

Anatomical variants**DOUBLE TRANSVERSE FORAMEN IN INFANTILE CERVICAL VERTEBRAE OF THE LATE 17TH AND EARLY 18TH CENTURIES****Laura Quiles-Guiñau, Marcos Miquel-Feucht***Department of Human Anatomy and Embryology, School of Medicine. University of Valencia. Spain***RESUMEN**

La presencia de doble foramen transverso es una variante anatómica que se observa con frecuencia en el raquis cervical inferior y que presenta implicaciones clínicas en relación con la arteria vertebral. Aunque en la actualidad existe cierta controversia acerca de si el origen de esta variante cervical es congénito o adquirido, y a pesar de que la presencia de doble foramen transverso en población infantil descartaría un origen degenerativo-artroscico, se encuentra una falta de información acerca de su presencia en vértebras cervicales infantiles. En este sentido, nuestro objetivo ha sido analizar la presencia de doble foramen transverso en vértebras cervicales infantiles procedentes de restos óseos, datados entre finales del siglo XVII y principios del XVIII, y exhumados de una fosa común en la Iglesia de Nuestra Señora de los Ángeles en Castielfabib (Ademuz, España). Se encontraron 10 vértebras infantiles con doble foramen transverso, 4 de ellas pertenecientes a niños de menos de 4 años y 6 a niños de más de 4 años. Este hallazgo apoya un origen congénito de esta variante anatómica, frente a la hipótesis degenerativo-artroscica, y apunta a la necesidad de un estudio en profundidad acerca de la prevalencia actual de esta variante anatómica en población pediátrica, dadas sus posibles consecuencias clínicas.

Palabras clave: niño, raquis, anomalías congénitas, España.

ABSTRACT

Double transverse foramen is an anatomical variant often observed in the inferior cervical spine and which may present clinical considerations with regard to the vertebral artery. There is some current controversy as to whether the origin of this cervical variant is congenital or acquired. Despite the fact that its presence in children would discard a degenerative osteo-arthritis origin, there is a lack of information on

the origin and physiological meaning of this anatomical feature. In this respect, our objective was to analyse the presence of double transverse foramen in infantile cervical vertebrae in skeletal samples dating from the end of the 17th and early 18th centuries and exhumed from a common grave in the "Nuestra Señora de los Ángeles" church, in Castielfabib (Ademuz, Valencia, Spain). Ten infantile vertebrae with double transverse foramen were discovered: 4 from children under 4 years old and 6 from children over 4 years old. This finding supports more a congenital origin than a degenerative osteoarthritis process. Therefore, it is fundamental to further study its prevalence in a current paediatric population and its clinical significance.

Keywords: child, spine, congenital abnormalities, Spain.

INTRODUCTION

Double transverse foramen (DTF) denotes the presence of an additional transverse orifice in the cervical vertebrae, which is usually smaller and posterior to the principal one (Kaya et al, 2011). This anatomical variant may be found in the inferior cervical spine either unilaterally or bilaterally, and located more often in vertebra C6 (Murlimaju et al, 2011; Chandravadia et al, 2013). The high prevalence of DTF at the cervical spine (up to 50%) makes it a common anatomical variant and, therefore, should be evaluated in clinical practice (Rios et al, 2014).

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Transverse foramen (TF) nosological status is determined in relation to the vertebral artery, depending on whether or not it affects normal blood flow (Taitz et al, 1978; Yilmazlar et al, 2003; Nayak, 2008). Variations in TF number and size may cause headaches, blackouts due to low blood pressure on the vertebral artery (Nayak, 2008) and vertebrobasilar insufficiency as a response to certain neck movements (Bulsara et al, 2006). In children, DTF presence may be associated with a higher risk of vertebral artery sectioning and cerebral infarction (Sedney and Rosen, 2011). Furthermore, detailed anatomical knowledge on FT would be useful in X-ray analysis and cervical surgery involving pedicles and transverse processes.

There is some controversy on DTF origin, from hereditary congenital (Aydinoğlu et al, 2001; Kaya et al, 2011), or congenital associated to differential development of the vertebral artery (Das et al, 2005), to osteoporotic degenerative process involving formation of osteophytes (Sanchis-Gimeno et al, 2005; Katzenberg and Saunders, 2008). And the fact that most studies focus on adult cervical vertebrae (Taitz et al, 1978; Nagar et al, 1999; Aydinoğlu et al, 2001; Das et al, 2005; Sanchis-Gimeno et al, 2005; Sharma et al, 2010; Kaya et al, 2011; Murlimanju et al, 2011; Agrawal et al, 2012; Chandravadiya et al, 2013; Rathnakar et al, 2013; Katikireddi and Setty, 2014) does not help to clarify DTF origin.



Figure 1. Infantile cervical vertebrae. (A) Perinatal. (B) Child aged 2 to 4 years old, without TF anterior closure. (C) Child aged 2 to 4 years old, with TF anterior closure. (D) Child aged over 4 years old.

MATERIAL AND METHODS

Infantile vertebrae samples used in this study were discovered between 1999 and 2005, during restoration works at the fortress-church "Nuestra Señora de los Ángeles", in Castielfabib (Rincón

de Ademuz, Valencia, Spain). This fortress-church was initially built as a castle in the 12th century over ruins of first a Roman and then Arab fortress. During the 13th and 14th centuries, one of the defensive towers was enlarged and converted into a palace; later, its upper level

became the current church (López-González and García-Valldecabres, 2012). The buried osseous remains discovered under its floors and within its walls date from late 17th to the early 18th centuries, as has been revealed by the layette items found in the tombs as well as by the historical context.

Skeletal samples were labelled according to their stratigraphic unity status, and analyzed at the Anthropometry and Paleopathology Laboratory, Department of Anatomy and Human Embryology, University of Valencia, as recommended by the Spanish forensic anthropology and dentistry association known as "Asociación Española de Antropología y Odontología Forense" (Serrulla, 2013).

Bone samples were initially cleaned with a soft-bristle brush under a constant low-pressure water jet and then dried under mechanical ventilation at room temperature (Serrulla, 2013). After drying no consolidation procedures were necessary.

Bone samples came from common graves, which presented a mix of bones. Therefore, it was necessary to calculate the minimum number of individuals. The species and anthropological identification was based on a morphological and morphometric analysis (Reverte-Coma, 1991). Then, bones samples were separated as adult or child bones. Since children skeletons were partially incomplete, their gender could not be determined.

Children skeletons were separated in 5 age ranges: foetal, neonate, child 1 (0 to 7 years old), child 2 (8 to 14 years old) and adolescent (15 to 18 years old). In categories child 1, child 2 and adolescent, age range was determined by analyzing synostosis, epiphysis growth (Reverte-Coma, 1991) and degree of dental eruption (Demirjian and Goldstein, 1976). For perinatal and neonatal categories, age was determined by diaphysis lengths of humerus, radius, femur and tibia (Hoffmann, 1979).



Figure 2. Cervical vertebrae of children aged 1 to 4 years old with DTF. (A) Bilateral DTF. (B) Bilateral DTF. (C) Bilateral DTF. (D) Unilateral DTF (right).

Classification of infantile cervical vertebrae and their age range were established by analyzing vertebral anatomical changes involved in bone growth; this does not solely depend on vertebral size (Fig. 1), but also on the presence of striations on vertebral bodies joint surfaces (Reverte-Coma, 1991; Baker et al, 2010), anterior closure of TF (starting at age of 2 years and finishing around age of 3 to 4 years) and fusion of vertebral arch with vertebral body (occurring after closure of TF) (Baker et al, 2010).

Given that the complete cervical spine of the exhumed children were not available, the analysis was limited to the set of the observed cases of infantile vertebrae with DTF. It was not possible to determine the exact level of inferior cervical spine in each vertebra.

The study was approved by the ethics committee for research on humans from the University of Valencia; furthermore, the corresponding authorization was obtained from the local authorities at "Conselleria de Cultura de la Comunidad Valenciana".



Figure 3. Cervical vertebrae of children over 4 years old with DTF. (A) Right-side unilateral DTF. (B) Right-side unilateral DTF. (C) Right-side unilateral DTF. (D) Left-side DTF. (E) Left-side DTF. (F) Possible left-side DTF.

RESULTS

Exhumed bone samples from the "Nuestra Señora de los Ángeles" church-fortress were all of human origin, from 154 children and 177 adults (46.52% and 53.47% of total individuals, respectively). Age distribution of the 154 children was as follows: 4 (2.59%) fetuses, 40 (25.97%) newborns, 83 (53.89%) child 1, 14 (9.09%) child 2 and 5 (3.24%) adolescents. In 8 cases (5.19%) age range could not be established due to the poor state of the sample.

The exhumed infantile bones included 96 typical cervical vertebrae (from C3 to C7), from which 10 (10.41%) presented DTF: 4/10 corresponded to

children under 4 years old (Fig. 2), while the other 6/10 were from children older than 4 years old (Fig. 3). In 3/10 vertebrae, DTF was bilateral, whereas in 4/10 they were unilateral. It was not possible to determine if DTF was unilateral or bilateral in 3 vertebrae, because the right transverse process was missing.

DISCUSSION

Presence of DTF in the inferior cervical spine has been linked to ossification of structures close to TF (Katzenberg and Saunders, 2008) and

formation of osteophytes (Sanchis-Gimeno et al, 2005). One of the studies focused in an osteoporotic-degenerative origin of DTF (Sanchis-Gimeno et al, 2005) observed osteophytes in TF of C3-C7 in numerous vertebrae from post-mortem donors. In that study, degenerative changes were most frequently found in C5 and C6, the vertebrae with the highest prevalence of DTF.

Association between DTF and fenestration or duplication of vertebral arteries has been described previously (Sim et al, 2001). Since embryonic vertebrae develop together with vertebral arteries, alterations in vertebral artery development may affect TF morphology and vice-versa (Taitz et al, 1978), which would point to a congenital origin of DTF variant (Sim et al, 2001). Furthermore, the fact that cervical DTF is by far more common in humans than in primates, may point to an evolutionary consequence of the upright posture and biped walking (Rios et al, 2014). In embryological terms, TF is a result of fusion of the double costal vestige with the vertebral body and the transverse process (retaining within them vertebral vessels and nervous plexus). Therefore, DTF could be a consequence of double costal vestige fusion, resulting in an unusual number of TF (Taitz et al, 1978).

The presence of DTF affecting C3 and C6 vertebrae has been described in foetal skeletons (Rios et al, 2014), supporting the hypothesis of a congenital origin. There is nonetheless a lack of studies focusing on child population, perhaps partly due to the fact that these kinds of osseous pieces are usually not available and they tend to be extremely fragile, mainly if they come from ancient burial sites.

Our findings on various infantile vertebrae with DTF offer further information on DTF presence in a paediatric population, in agreement with fetal DTF reported by Rios et al (2014). Even though these bone samples are indicative of a congenital origin, additional degenerative arthritis origin could develop after childhood (Sanchis-Gimeno et al, 2005). To clarify this issue, it could be useful to compare DTF prevalence between a healthy adult population and an adult population with cervical arthritis.

In any case, our finding of DTF in infantile cervical vertebrae contributes to a greater clinical meaning of this anatomical variant in today's paediatric population, especially because DTF might be associated with a greater risk of vertebral artery sectioning and cerebral infarction due to osseous cervical anomalies (Sedney and Rosen, 2011). Clinical importance of DTF contrasts with a lack of data on DTF prevalence in today's paediatric population, since research

studies has exclusively focused on both present adult populations (Aydinoğlu et al, 2001; Das et al, 2005; Sanchis-Gimeno et al, 2005; Sharma et al, 2010; Murlimanju et al, 2011; Agrawal et al, 2012; Chandravadiya et al, 2013; Rathnakar et al, 2013; Katikireddi and Setty, 2014) and ancient adult populations from archaeological samples (Taitz et al, 1978; Nagar et al, 1999; Kaya et al, 2011).

Since vertebrae with DTF analyzed in this study were ancient, fragile and incompletes, it was not possible to study DTF prevalence in our subject population. Nevertheless, our findings on DTF in infantile cervical vertebrae supports a congenital origin of this anatomical variant and highlights the need for further research on today's DTF prevalence in paediatric population.

Conflict of Interests

The authors declare they have no conflict of interest.

Funding

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Ethical Approval

The study was approved by the Research Ethics Committee at the University of Valencia.

Informed consent

Not necessary.

Contribution

This study was designed by LQ; MM collected the data which was analysed by LQ and MM; and the manuscript was written by LQ and MM.

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