, it was believed that the CRs observed in fetuses were ossification centers that fuse with the transverse process and do not persist in adults (O'Rahilly et al., 1990). In recent times, it was demonstrated that supernumerary ribs do not disappear after development (Galis, 2006; Bots et al., 2011). Although clinical symptoms may not present until adulthood due to the growth of the rib (Black and Scheuer, 1997), they can appear in relation with pathologies of different origin and severity (Steigenga et al., 2006). In many cases, fetuses and infants with CRs are affected by early childhood cancers (Bots et al., 2011), but another association is thoracic outlet syndrome (TOS). In TOS cervical ribs may compress the portions of the brachial plexus and in severe cases, the subclavian vessels (Roos et al., 1999, Brewin et al., 2009). According to some authors, the low frequency of CRs in adult individuals with respect to infants is due to the early mortality of individuals with negative pleiotropic effects (Galis et al., 2006). The aim of this study was to investigate and describe a case of CR in an individual from a Roman period settlement in northern Latium (Viterbo, Central Italy).

MATERIALS AND METHODS

The skeletal remains come from a large Roman site (Graffignano, Viterbo, Central Italy), where a group of four tile-covered ("cappuccina") tombs were found. The burials have been dated to the late antique period by the presence of tile-stamps dating to 4th -5th century CE.

The skeleton termed PLG2 was in a good state of preservation. The state of preservation was quantified in accordance with Bello et al. (2006), who consider three indices: the anatomical preservation index (API), the qualitative bone index (QBI), and the bone representation index (BRI). In PLG2, about 85% of the bones are present (BRI) and their cortical surfaces are in a good state (based on the first two indices, API and QBI= class 4). The age at death and sex assessment were made according to the international standards of Buikstra and Ubelaker (1994). The age at death was estimated on the degree of fusion of the spheno-occipital synchrondrosis -sutura sphenooccipitalis- (Gray, 2000), on the morphological changes of the pubic symphyseal face (Brooks and Suchey, 1990) and on the modifications of the auricular surface of the os coxae (Lovejoy et al., 1985; Meindl and Lovejoy, 1989). These last two parameters are the most used in forensic and anthropological analysis, because they produce good results (Martrille et al., 2007). Moreover, the fourth rib was also considered (Schmitt et al., 2006). The sex assessment was made on the discriminant morphology of the os coxae (Buikstra and Mielke, 1985; Krogman and Iscan, 1986) and skull (Acsadi and Nemeskeri, 1970).

RESULTS

The individual PLG2 is a female of about 30-40 years old.

Description of the pathological changes on PLG2

Vertebrae – The thoracic region shows interesting changes in the costo-vertebral and sternocla-

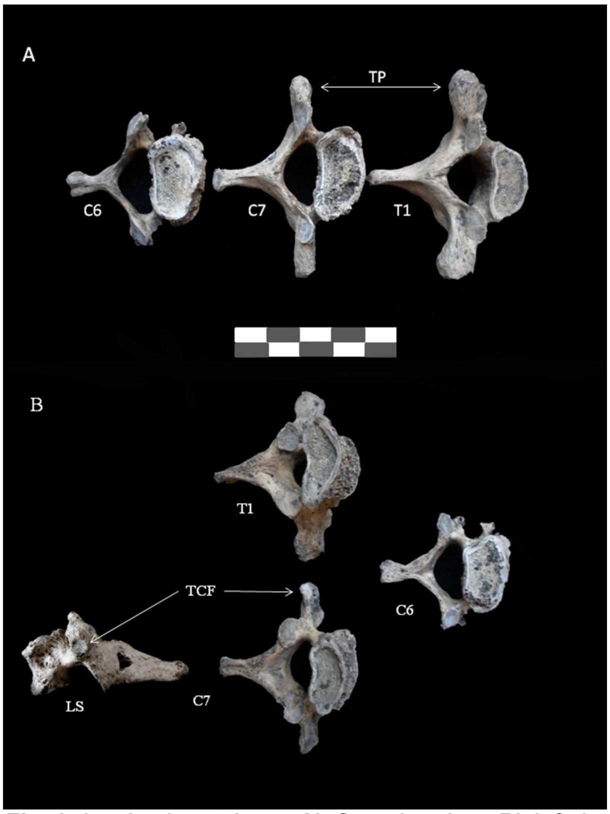

Fig 1. Involved vertebrae: **A)** Superior view; **B)** Inferior view. C6= sixth cervical vertebra. C6 shows an important osteophytosis along the left edge of the body; C7= seventh cervical vertebra. C7 displays an absence of the transverse foramen, with the transverse processes (TP) elongated and similar to those of the T1; T1= first thoracic vertebra; TP= transverse process; TFC= Transverse costal facets; LS= Left side.

Fig 2. A) Cervical rib (CR) and first ribs with the sternum. DE=Distal end of the CR with roughness of the concavity for the insertion of the fibrous bundle and ostephytosysis of the edge; **B)** Reconstruction of the articulation between the cervical rib and first rib; **C)** 3D pattern of the fibrous bundle that articulates the two ribs. IA=insertion area.

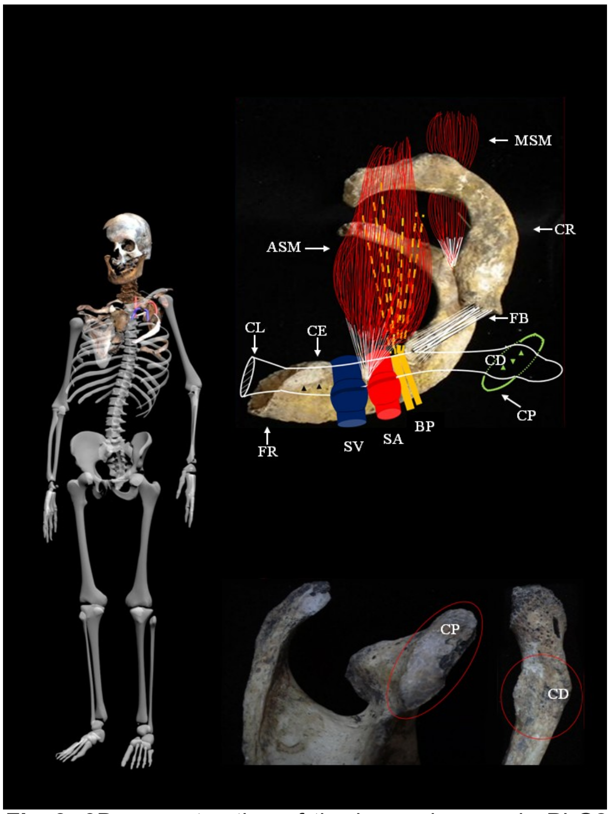

Fig 3. 3D reconstruction of the bone changes in PLG2 skeleton. MSM= Middle scalene muscle; ASM= Anterior scalene muscle; CR= Cervical rib; FB= Fibrous bundle; CP= Coracoid process; CD= Clavicle dilatation by contact with CP; CL= Clavicle; CE= Contact erosion (pseudofacet) 1st rib and clavicle; FR= 1st rib; SA= Subclavian artery, SV= Subclavian vein; BP= Brachial plexus.

vicular region. The 7th cervical vertebra (C7) displays an absence of the transverse foramen, while the transverse processes (TP) are elongated, with a morphology similar to those of the 1st thoracic vertebra (T1) (Fig. 1A). Furthermore, C7 shows the asymmetrical bilateral presence of transverse costal facets (TCF): the right facet is in the regular position, while the left is on the inferior surface of the transverse process (Fig. 1B). On the left side of the vertebral body there is a superior costal facet (Fig.1B, LS) that articulates with the head of the cervical rib.

Ribs – A supernumerary left cervical rib is present and in articulation with the C7. This rib is similar in morphology to a 1st thoracic rib (Fig. 2A), but with a smaller size (length 62.5 mm). The inferior surface shows vascular and muscular prints that normally are present on the superior surface of the 1st thoracic rib. The distal end is not fused with the 1st thoracic rib, but it shows a functional morphology with concave surface probably due to a fibrocartilaginous insertion and roughening of the edge caused by mechanical strain (Fig. 2A). The superior body of the left 1st thoracic rib presents an insertion area (tubercle) that probably was in articulation through a fibrous band with the cervical rib (Fig. 2B, C). Also a contact facet with the clavicle on the upper edge of the costal epiphysis is present. The distal end of the 1st left thoracic rib is larger than the right. Close to the distal end of the rib, the imprint of the subclavian vein is very evident with a large depression that could include also the subclavian artery. The 1st right thoracic rib is ill-formed shaped similar a 2nd thoracic rib with the absence of the muscular and vascular prints (Fig. 2A).

Clavicles – Both clavicles show entheseal changes at the attachment site for the costo-clavicular ligament, more accentuated on the left. They have a flattening (with dilatation) close to the conoid to which on the inferior surface corresponds a contact facet with the coracoid process of the scapula (Fig. 3).

DISCUSSION AND CONCLUSIONS

The PLG2 female individual shows an axial anomaly, the presence of a cervical rib. The etiology of the cervical rib, although not yet completely known, results from a malfunction of the Hox genes. What is not yet known in humans is whether the negative state of general health of the mother during early pregnancy can influence the behavior of the Hox genes (Boots et al., 2011), as seen

Table 1. Summary of the compression sites in the costo-clavicular space between the first thoracic rib and the clavicle and between the coracoids process and clavicle

Skeletal region	Anomalies PLG2	Anatomical variants PLG2	TOS
Vertebrae	Elongated C7 transverse processes	Presence of an incomplete articular costal facet on the inferior surface of the transverse process	
	T1 left transverse process without articular facet		
Ribs	Left cervical rib presence	Presence on the inferior surface of the body of cervical rib of a longitudinal transverse groove. Presence of a functional “sternal end” with concave surface (due to fibrocartilaginous insertion) and mild degenerative changes	The transverse groove indicates a muscular contact (anterior, middle and minimum scalene)
	First left thoracic rib without tubercle	Presence of a circular roughness with entheseal changes associated to the muscular insertion (tubercle) near the margin of the superior surface of left 1st thoracic rib. This “tubercle” could represent traces of fusion or insertion of a fibrocartilaginous bundle.	The fibrous bundle indicates the connection between the cervical rib and first thoracic rib. This bundle could narrow the space and compress the brachial plexus
		Presence of contact facet in the edge of the superior surface of the 1st thoracic rib	Erosion indicates contact between the rib and the region near the sternal end of the clavicle. By lowering and pressing back the clavicle, the subclavian vessels are compressed
	Absence of the tubercle of the anterior scalene muscle	Presence of a large depression on the superior surface of the firs thoracic rib (it can include both the subclavian artery and vein)	Depression can indicate a large poststenotic vascular dilatation
Acromioclavicular joint		Presence of flattening and dilatation in the area of insertion of the coracoclavicular ligament (between the coracoid process and the clavicle)	Coracoclavicular dilation is a compression anomaly due to ligamentous changes with narrowing of spaces and contact between the various joint components.
		Presence of contact facet of the superior surface of the coracoid process	
		Presence of an accentuated articular on the lateral end of the clavicle	

in mouse models (Kim et al., 2015). According to some authors CRs are more frequent among females (Palma and Carini, 1990; Gulekon et al., 1999) and in white populations (Brewin et al., 2009). The CRs could be complete or incomplete. They are complete when they are directly in articulation with the breast, while they are incomplete when they are smaller and often are in articulation with the 1st rib. In our case, the morphology of the supernumerary rib suggests that it is incomplete and could be classified as type 3 on the Gruber scale (see Introduction and Table 1). The articulation with the 1st thoracic rib indicates a position able to compress nerves and vascular structures as happens in TOS (Chang et al., 2013) (Fig. 3). The CRs are an important cause of neurovascular compression (Brewin et al., 2009). In fact, the trunk of the brachial plexus and the subclavian artery pass through the interscalene triangle toward the axilla. A cervical rib may articulate with the 1st thoracic rib in the base of this triangle through a fibrous or osseous connection. This connection may narrow the space through which the neurovascular structures can pass (Brewin et al., 2009). In our case there is an area of erosion, located on the upper edge of the sternal end of the 1st thoracic rib. This erosion may be related to the dilatation of the clavicular body near the conoid tubercle (achromial direction) corresponding to a flattening of the superior surface of the coracoid process of the scapula. These anatomical changes show the presence of different points of contact with a narrowing of the costo-clavicular space (Fig. 3). This aspect may suggest neurovascular impingement involving the subclavian vessels and the brachial plexus in PLG2 individual. In TOS three anatomic levels of neurovascular compression are documented:

Scalene triangle called scalene syndrome. The anterior and middle scalene muscles with the 1st thoracic rib are involved.

Costo-clavicular space called cervical rib syndrome. The clavicle and the 1st thoracic rib are involved.

Sub-coracoid space called hyper-abduction syndrome. The pectoralis minor, the 2nd thoracic rib and the coracoid process are involved.

TOS is a condition characterized by pain and paresthesia in the shoulder and arm. In addition, it can produce vascular signs such as edema and/or upper limb fatigue. These disorders originate from a compression of the vascular and/or nervous structures that travel from the mediastinum to the upper limb. The compression of the brachial plexus (and of the subclavian vessels) at the point of passage of these structures into the thoracic strait can be caused by malformations such as accessory ribs and scar results after trauma. Other conditions, such as peripheral neuropathic entrapment, neoplasms, and cervical spine and shoulder disorders, are often considered in the differential diagnosis. In PLG2, the presence of a cervical rib with evident signs of alterations in vascular prints and in articular facets associated with an absence of trauma makes these last factors more likely to be a cause of a possible TOS (Table 1) that involve the costo-clavicular space and probably also the scalene muscle triangle (points a and b). Undoubtedly, besides a possible syndrome, PLG2 suffered from a neurovascular compression, which, as mentioned above, produces a series of disorders in the upper limbs during the life. These disorders may have affected the individual as, precisely because of compressive phenomena, the upper limbs were easily tired out after short activity. In conclusion, our case seems to have all the skeletal features of a cervical rib associated with TOS a syndrome that is relatively invalidating but compatible with life.

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