Cervical rib in a young individual from the late medieval cemetery of Corfinio (XII–XIII centuries CE, Italy): a case report and review of the literature

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With 7 figures and 1 table

Abstract: Cervical ribs constitute the most important variants among ribs, particularly because of their proximity to the brachial plexus nerve network and subclavian artery. Thus, cervical ribs may exert excessive pressure upon these structures, producing a variety of symptoms that may be considered as a neuro-vascular complex. Cervical ribs are not uncommon in the catalogue of anatomic abnormalities in modern medicine, but few cases have been reported from historic skeletal material. We conducted a review with the existent articles published in palaeopathological literature, the historical background with the most important milestones regarding the studies on cervical ribs, the embryological processes in their development, and their prevalence and principal clinical features. Finally, we described a case of a cervical rib that was observed in the skeletal remains of a young individual from the late medieval cemetery of Corfinio (XII–XIII centuries CE, Italy).

Keywords: supernumerary rib; congenital anatomical variant; thoracic outlet syndrome; archaeology; palaeopathology; *Corfinium*

Introduction

Anatomical 'variant' is defined as the normal flexibility of size, form, structure and position of body structures (Sañudo et al., 2003). They represent a small departure of the normal presentation, and therefore, usually do not require therapeutic treatments in the clinical setting; however, they may become symptomatic under certain circumstances (Georgiev, 2017). Thus, anatomical variants represent an embryological and comparative framework for disciplines such as medicine and life sciences, whose objectives are to understand the morphological properties of the human body and its related structures (Sañudo et al., 2003; Viciano et al., 2017b). According to Hennekam et al. (2013), an anatomical structure is defined as a 'variant' when it is within the normal range of variation (i.e., when the prevalence is between 2.5% and 10% for an appropriate reference population), while it is defined as an 'anomaly' when it refers to a particular abnormality found in a small proportion (i.e., a prevalence < 2.5%).

The thoracic cage of the human skeleton consists of 12 pairs of ribs with their costal cartilage and the sternum (White et al., 2011), but this can vary by either agenesis or the presence of supernumerary bone elements (Black & Scheuer, 1997; Capasso et al., 1999). Congenital agenesis of the ribs is relatively a rare condition (Chen, 2007; Shalaby & Elnagdy, 2020; Zhang & Wang, 2018); however, the presence of supernumerary ribs in both the cervical and lumbar regions of the spine is well-reported in the literature (e.g., Anderson, 1996; Barnes, 2012; Black & Scheuer, 1997; Brintnall et al., 1956; Capasso, 2001; Chengetanai et al., 2017; Cumming, 1926; Eaves-Johnson, 2010; Hertslet & Keith, 1896; Kammerer, 1901). The presence of supernumerary ribs is considered a phenomenon of atavism due to the general evolutionary tendency towards the reduction in the number of ribs in humans and higher primates (Black & Scheuer, 1997; Gladstone & Wakeley, 1932; Jones, 1913). This evidence is based on the fact that vertebrate animals that develop limbs require greater mobility in the region of the pectoral and pelvic girdles; therefore, the presence of supernumerary ribs in these locations could limit the flexibility and mobility of these regions (Aiello & Dean, 2002; Black & Scheuer, 1997; Scheuer & Black, 2004). For this reason, supernumerary ribs are considered indicators for disadvantageous developmental events and, therefore, are subject to strong negative selection during the evolutionary process (Furtado et al., 2011; Galis, 1999a; Galis et al., 2006). In contrast, limbless vertebrate animals, such as snakes, retain two ribs associated with each vertebra (Adson & Coffey, 1927; Davis & King, 1938; Jones, 1913). Jones (1913, 1911) proposed that the morphology and mechanical pressure of the brachial plexus on developing ribs influence the formation of cervical ribs, preventing or limiting their growth; however, Todd (1911) did not agree with this proposition and stated that the brachial plexus alone is insufficient for the presence of cervical ribs. On the other hand, Davis & King (1938) suggested that the development of cervical ribs was constrained by a distinctive trajectory of spinal nerves. Be that as it may, as Black & Scheuer (1997) noted, if these were the possible explanations for the presence of cervical ribs, one would also expect to find ribs in the upper cervical vertebrae. However, as highlighted by the same authors, in the literature, there are no reported cases of supernumerary ribs from first to the fourth cervical vertebrae. Thus, these hypotheses based on anatomical and mechanical factors associated with the brachial plexus and spinal nerves are currently controversial and not widely accepted.

We describe a case of a cervical rib that was observed in the skeletal remains of a young individual from the late medieval cemetery of Corfinio (XII–XIII centuries CE, Italy), and we briefly review the existent articles published in palaeopathological literature, the historical background with the most important milestones regarding the studies on cervical ribs, the embryological processes in their development, and their prevalence and principal clinical features.

Historical background

The most important milestones regarding the studies on cervical ribs are briefly described in Table 1. The initial description of a cervical rib apparently dates back to the Roman period (Roos et al., 1999). Other cases have been reported by Andreas Vesalius in the XIV century and by J.F. Hunauld in the XVIII century (Roos et al., 1999). Since the XIX century, clinical studies have described cases of cervical ribs and correlated this anatomical variant with neurovascular symptoms (e.g., Cooper, 1818; Peet et al., 1956).

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Review of the palaeopathological literature

Hrdlička (1899) described the anomalous skeleton of an adult male individual from the Valley of Mexico, belonging apparently to the Aztec period. The skeletal remains showed a pair of cervical ribs articulating with the seventh cervical vertebra and concluded that all the thoracic structures (the blood vessels included) were elevated, assuming the supernumerary ribs the anatomical relations which ordinarily belong to the first thoracic pair. Barclay-Smith (1911) provided a detailed report of multiple abnormalities in the vertebral column of a young adult female skeleton from excavations at Sakkara in Egypt (dated to a period between 600 and 500 BCE). In this specimen, the neural arch of the seventh cervical vertebra was divided by the spinous process, and an extra cervical vertebra (eighth cervical vertebra) had an associated small cervical rib on the right side. MacCurdy (1923) described an adult male skeleton from Peru, in which there was a supernumerary rib associated with hemivertebrae of the right side. Denninger (1931) provided a detailed description of a specimen from a prehistoric Indian mound in Illinois. The skeleton belonged to a young adult male, in which the body and arch of the seventh cervical vertebra showed winged rib-like processes. On the right side, the supernumerary rib was fused with the transverse process and body of the vertebra. On the left side, the cervical rib was roughly L-shaped and was not fused with the body of the vertebra, but it possessed two distinct articular facets that corresponded with the similar ones on the left side of the body and transverse process of the vertebra. Denninger concluded that the cervical rib was of sufficient length to come into contact with the left brachial plexus, basing his observation on the fact that there were two smooth grooves transversely crossing the inner and upper sides of the body the rib near its anterior end. The latter was diffusely pitted and tapered in such manner to suggest that a fibrous cord or tendon connected this rib with the sternum on the right. Angel (1946) noted a left cervical rib in an ancient Greek skeleton, but without a detailed description of this anatomical variant. Finnegan (1976) reported a case of a cervical rib with associated disuse bone atrophy in an Archaic American male aged 45–55 years from the Ken Caryl Range, Colorado (dated 490 BCE). Wells (1979) reported one case of a female Anglo-Saxon dwarf from a northern English monastery who also had a cervical rib, suggesting its possible association with atrophy of her arm. Anderson (1996) published a short note addressed to archaeologists, with special emphasis on the importance of carrying out an excavation of the skeletal remains avoiding subsequent damage for the correct analysis by the osteologist, presenting photographs of several skeletal abnormalities. Of the 13 cases presented, one of them referred to a cervical rib and another referred to a lumbar rib. Black & Scheuer (1997) reported a case of a cervical rib from the post-medieval St. Bride's documented skeletal collection (London). Capasso (2001) reported three cases of cervical ribs in the seventh cervical vertebra from the victims who died while trying to flee by sea from the ancient beach of Herculaneum (Naples, Italy), during the eruption of Mount Vesuvius on 24–25 October 79 CE. Individual E40 (male individual aged 30–35 years) presented with a left cervical rib fused with the first thoracic rib; individual E57 (female individual aged 40–45 years) showed a bilateral cervical rib, with the right one articulating with the first thoracic rib; and individual E65 (female individual aged 45–50 years) had a left cervical rib. The same author also described a case of a left supernumerary lumbar rib anchylosed to the transverse apophysis of the first lumbar vertebra (E52, female individual aged 20–25 years). Jankauskas (2001) noted three cases of cervical ribs in recent archaeological excavations in Lithuania, but without a detailed description of this anatomical variant. Nagar (2002) observed a seventh cervical vertebra in an adult individual (over 50 years old) manifesting as a bony extension from its left transverse process, which was interpreted as a cervical rib. The skeletal remains of this individual, dated to the Roman period, were found in the excavation of Cave VIII/28 (‘Cave of the Sandal’) in the northern Judean Desert. Fernandes & Costa (2007) analysed the skeletal remains of an adult male exhumed from a Portuguese graveyard dating from the XIII to XV century, showing several malformations in the cranium and spine corresponding to Klippel-Feil syndrome. Among the skeletal abnormalities, the researchers observed a rudimentary cervical rib in the left transverse process of the seventh cervical vertebra. Lewis (2013) observed a possible cervical rib in the skeletal remains of a young individual (16 years of age) in England. Piombino-Mascali et al. (2017) observed a large cervical rib in a mummy held in a crypt beneath the Mother Church of Piraino, Italy. This cervical rib started from the seventh cervical vertebra with synostosis to the transverse process and reached the sternum. Rubini et al. (2019) described an adult female skeleton, aged 30–40 years, from an Italian site dating to the Late Antiquity period (IV–V centuries CE), in which there was a cervical rib articulated with the first thoracic rib through a probable fibrous band. Partiot et al. (2020) provided a detailed report of 43 unilateral and bilateral cervical ribs in 27 deceased individuals between 24 weeks of amenorrhoea and 2 years of age from the 8B-51 necropolis of Saï Island (Sudan), dated to the Classic Kerma period between XVIII and XVI centuries BCE. Finally, there are several theses and archaeological and anthropological reports (e.g., Caffell & Holst, 2012; Connell & White, 1998; Dinwiddy, 2009; Malnasi, 2010) that show the presence of cervical ribs. However, these reports make neither a brief nor detailed description of this skeletal variant, as they simply record their presence in the skeletal individual.

Cervical ribs are a common modern clinical finding (Brewin et al., 2009; Checa, 2020; Spadliński et al., 2016; Viertel et al., 2012; Walden et al., 2013). However, it seems to be evident that the few cases of cervical ribs so far reported in the palaeopathological literature might be due to several factors, such as (i) scarce attention to this anatomical variant in past years, (ii) the inadequate state of preservation of skeletal remains, (iii) the loss of the cervical rib during storage, or (iv) its easy confusion with the eleventh or twelfth thoracic rib (especially if the ribs are damaged) (Partiot et al., 2020; Waldron, 2009). In the latter case, Waldron (2009) suggests that, if it seems that there are 13 ribs, the seventh cervical vertebra should be examined for an articular facet, since cervical ribs are more common than 13 thoracic or lumbar ribs. Thus, although the rib may be damaged or may not be recovered, a costal facet on the vertebra is evidence for its presence.

Embryology

Several authors have suggested that cervical ribs result from a secondary ossification centre that appears within the cartilaginous costal elements of vertebrae around the sixth month of gestation (Black & Scheuer, 1997; Meyer, 1978) and remain separate until the age of 10 years (Black & Scheuer, 1997). During the development of the embryo, lateral costal processes form in each of the vertebrae along the entire spine (Donahue, 2013). However, only in the thoracic region do these processes separate from the developing vertebral mass and continue to grow to develop into ribs, whereas in the rest of the spine, they fuse with the vertebrae to become transverse processes (Cave, 1975; Donahue, 2013; Scheuer & Black, 2004; Sherk, 2005). Abnormal development of these costal processes is a common condition at the seventh cervical vertebra, resulting in either a cervical rib or an elongated transverse process that extends beyond the transverse process of the first thoracic vertebra (a condition known as ‘transverse apophysomegaly’) (Donahue, 2013; Galis, 1999a; Merks et al., 2005); nevertheless, it can also be found less frequently associated with the fifth and sixth cervical vertebrae (Adson & Coffey, 1927; Barnes, 2012; Donahue, 2013; Hrdlička, 1899; Scheuer & Black, 2004). Currently, this hypothesis on the development of cervical ribs is the focus of debate and it is not widely accepted by the scientific community (Bots et al., 2011; Galis et al., 2006; Partiot et al., 2020).

Recent investigations suggest that the presence of cervical ribs is caused by a disturbance in the *Hox* gene expression during early development (Böhmer et al., 2018; Furtado et al., 2011; Galis et al., 2006; Partiot et al., 2020). This hypothesis is based on the fact that, in all vertebrates, *Hox* genes are key regulators in the patterning of the axial skeleton, involving the development of each vertebral element into a specific type of vertebra (i.e., cervical, thoracic, etc.) and development of the nervous system (Mallo et al., 2010; Wellik, 2009). A group of 40 highly conservative evolutionary transcription factors in four clusters (*HoxA*–*HoxD*) distributed in different chromosomes control the *Hox* genes (Oostra et al., 2005). Thus, disruptions in *Hox* gene expression often present as abnormalities of the spine, including cervical ribs (Böhmer et al., 2018; Bots et al., 2011; Galis, 1999a, 1999b; Oostra et al., 2005; Wellik, 2009). According to Furtado et al. (2011) and ten Broek et al. (2012), cervical ribs would be the result of a homeotic transformation leading to a change in the phenotype in which the identity of the seventh cervical vertebra is transfigured into a rib-bearing thoracic vertebra.

Epidemiology and clinical significance

Several studies show that the prevalence of cervical ribs is extremely higher in deceased foetuses and diseased children than in adults. These studies report high prevalence rates in foetuses (ranging from 19% to 63%) (Bots et al., 2011; Chernoff & Rogers, 2004; Furtado et al., 2011; Galis et al., 2006; McNally et al., 1990; Partiot et al., 2020); however, much lower prevalence rates have been found in adults (ranging from 0.03% to 8.0%) (Brewin et al., 2009; Furtado et al., 2011; Galis et al., 2006; McNally et al., 1990; Walden et al., 2013). Several studies state that the significant discrepancy between foetal and adult prevalence would be explained by the strong selection against the appearance of these anatomical variants as a consequence of a homeotic transformation (see 'Embryology' section), based on the extremely high prevalence of cervical ribs found in foetuses who die before or around birth and in infants before reaching the age of 1 year (Bots et al., 2011; Furtado et al., 2011; Galis et al., 2006; Partiot et al., 2020).

Currently, the prevalence of cervical ribs in adults is uncertain as a result of the difficulty of detecting this congenital variant in clinical medicine through routine radiology (Spadliński et al., 2016; Viertel et al., 2012). Research has reported a wide variety for the prevalence rates in different populations, from 0.05% in the US population (Steiner, 1943) to 3.0% in the Anatolian population (Gülekon et al., 1999). However, recently, Erken et al., (2002) reported a prevalence of 6.2% in a population sample from Turkey, but the largest conducted studies in largely Caucasian populations produced frequencies in the range of 0.05% to 0.54% (Bokhari et al., 2012). Although this discrepancy of prevalence rates in adults may represent real differences between populations, part of the variation may be a consequence of the sex of the individuals studied, the sample size, the imaging techniques used for diagnosis (Brewin et al., 2009) and the discrepancy about the definition of what constitutes a cervical rib (detailed explanation in 'Classification system' section).

Cervical ribs are usually associated with the seventh cervical vertebra and are more common in females than males, in a ratio from 2:1 (Bokhari et al., 2012; Brewin et al., 2009; Viertel et al., 2012) to 3:1 (Gülekon et al., 1999) in adult individuals; in foetuses, an equal sex prevalence is observed (Furtado et al., 2011; McNally et al., 1990). It is present bilaterally in 40.3% to 80.0% of cases in adults (Adson & Coffey, 1927; Bokhari et al., 2012; Gülekon et al., 1999; Viertel et al., 2012) and between 59% to 78.3% in foetuses (Bots et al., 2011; Furtado et al., 2011; Galis et al., 2006; McNally et al., 1990), but the two sides are often asymmetrical (Du Toit & De Muelenaere, 1982). When unilateral, it is more frequently on the left side than on the right side in adults and foetuses (Adson & Coffey, 1927; Bots et al., 2011; Brewin et al., 2009; Furtado et al., 2011; Galis et al., 2006). Although the aetiology is uncertain, there is some evidence of a familial connection, and therefore, a tendency towards a genetic predisposition, reporting autosomal dominant transmission with variable expression (Schapera, 1987; Weston, 1956).

The presence of cervical ribs commonly causes the development of thoracic outlet syndrome in adults (Chang et al., 2013), a condition characterized by compression of and diminished blood flow in the subclavian vessels and carotid arteries and alteration of the position of the stellate ganglia, sympathetic ganglia, and seventh cervical and first thoracic nerve roots (Chernoff & Rogers, 2004); however, this condition is poorly described in childhood (Haroun, 2016). In clinical medicine, about two-thirds of cervical ribs in adults are asymptomatic and are identified on radiographs obtained for unrelated reasons. The ribs may continue to grow until the age of about 25 years, so symptoms from cervical elements may not be present until late adolescence or even early adulthood (Black & Scheuer, 1997). The symptomatology of thoracic outlet syndrome is of vascular and neurological origin. The vascular effects include cerebral and distal embolism (Bearn et al., 1993; Jusufovic et al., 2012; Naz & Sophie, 2006), whereas neurologic symptoms include extreme pain, migraine and Parkinson's disease (Evans, 1999; Fernandez Noda et al., 1996; Saxton et al., 1999). The presence of cervical ribs is a characteristic of the Klippel-Feil syndrome, also known as synostosis of the cervical spine (Fernandes & Costa, 2007; Tubbs et al., 2006). This disorder results in the congenital fusion of two or more cervical vertebrae. A strong relationship between the presence of axial skeletal abnormalities and cancer has been reported in several studies (Anbazhagan & Raman, 1997; Bots et al., 2011; Galis, 1999a; Henry et al., 2018; Loder et al., 2007; Merks et al., 2005; Schumacher et al., 1992; Zierhut et al., 2011), indicating a highly significant association with rib abnormalities, particularly cervical ribs, and a series of cancers, including germ cell tumours, neuroblastomas, brain tumours (astrocytoma and medulloblastoma), acute lymphoblastic and myeloid leukaemia, soft tissue sarcoma, Wilms tumour, and Ewing sarcoma; however, these severe medical conditions have only been reported in foetuses and infants in the aforementioned studies. These associations may be due to effects on genes that play a role in the development of the axial skeleton and are also involved in the regulation of cell proliferation. Therefore, it has been hypothesized that disruptions in *Hox* gene expression can lead to disturbances of cell proliferation resulting in childhood cancers, which would explain the high association observed between the presence of cervical ribs and the occurrence of oncogenic activity in children (Anbazhagan & Raman, 1997; Galis, 1999a; Loder et al., 2007; Merks et al., 2005; Oostra et al., 2005; Quinonez & Innis, 2014; Spadliński et al., 2016). Thus, the presence of cervical ribs during early life reduces the chances of survival of individuals to develop into viable adults, and being considered, the presence of this anatomical variant is an indicator of a strongly compromised health status of the individual (Galis et al., 2006; Partiot et al., 2020).

Classification system

In 1869, Gruber (quoted in Beck, 1905) proposed a classification of cervical ribs for adults and older children, differentiating the different grades of this anatomical variant according to several stages of development. On the basis of his viewpoint we may divide the different classes as follows (Barnes, 2012; Beck, 1905; Honeij, 1920):

- *Class I (slight degree)*. The cervical rib is a small tubercle or small separate ossicle at the end of the transverse process of the cervical vertebrae.
- *Class II (more advanced)*. The cervical rib reaches beyond the transverse process, either with a free end or touching the first rib. This cervical rib is a blunt projection of bone 40–50 mm in length.
- *Class III (almost complete)*. The cervical rib extends beyond the transverse process to articulate with the first rib (by means of a distinct band or by the end of its long body) or is even attached to the sternum (by means of a fibrous band of cartilage).
- *Class IV (complete)*. It has become a true rib and possesses a true cartilage, which unites with the cartilage of the first rib. Sometimes the cervical rib imitates a first thoracic rib; however, the cervical rib appears more constricted and narrower than the first rib, and the width and length vary from 20 mm to 85 mm.

Although historically classified into these four groups, from a clinical point of view, cervical ribs can be divided into two types (Powell & Illig, 2015):

- *Type I (complete).* The cervical rib attaches to the first rib by fusion or by a true joint.
- *Type II (incomplete).* The cervical rib attaches to the first rib instead by way of a thick, tight ligament.

A problem detected during the implementation of this study was the controversy about the definition of what constitutes a cervical rib. In clinical medicine, there has been no consensus reached on what constitutes a well-developed elongated transverse process or rudimentary first rib as opposed to a cervical rib (Bokhari et al., 2012). The lack of consensus on this definition could be translated in the discrepancy data about the prevalence of true cervical ribs in the different populations reported in the literature based on radiographs (see section 'Epidemiology and clinical significance'). According to De Luca et al. (2013), it is important that the basis of palaeopathological diagnosis of skeletal remains is as close as possible to that in modern clinical medicine. Therefore, despite the few cases reported in the palaeopathological literature, some of them may have misidentified true cervical ribs using the different definitions available in the medical literature. To avoid errors in future palaeopathological studies, we recommend using the criteria for diagnosing cervical ribs widely used in medical literature suggested by Merks et al. (2005) and Brewin et al. (2009), which are not only based on radiographs, but also are easy to use on dry bone remains. These are as follows:

- The *cervical rib* must articulate with the seventh cervical vertebra and project either caudally or laterally from the spine, as opposed to the transverse process, of the first thoracic vertebra, which projects diagonally upward from the point of origin.
- For a rib to qualify as *cervical rib*, it must not articulate with the manubrium sterni, but it may be joined to the first thoracic rib. This serves to differentiate it from the *rudimentary first rib*. The *rudimentary first rib* differs from the *typical thoracic rib* in that it attaches to the second rib instead of the sternum.
- The *cervical rib* must be separated from, but articulate with, the transverse process of the seventh cervical vertebra with a well-defined joint. If the rib is fused with the vertebra, it is considered an *elongated transverse process (transverse apophysomegaly* or *transverse mega-apophysis)*. The transverse process of the seventh cervical vertebra is considered to be elongated if it projects beyond the lateral margins of the first thoracic vertebra.

Material and Methods

Archaeological context

From the pre-Roman to the medieval age, *Corfinium* constituted the main settlement of the southern zone of the *Valle Peligna,* located on the route of the consular road *Claudia Valeria* inland of the region of Abruzzo (southern Italy) (Figure 1).

-----INSERT FIGURE 1 ABOUT HERE-----

Between 1988 and 1994 and more recently between 2013 and 2018, a series of archaeological excavation campaigns were started near the Romanesque complex of the Cathedral of San Pelino aimed at reconstructing the methods of construction and characteristics and times of use of the Late Antiquity and medieval funerary area (Cavallari, 2015; Colleti et al., 1990; De Nino, 1879; Somma, 2015; Somma et al., 2018; Somma & Antonelli, 2015; Van Wonterghem, 1984). It seems clear that the most significant sepulchral nucleus was located in the space immediately behind the apse of Sant'Alessandro's oratory. In this area, approximately 92 burials and six ossuaries were identified, of which two of them (tombs T39 and T47) were made by re-using the stone coffins relating to the previous use of the funerary area in the Late Antiquity period, and were still present *in situ* during the phase of frequentation of the late medieval cemetery (Giuntella, 1989). Based on the first data that emerged from the stratigraphic analysis and from the preliminary study of the very few grave goods found in the tombs, it is hypothesized the use of this late medieval cemetery was from the XII to XIII century CE. Although the archaeological excavation is not yet complete, it can clearly be seen how the area in which the medieval cemetery was initially arranged was object to some organization in the arrangement of the burials; later, the space was intensively exploited for depositional purposes. The constant need to free up space for new burials should have also led to the periodic dismantling and removal of older tombs with the consequent construction of ossuaries, in addition to the reuse of pre-existing monumental tombs for the burial of a different number of deceased. Specifically, in two of the three stone coffins of the early Middle Ages (tombs T39 and T47), a rather complex depositional situation has been found. In both tombs, the superficial layers involved commingled skeletal remains. In the immediate underlying layers skeletal remains (partially or totally articulated) were found belonging to different individuals deposited one on top of the other. In the current state of research, preliminary anthropological analyses carried out on the skeletal remains have calculated a minimum number of nine individuals for T39 and eight individuals for T47. Among the nine individuals of tomb T39 (Figure 2), the individual I44bis was of particular interest, of which only a few skeletal remains have been preserved.

-----INSERT FIGURE 2 ABOUT HERE-----

Brief description of individual I44bis

In recent years, during the anthropological examination of the skeletal remains belonging to the individual I44 (male individual, aged 50–55 years), three vertebrae (seventh cervical and first and second thoracic vertebrae) and one rib belonging to a juvenile individual were found. These bones articulated correctly between them; therefore, they were identified as belonging to the same individual (designated I44bis). No other bone remains were preserved or could be assigned to this juvenile skeleton. Sex could not be estimated. The age of death was estimated through the morphological and dimensional features of the vertebrae (Albert & Maples, 1995; Scheuer & Black, 2004). The internal structure of the bone elements was observed through X-ray analysis.

Results and discussion

The three vertebrae showed the centres of ossification completely fused with the transverse processes. In addition, there was no evidence of the presence of the epiphyseal ring and regular undulations were present on the edges of the vertebral body of all three vertebrae. Thus, the individual was aged between 10 and 16 years according to the descriptions of Albert & Maples (1995) and Scheuer & Black (2004).

Accurate inspection of these bones showed that the first and second thoracic vertebrae were normal in morphology and size. However, the seventh cervical vertebra showed a well-defined articular facet on the transverse process on the left side (Figure 3). The rib (Figure 4) had a well-developed shaft, measuring 49 mm in length, 40 mm in chord length, and a maximum breadth of 4.5 mm. The rib presented a free rounded distal extremity with no evidence suggesting a fibrous band in connection to the first thoracic rib below. The vertebral end showed an articular facet that match with the one on the left transverse process of the seventh cervical vertebra. In other words, the rib articulated with the vertebra via a well-defined joint with the left transverse process, and it could truly be a cervical rib in origin. The right transverse process was partially broken due to postmortem damage, precluding any observation of a contralateral joint. Thus, there was no reason to presume that this was a case of a bilateral cervical rib.

-----INSERT FIGURES 3 AND 4 ABOUT HERE-----

The cervical rib did not follow the normal curve of the first thoracic rib but tended to be straight, running nearly vertically, inferiorly and anterolaterally. According to length and the absence of osseous signs of a fibrous band, the cervical rib can be classified as Class II (more advanced) according to Gruber's classification. Figure 5 shows the spatial orientation of the cervical rib in association with the three articulated vertebrae. In addition, in the left transverse process of the seventh cervical vertebra, a complete double foramen transversarium was observed, where the foramen was subdivided by a thin bony septum (Figure 3). Both foramina were of small size compared to the contralateral one. Conversely, the foramen transversarium on the right side corresponded to an incomplete double foramen transversarium. Figure 6 shows the radiographic images of the seventh cervical vertebra and its associated cervical rib, precluding any type of pathologies (e.g., antemortem fractures related to traumatic events, infections, etc.).

-----INSERT FIGURES 5 AND 6 ABOUT HERE-----

According to the anatomical model (Du Toit & De Muelenaere, 1982; Rockwood & Matsen III, 2009; Testut, 1943), the scalenus anterior muscle is inserted into the scalene tubercle on the inner border of the first thoracic rib, while the scalenus medius muscle is inserted about halfway along the arc of the first rib into the upper surface, between the tubercle and the subclavian groove (Figure 7). The subclavian artery and brachial plexus pass between the anterior and middle scalene muscles, whereas the subclavian vein passes anterior to the scalenus anterior as the muscle crosses over the first rib. Together, the subclavian vessels (vein and artery) and brachial plexus leave the neck for the arm, passing posterior to the clavicle, and then between the clavicle and the first thoracic rib, where the divisions form the lateral, posterior, and medial cords of the plexus. Laterally, the neurovascular structures lie inferior to the coracoid process and deep to the pectoralis minor muscle. In the presence of a complete cervical rib or the fibrous extension of an incomplete rib, it implants on (or close to) the scalene tubercle of the first thoracic rib. This makes the interscalene triangle even smaller, causing the above-mentioned structures to ‘hang over’ the cervical rib or its fibrous extension.

-----INSERT FIGURE 7 ABOUT HERE-----

Many clinical symptoms may be attributed to a cervical rib, whose main manifestations are located at the base of the neck, anterior chest wall, shoulder, arm, elbow, forearm, wrist, hand, or any combination of these. The characteristic symptoms are pain, atrophy, circulatory abnormalities and disturbance of sensation (Adson & Coffey, 1927; Du Toit & De Muelenaere, 1982; Gilliatt et al., 1970; Patterson, 1940; Sanders & Hammond, 2002). The pain may be associated with hyperesthesia, paraesthesia or anaesthesia, depending on the degree of involvement and varying intensity. This is usually referred to as tingling, burning and numbness along the inner side of the arm, hand and fingers. Atrophy is late to occur and is rarely complete. Circulatory symptoms are rarely severe, but they may manifest themselves in a dusky hue of the arm and hand. Other symptoms include occipital headaches; tiredness and weakness of the arm and forearm; cramps in the fingers; numbness, tingling or coldness of the hand; shrinking of some of the muscles of the hand; a lump at the base of the neck; tremor of the fingers; discoloration of the fingers; and gangrene of the distal parts (e.g., a fingertip or even a whole digit) (Adson & Coffey, 1927; Du Toit & De Muelenaere, 1982; Gilliatt et al., 1970; Patterson, 1940; Sanders & Hammond, 2002).

The size of a cervical rib is associated with the severity of medical problems (Bots et al., 2011) and, in a well-developed case such as the individual I44bis from Corfinio, with an almost complete left cervical rib 49 mm in length extending far forward in the posterior triangle of the neck, and with no evidence of a thin fibrous band connected to the first rib, this supernumerary rib can cause a bony obstruction of the subclavian artery as it curves over the thoracic outlet to reach the neck and axilla. This can happen if the left subclavian artery is elevated so that it passes over the cervical rib, which assume the anatomical relations that ordinarily belong to the first thoracic pair. This anatomical condition could be related to considerable compression of the subclavian artery, leading to diminished blood flow that could manifest clinical symptoms. The case of individual I44bis from Corfinio is in agreement with Chang et al. (2013), who highlighted that more advanced, almost complete and complete cervical ribs (Classes II, III and IV) lead to vascular compromise, including arterial compression, thrombosis, and aneurysm formation. Besides, Ross (1959) noted that complete ribs were associated with vascular complications, whereas if the cervical rib was a short pointed structure and there was a fibrous band connected to the first rib, the situation could lead to neurologic complications due to the compression of the brachial plexus. When the blood vessels are compressed, thrombosis, leading to gangrene of the fingers, may occur (Wells, 1979). Bearn et al. (1993) and Du Toit & De Muelenaere (1982) observed that complete cervical ribs were more commonly associated with arterial insufficiency than with neurological or venous symptoms. Nevertheless, only 5–10% of people with cervical ribs experience symptoms (Gülekon et al., 1999), which are manifested in older children and adults, whereas cervical ribs are rarely symptomatic in early childhood (Glass et al., 2002; Todd, 1911). The individual I44bis described from the late medieval cemetery of Corfinio had an estimated age of 10–16 years and could have manifested certain symptoms, but in a very mild degree. However, evidence of symptoms is difficult, if not impossible, to diagnose and interpret on skeletal human remains, particularly in this reported case. According to Ortner (2003), evidence of disease on human skeletal remains in past populations is a strong challenge in palaeopathology, and the inference of signs and symptoms can be subjective and difficult to associate with specific pathological conditions in certain contexts. Due to the absence of other skeletal elements belonging to this same individual, it was not possible to conduct a more exhaustive study; therefore, interpretations about the possible symptoms that it could have suffered were avoided.

In summary, the analysis of human skeletal remains not only allows the study of the history and evolution of the disease but also the reconstruction of health conditions in past populations (D'Anastasio et al., 2013; De Luca et al., 2013; Viciano et al., 2017a, 2015a, 2015b, 2012; Viciano & D'Anastasio, 2018). The examination of human skeletal remains has demonstrated that anatomical variants may be potentially associated with pathological conditions (Barnes, 2012; Nikolova et al., 2014; Ortner, 2003). Thus, the palaeopathological study of ancient human remains allows to highlight if anatomical variants could be related to life and health conditions in past populations. However, although specific case studies do not contribute by themselves to the advocated 'health status population approach', these case studies provide an important source of data for particular palaeopathological conditions; without them, there would be less data available for future studies, and therefore, we would have a less comprehensive picture of the impact that these pathological conditions could have had in past individuals/populations.

Conclusion

The palaeopathological cases of cervical ribs are difficult to diagnose on ancient human remains. Curiously, this anatomical variant is also silent in modern populations, as many cases are asymptomatic. However, the discovery of cervical ribs on ancient human remains can be interesting, as it could be related to complex symptomatology found in some clinical cases through X-ray images.

The skeletal remains of individual I44bis from the late medieval cemetery of Corfinio have all the anatomical features necessary to classify it as a true cervical rib (Class II, more advanced) according to the diagnostic criteria suggested in the scientific literature. This supernumerary rib could be related to considerable compression of the subclavian artery, leading to diminished blood flow of the subclavian artery, which could manifest clinical symptoms. Moreover, this study has given us the opportunity to review the palaeopathological and clinical cases in order to highlight the difficulties of classifying a true cervical rib and suggest diagnostic criteria for its diagnosis.

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Fig. 1. Geographic location of late medieval cemetery of Corfinio (XII–XIII centuries CE) in the region of Abruzzo (Italy).

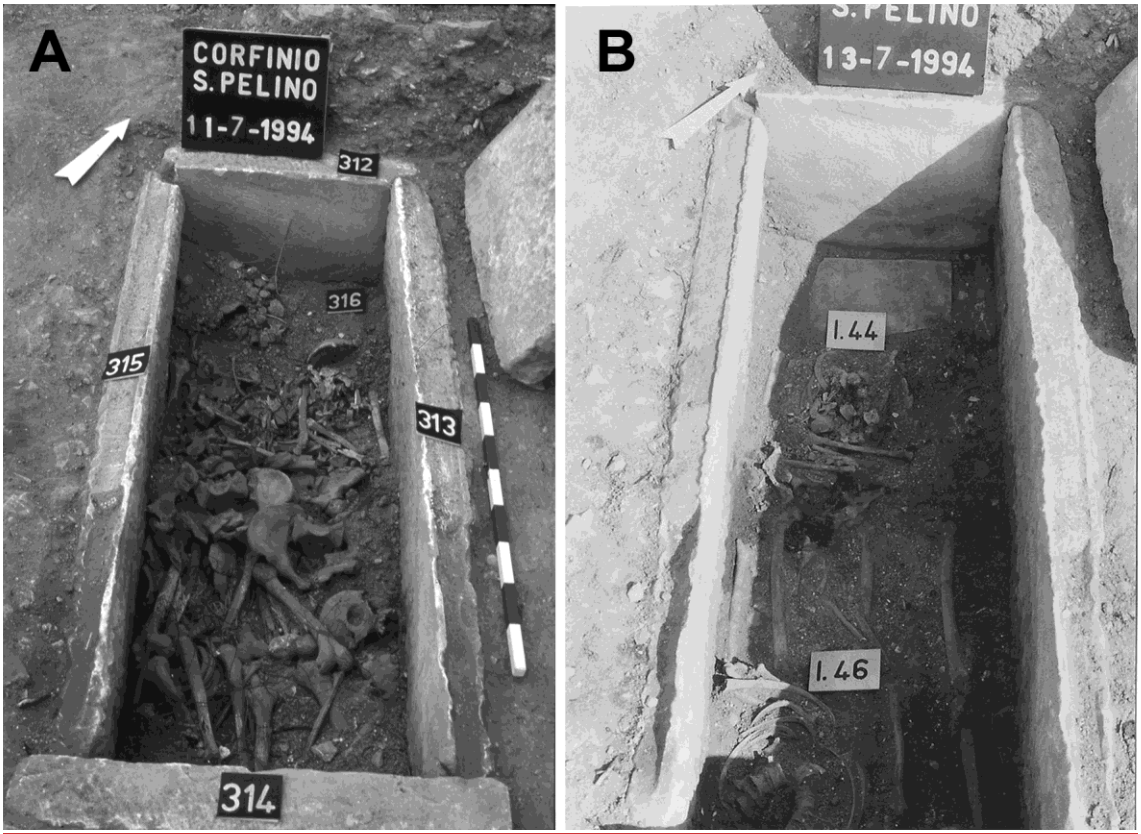

Figure 2. A. Excavation context in the stone coffin T39 with commingled human remains. **B.** Inside of the stone coffin after the excavation was almost complete, with the presence of two completely articulated skeletons (I44 and I46) and a partially spine of one individual not identified in the image. The skeletal remains of I44bis are not visible in this image (images courtesy of M.C. Somma).

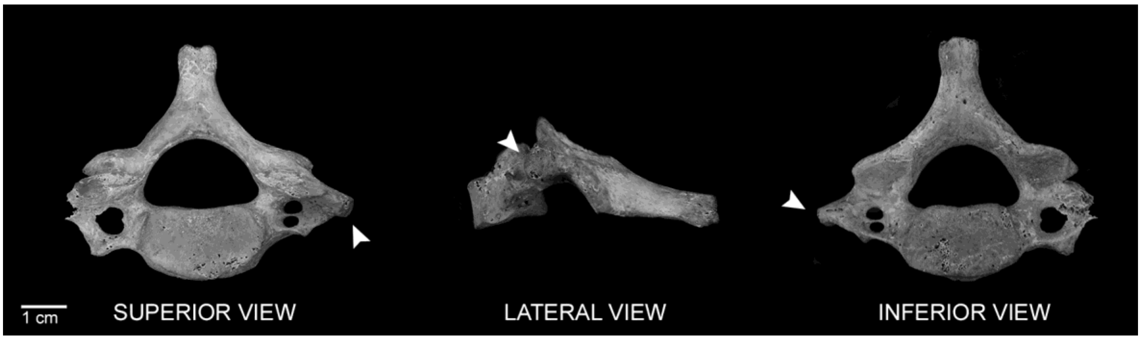

Figure 3. Different views of the seventh cervical vertebra, showing a well-defined articular facet (arrowhead) and incomplete (right) and complete (left) double foramen transversarium on the transverse process.

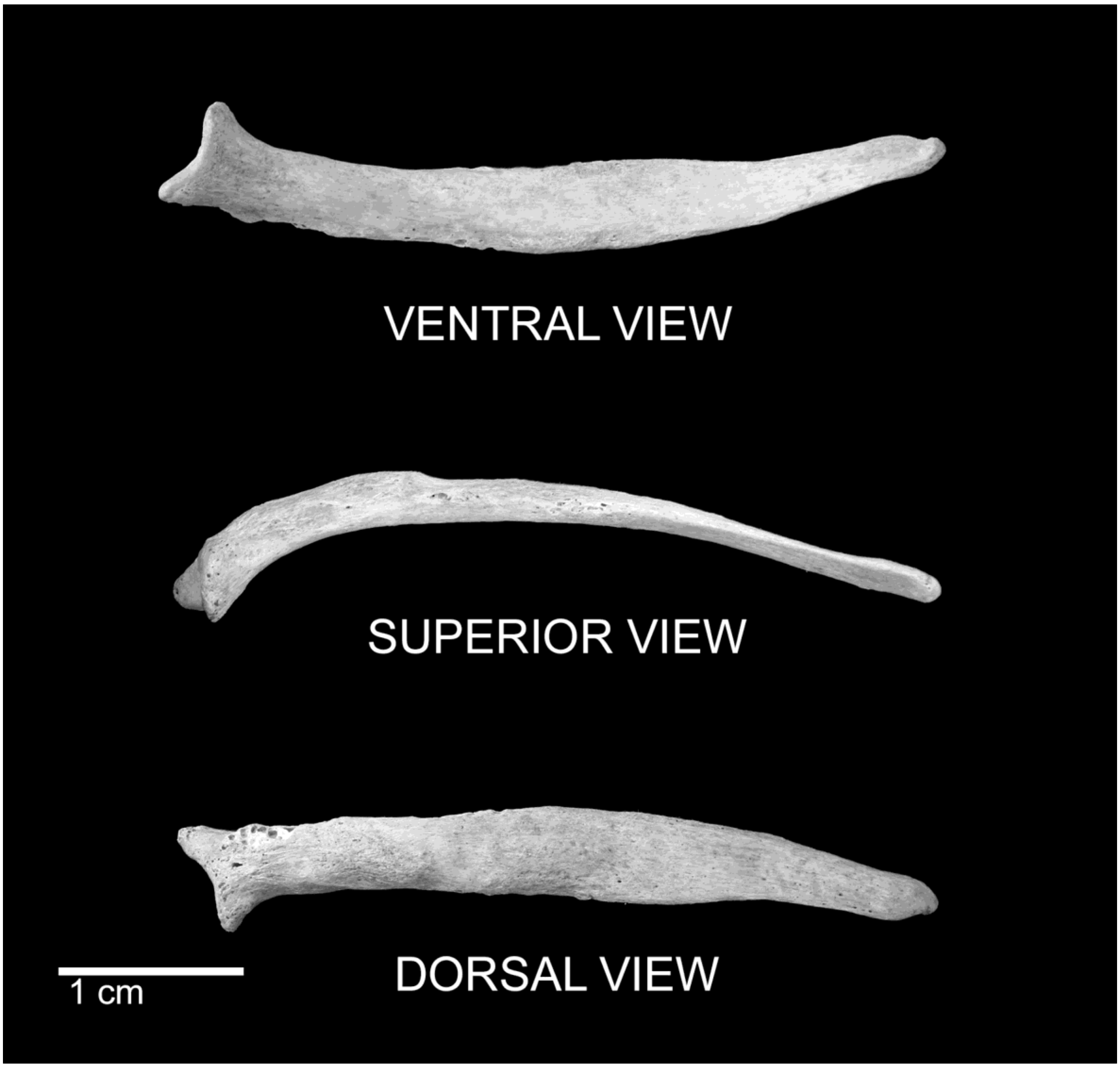

Figure 4. Different views of the cervical rib.

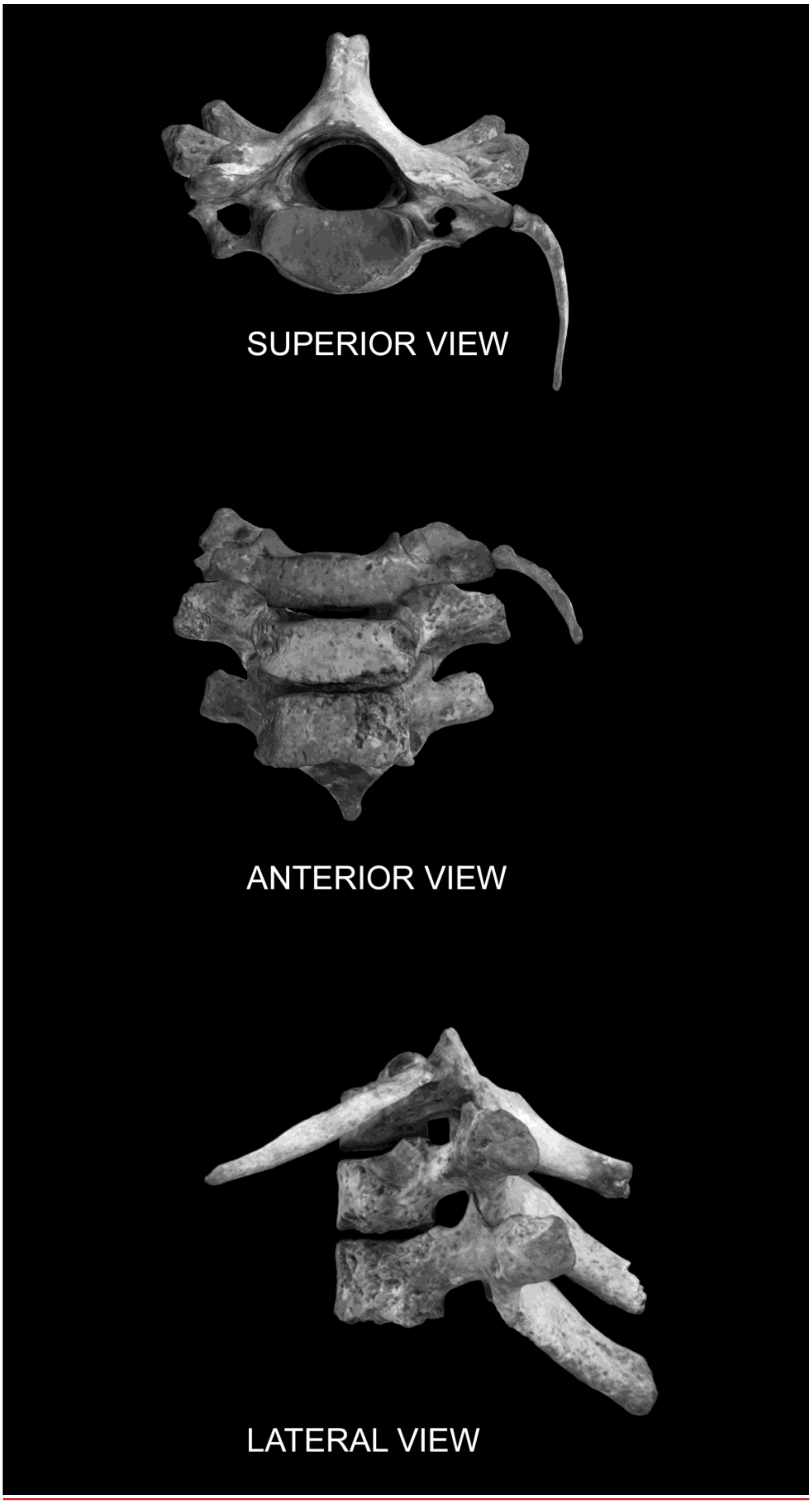

Figure 5. Different views of the spatial orientation of the cervical rib in association with the three articulated vertebrae (seventh cervical and first and second thoracic vertebrae).

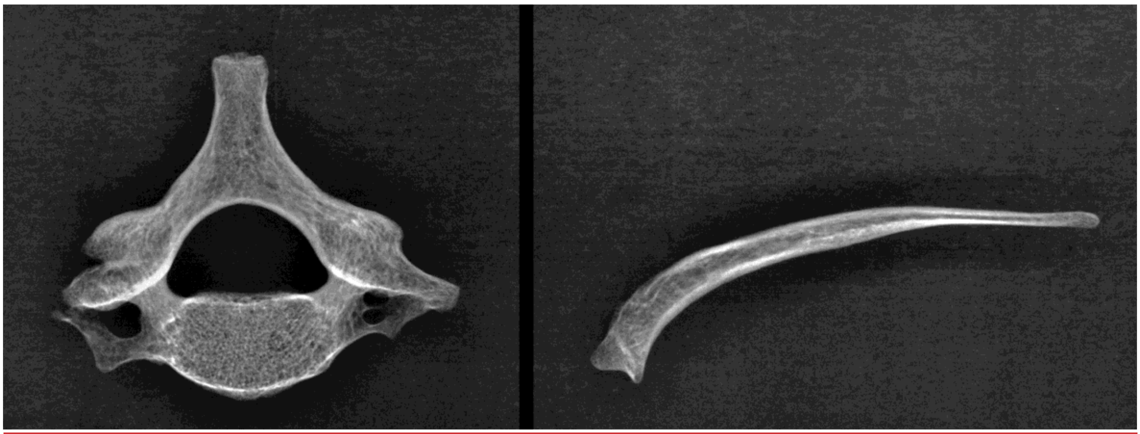

Figure 6. Radiographic images of the seventh cervical vertebra and its associated cervical rib.

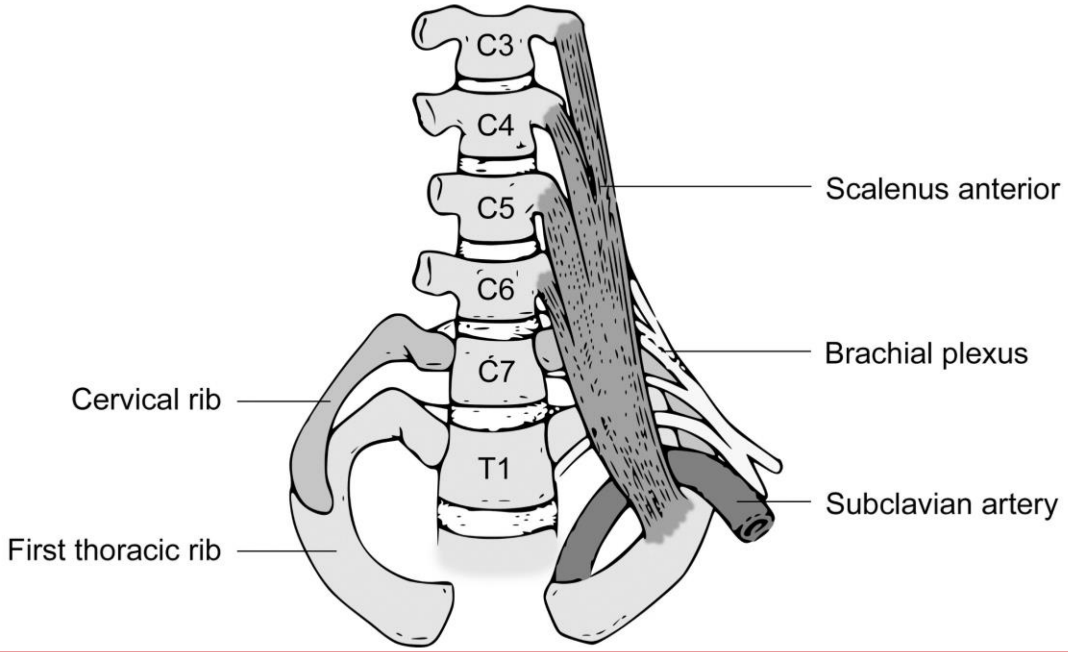

Figure 7. Relevant anatomy of the neck. In the presence of a complete cervical rib the interscalene triangle is smaller, causing compression of the neurovascular structures (adapted from Ellis & Mahadevan, 2019).

Table 1. Historical background with the most important milestones regarding the studies on cervical ribs.

Year (CE)	Event	Source
130–201	The first description of a cervical rib was apparently made by Galen during dissections of human cadavers.	Roos et al. (1999)
1514–1564	Andreas Vesalius again described the cervical rib in human cadavers.	Roos et al. (1999)
1742	François-Joseph Hunauld described and categorized supernumerary ribs in humans, including the cervical rib.	Clare, Schilp, & Starr (1956); Wei Lum & Fresichlag (2014)
1818	Sir Astley Cooper first described arterial thoracic outlet syndrome in a young woman with an absent radial pulse and reduced sensation and pain in her hand and demonstrated the relationship of neurological and vascular symptoms in the upper limb with a cervical rib. His observation led gradually to the recognition of a 'cervical rib syndrome'.	Brewin et al. (2009); Cooper (1818)
1860	W.H. Willshire noted the relationship between a cervical rib and paraesthesia of the upper extremity.	Wei Lum & Fresichlag (2014)
1861	Richard Holmes Coote, at St. Bartholomew's Hospital in London, performed the first surgical operation to treat thoracic outlet pathology in a woman diagnosed as having an 'exostosis' of the seventh cervical vertebra with predominantly vascular symptoms and an absent radial pulse. Coote resected a portion of the transverse process, and the patient's symptoms improved.	Coote (1861)
1869	W. Gruber defined four types of cervical rib ranging from rudimentary ribs barely longer than the transverse process of the seventh vertebra to complete, fully articulated ribs.	Beck (1905); Honeij (1920)
1905	John Benjamin Murphy was the first surgeon to resect a cervical rib that was associated with a subclavian artery aneurysm.	Murphy (1905)
1907	William W. Keen, at Thomas Jefferson University in Philadelphia, published a review of 42 cases of resected cervical ribs and described a clinical definition and surgical treatment for this disorder.	Keen (1907)
1908	J.B. Roberts described numerous anatomic forms of the cervical rib and surgical importance of each form.	Roberts (1908)
1911–1913	Between the years 1911 and 1913, in a series of scientific papers outlining the anatomy and anatomical variability of the thoracic outlet, Thomas Wingate Todd described the potential causes of neurovascular compression in the thoracic outlet. Todd's anatomic dissections included descriptions of cervical ribs, anatomic variants of the first rib and scalene muscles, and the position of the clavicle with shoulder movements. Todd also postulated that the gradual descent of the shoulder girdle with aging causes narrowing of the space between the clavicle and rib, contributing to neurovascular compression.	Todd (1911, 1912, 1913)
1916	W.S. Halsted evaluated 716 case reports of cervical ribs, in which 125 had vascular symptoms and 27 had aneurysms distal to the point of compression by the cervical rib and its attachments.	Halsted (1916)

1927	Alfred W. Adson and Jay R. Coffey suggested the mechanism of the anterior scalene muscle causing upper extremity neurovascular compression in patients with cervical ribs and for the first time demonstrated that scalenotomy could relieve symptoms of neurovascular compression without resection of the cervical rib.	Adson & Coffey (1927)
1934	T. Lewis and G.W. Pickering reported that subclavian artery thrombosis and embolization could occur due to trauma caused by a cervical rib.	Lewis & Pickering (1934)
1956	R.M. Peet and colleagues first use the term ‘thoracic outlet syndrome’ to unify all the upper extremity neurovascular compression syndromes.	Peet et al. (1956)