# THE INCREASED FEMORAL NECK ANTEVERSION IN MEDIEVAL CEMETERY OF PECENJEVCE. AETIOLOGY AND DIFFERENTIAL DIAGNOSIS IN ARCHAEOLOGICAL CONTEXT

## ANTEVERSIÓN FEMORAL AUMENTADA EN EL CEMENTERIO MEDIEVAL DE PECENJEVCE. ETIOLOGÍA Y DIAGNÓSTICO DIFERENCIAL EN EL CONTEXTO ARQUEOLÓGICO

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The femoral neck anteversion (FNA) is defined as the angle between the longitudinal axis of the neck of a femur and the axis passing horizontally through femoral condyles. However, there is no data regarding this feature in archaeological populations. Therefore, the aim of this study was to investigate FNA in a medieval skeletal population from Serbia. According to the results the analysed angle ranged from 11 to 24 degrees in adults, apart from only one individual with significantly increased femoral neck anteversion of nearly 60 degrees. The discussion of the present paper is focused on the differential diagnosis of this condition and its aetiology, especially outlining diagnostic limitations when dealing with dry bones. Finally, the most probable aetiology of increased FNA in our case is the asymmetric form of cerebral palsy. Overall, the traces of various orthopaedic and neuromuscular disorders in past human populations could be revealed by systematic recording of the femoral neck anteversion during anthropological analyses.

**Key words:** femoral neck anteversion; medieval skeletal population; cerebral palsy; orthopaedic disorders; neuromuscular disorders.

*Anteversión femoral se define como el ángulo entre el eje cérvico-cefálico y la tangente posterior bicondílea que pasa por el plano horizontal. Dado que no hay datos sobre el diagnóstico diferencial de esta característica en las poblaciones arqueológicas, la intención de este estudio fue investigar anteversión femoral (FNA) en los esqueletos de una población medieval de Serbia. Los resultados demostraron valores de este ángulo de 11 a 24 grados en adultos, pero una persona tenía el ángulo de anteversión femoral significativamente aumentada (casi 60 grados). En la discusión hemos enfocado en el diagnóstico diferencial de alta FNA causada por diferente etiología, así como sobre las limitaciones diagnósticas en los huesos secos. La más probable etiología de aumento de FNA en nuestro caso es la forma asimétrica de parálisis cerebral. Revisión sistemática de la anteversión femoral puede demostrar las huellas de diversos trastornos ortopédicos y neuromusculares en las poblaciones humanas del pasado.*

***Palabras claves:*** *anteversión femoral, población medieval, parálisis cerebral, enfermedades ortopédicas, enfermedades neuromusculares.*

Schoenecker and Rich (2006) defined the concept of *version* as a tilt or inclination within a bone. Thus, *femoral neck anteversion (FNA)* is represented by the angle between the longitudinal axis of the neck of a femur and the axis passing horizontally through femoral condyles (Crane 1959; Fabry et al. 1973). It depicts the degree of rotation of the femoral neck in reference to the coronal plane and influences the ability of internal thigh rotation (Schoenecker and Rich 2006).

The femoral anteversion is a result of fetal development, heredity, mechanical forces, and intrauterine position (Guidera et al. 1994). The value of FNA angle at birth is commonly about 40 degrees and decreases gradually to approximately 20 degrees by the age of 10, to finally achieve value around 8 to 15 degrees in adulthood (Fabeck et al. 2002; Fabry et al. 1973; Harkess 2003; Hefti et al. 2007; Schoenecker and Rich 2006). It is believed that physiological decline in torsion of the femoral neck in children is influenced by dynamic forces produced during upright walking. Therefore, the magnitude of anteversion angle is attributed to appropriate motor control, muscle balance, ligamentous integrity,

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normal ambulation and even unconscious necessity to position the feet on the ground in parallel (Hefti et al. 2007; Renshaw and Deluca 2006).

In newborns, Hefti et al. (2007) considered the value of the FNA above 50 degrees as pathological. The deformity present in childhood, if left uncorrected, leads to adaptive changes in muscles, tendons, ligaments, joint capsules and finally to bone deformities in growing children (William and Warner 2003a). Higher values of FNA angle can be caused by different reasons, from fairly benign and temporary to more severe with long-term consequences (Fabry et al. 1973; Ruby et al. 1979). Thus, following literature the unilateral presentation of this condition could be observed in cases of asymmetric form of developmental dysplasia of the hip – DDH (Fabry et al. 1973; Fitzgerald et al. 2002; Subasiet al. 2008; Sugano, Noble, Kamaric et al. 1998), unilateral type of Legg-Calve-Perthes disease also known as coxa plana (Aufderheide et al. 1998; Thompson and Salter 1987) and various neuromuscular disorders. According to the publications, differential diagnosis of neuromuscular disorders should involve: hemiplegic cerebral palsy, myelomeningocele, chronic stage of poliomyelitis, and central motoneuron lesions other than cerebral palsy such as brain or spinal cord tumors or infections (Beals 1969; Bobroff et al. 1999; Cibulka 2004; Fabry et al. 1973; Glyn et al. 1966; Heftiet al. 2007; Lau et al. 1986; Lewis et al. 1964; Morrissy and Weinstein 2006; Murray and Robb 2006; Noonan 2006; Renshaw and Deluca 2006; Rogers 1934; Parsons and Seddon 1968; Samilsonet al. 1972; Sauser et al. 1986; Shefelbine and Carter 2004; Skinner 2003; Staheli 1977; Tachdjian and Minear 1956; William and Warner 2003a; William and Warner 2003b).

There are a number of clinical studies about femoral anteversion focused either on technical complexity of measuring the angle in patients (Magilligan 1956; Miller et al. 1997; Murphy et al. 1987; Ruby et al. 1979; Sugano, Noble and Kamaric 1998) or on therapeutic issues (Baker et al.1962; Beals 1969; Cobeljic et al. 2005). Anthropological and archaeological studies also mostly focused on the technical aspects of measuring it in dry bones. Stirland (1994) noted that very large variations in measurements have been probably due to two reasons: difficulties in measuring and inconsistency in the methodology. The study by Shrikant and Arati (2009) gives comparative data on femoral anteversion

obtained by centre head neck line to retrocondylar line method toward anterior head trochanter line to retrocondylar line method. The authors reported significantly different values between data acquired by tested two methods.

Heretofore, there are no guidelines for differential diagnosis of this condition in archaeological context. Therefore, in this paper we investigated FNA in the medieval skeletal population and presented the case with increased femoral anteversion, focusing on the differential diagnosis and diagnostic limitations in dry bones.

## Material and Methods

The skeletal material investigated in this study derived from the late medieval cemetery (XII - XIII Century AD) of Pecenjevce – Tapan near the city of Leskovac, located in the Southern Serbia (Figure 1).

According to the preliminary archaeological report the cemetery was comprised of 29 graves. The estimated minimum number of individuals



Figure 1. Location of the archaeological site.
*Localización del sitio arqueológico.*

(MNI) was 35. According to the conducted standard anthropological analysis the demographic distribution of skeletons involved 17 non-adults (48.57%), and 18 individuals that comprised category of adulthood whereof 2 were males (5.71%), 8 females (22.86%), and 8 individuals of undetermined (22.86%). Age categories were estimated according to the recommendation of Roksandic and Amstrong (2011) while the sex of the individuals was assessed following standard criteria of dimorphic features of the os coxae (Buikstra and Ubelaker 1994). Angle measurement of the femoral anteversion was done electronically analysing digital photography by KVI-POPOVAC Pro software (Version 2.2, Copyright Leica Imaging Systems). Palaeopathological analysis included macroscopic observation and radiographic analysis.

## Results

Analysis of the skeletal material revealed that only five adult individuals had completely preserved femora. The angle of the femoral neck anteversion in all cases ranged from approximately 11 to 24 degrees, except the skeleton of an adult woman (Nº 22) with remarkably increased femoral neck anteversion on the left side. According to the conducted analyses the age category of individua Nº 22 was estimated at full adulthood.

The skeleton Nº 22 was relatively well preserved. The skull, both clavicles and humeri were presented and in well condition. Among the vertebrae, only the first, second, fourth, fifth and seventh cervical, as well as the second and third thoracic vertebrae were preserved. Both femora were of physiological morphology, without any traces of trauma and degenerative disease. Pelvic bones were fragmented with preserved acetabular surfaces. The right acetabulum was morphologically normal. On the left acetabulum, the anterior half of the lunate surface was missing, while the rest appeared normal except the margin of the acetabulum which was taphonomically damaged. The analyses revealed relatively well preserved tibiae with fragmented left tibial plateau. The tarsal bones were presented on both sides with some post-mortem defects. In addition, the acquired measurements of the preserved long bones are given in Table 1.

The left femoral head and neck of the skeleton Nº 22 were rotated anteriorly with respect to the plane of the femoral condyles, which caused internal rotation in the hip joint. The observed femoral neck rotation resulted in unusual position of the left femur in the grave (Figure 2). FNA angle on the left femur was very high – 60 degrees (Figure 3), while recorded value of the contralateral femur was relatively low (17 degrees). In addition, the fovea of

Table 1. Measurements of the left and right long bones.
*Dimensiones de los huesos largos izquierdos y derechos.*

| Bone | Measurement | R | L |
| --- | --- | --- | --- |
| Femur | Maximum Length | 39.6 | 39.7 |
| | Maximum Head Diameter | 4.0 | 3.9 |
| | Sagittal Subtrochanteric Diameter | 2.9 | 2.8 |
| | Transverse Subtrochanteric Diameter | 2.9 | 3.2 |
| | Sagittal Midshaft Diameter | 2.9 | 2.6 |
| | Transverse Midshaft Diameter | 2.4 | 2.1 |
| | Neck-shaft Angle | 141 | 132 |
| | **FNA Angle** | **17** | **59.88** |
| | Epicondylar Breadth | 6.9 | 6.1 |
| Tibia | Maximum Length | 34.2 | 34.4 |
| | Transverse Diameter | 2.3 | 3.0 |
| | Sagittal Diameter | 2.9 | 2.6 |
| | Minimum circumference of Shaft | 6.8 | 6.7 |
| | Circumference at the Nutrient Foramen | 8.6 | 8.8 |
| Humerus | Maximum Length | / | 28.8 |
| | Minimum Circumference of Shaft | 6.1 | 5.5 |
| | Minimum Diameter | 1.9 | 1.6 |
| | Maximum Diameter | 2.2 | 1.8 |

Figure 2. Skeleton in situ: medial rotation of the left femur in the hip joint with possible equinovarus position of the left foot.
*Esqueleto* in situ*: la rotación medial del fémur izquierdo en la articulación coxofemoral con posible equinovaro del pie izquierdo.*
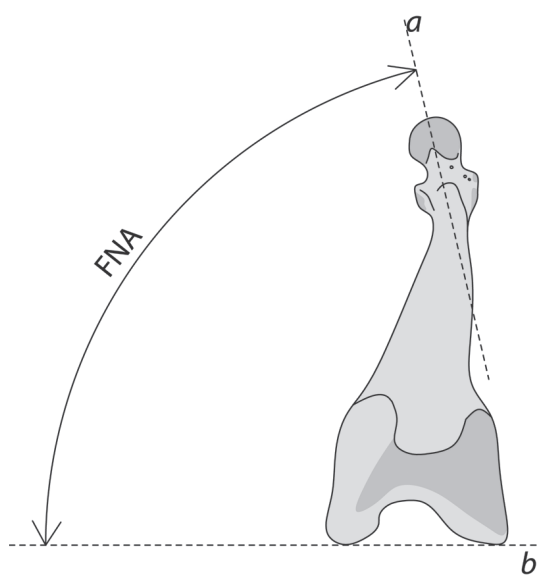


Figure 3. FNA angle measured on the left femur – 59.88 degrees.
*Anteversión femoral del fémur izquierdo – 59,88 grados.*

the left femoral head was positioned eccentrically, suggesting subluxation in this hip joint. Furthermore, no signs of tibial torsion were recorded on the left body side, and the present tarsal bones were without clear signs of pathological changes. In addition, the right clavicle displayed signs of a healed fracture. Overall, apart from the changes of the proximal femur and the right clavicle no further evidence of pathological changes were found.

### Discussion

Abnormal femoral anteversionis associated with clinical manifestations ranging from benign intoeing gait to severe orthopaedic problems. Table 2 summarizes the morphological characteristics of various conditions with possible expressed increased FNA angle. However, anteversion can also be high on bones unaffected by any other pathological conditions.

One of the main forms of an increased femoral anteversion in childhood is physiological

Table 2. Guidelines for differential diagnosis of increased femoral anteversion in dry bones.
*Directrices para el diagnóstico diferencial de anteversión femoral aumentada en huesos secos.*

| | Physiological | | Developmental dysplasia of the hip | | Legg-Calve-Perthes disease | | Cerebral palsy | | Chronic phase of poliomyelitis | | Myelomeningoceles | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Finding | Level | Finding | Level | Finding | Level | Finding | Level | Finding | Level | Finding | Level |
| Symmetry of the disease | Bilateral Symmetric | C | Unilateral | U | Unilateral | U | Bilateral | U | Asymmetric | I | Bilateral | I |
| Increased FNA angle | Both sides (Childhood) | C | Both sides | U | Both sides (Slightly) | U | Affected side | C | Affected side | C | Affected side | R |
| NSA | Normal | I | Increased | U | Decreased | U | Normal | U | Increased | U | NA | NA |
| Morphological Changes — Femoral head | Normal | I | Small | C | Specific changes* | C | Flattening | R | Normal | U | NA | NA |
| Morphological Changes — Femoral neck | Normal | I | Short | C | Short, wide | C | NA | NA | NA | NA | NA | NA |
| Morphological Changes — Acetabulum | Normal | I | Shallow | C | Deformity adaptive to the head | U | Obliquity of the roof (Shallow) | R | Obliquity of the roof (Shallow) | R | Shallow | U |
| Other | Normal | I | NA | NA | NA | NA | NA | NA | Signs of genu valgum, tibial external rotation, genu recurvatum, pes equines | NA | Spina bifida (vertebral level T12–L4) | C |
| Leg: Long Bones Length, Bone Thickness | Normal | I | Normal | I | Normal | I | Decreased | U | Decreased | I | NA | NA |
| Hip Subluxation, Dislocation | No | I | Yes | I | Yes | I | Yes | U | Yes | U | Yes | U |
| Degenerative Hip Joint Disease (Adult age) | No | I | Yes | C | Yes | C | No | C | No | U | Yes | U |

* Deformation, flattening, elongation, irregular articular surface lower than trochanter, lack of fovea capitis, mushroom shape.

Level of importance for the diagnosis: C – Cardinal (necessary for the diagnosis), I – Important (frequently seen in that disease, so it is valuable for the diagnosis), U – Usual (usually happens in that disease, contributes to diagnosis, but its absence does not necessarily exclude it), R – Rare (sometimes or occasionally occur in that condition, so their absence does not exclude the diagnosis). NA - Not Available.

The table data are based on the following references: Aufderheide et al. 1998; Beals 1969; Cibulka 2004; Fitzgerald et al. 2002; Glyn et al. 1966; Hefti et al. 2007; Hubbard et al. 1988; Lau et al. 1986; Morrissy and Weinstein 2006; Noonan 2006; Parsons and Seddon 1968; Rogers 1934; Samilson et al. 1972; Sauser et al. 1986; Shefelbine and Carter 2004; Skinner 2003; Staheli 1977; Subasi et al. 2008; Sugano et al. 1998; Tachdjian 1997; Tachdjian and Minear 1956; Thompson and Salter 1987; Wedge et al. 1989; Wheeless Textbook of Orthopaedics; William and Warner 2003.

(developmental) anteversion. The skeleton from this cemetery did not meet the main criteria for this diagnosis (bilateral and symmetric anteversion, and complete correction of the deformity by skeletal maturity, Table 2), since the observed FNA angle was unilaterally increased and the person was of adult age without correction of the condition.

As it is previously stated, the unilateral presentation can speak in favour of asymmetric form of developmental dysplasia of the hip – DDH (Fabry et al. 1973; Fitzgerald et al. 2002; Subasiet al. 2008; Sugano, Noble, Kamaric et al. 1998), unilateral type of Legg-Calve-Perthes disease – coxa plana (Aufderheideet al. 1998; Thompson and Salter 1987) and neuromuscular disorders – hemiplegic cerebral palsy, myelomeningocele, chronic stage of poliomyelitis, and central motoneuron lesions other than cerebral palsy such as brain or spinal cord tumors or infections (Beals 1969; Bobroff et al. 1999; Cibulka 2004; Fabry et al. 1973; Glyn et al. 1966; Heftiet al. 2007; Lau et al. 1986; Lewis et al. 1964; Morrissy and Weinstein 2006; Murray and Robb 2006; Noonan 2006; Parsons and Seddon 1968; Renshaw and Deluca 2006; Rogers 1934; Samilsonet al. 1972; Sauser et al. 1986; Shefelbine and Carter 2004; Skinner 2003; Staheli 1977; Tachdjian and Minear 1956; William and Warner 2003a; William and Warner 2003b).

Prominently high value of anteversion angle, as well as a lack of specific morphological changes of the proximal femur (changes of the femoral head: deformation, flattening, elongation, irregular articular surface lower than trochanter, lack of the fovea capitis, mushroom shape; and the neck: shortening, widening and decreased neck-shaft angle) did not support Perthes disease as an aetiological possibility in our case.

In unilateral cases of DDH not only is FNA angle increased on the affected side, but unaffected side also displays similar or even higher anteversion (Fabry et al. 1973). Unilaterally high FNA angle in our case, as well as lack of morphological characteristics of DDH (such as small head, short neck, shallow acetabulum) on either side, made DDH a rather unlikely diagnosis. In addition, lack of signs of secondary hip osteoarthritis at adult age was not compatible with either Perthes disease or DDH, given the natural progression of those two conditions which alter biomechanical homeostasis and frequently lead to premature degenerative joint disease (Sugano, Noble, Kamaric et al. 1998).

Unaltered articular surfaces on the femoral head ruled out juvenile rheumatoid arthritis as another possible cause of increased FNA (Harhess 2003).

Therefore, neuromuscular disorder should be further considered as more appropriate explanation for high FNA angle of the female skeleton from Leskovac.

However, we have to note that various neuromuscular disorders may be encompassed in differential diagnosis. One such possibility would be mild monoplegia observed after meningitis. According to William and Warner (2003a) there is a very rare type of paresis which is usually observed after meningitis, and it is seen in the form of monoplegia. The authors described such monoplegia as a form of mild hemiplegia where other ipsilateral extremity is mildly involved. The results of the skeletal analyses in our case revealed just discrete side-specific differences in bone size of humeri and increased FNA of the left femur without any other signs of limb discrepancy which would be concordant to such grade of plegia. As the analyses did not reveal any visible sign of infection on the cranial skeleton, such specific diagnosis cannot be accepted.

Generally, a question arises about what would happened if an older individual (adolescent, young adult or adult) suffered any other type of central motoneuron damage. We considered this scenario as well, particularly as the lack of degenerative joint disease in our case might indicate a shorter time between the onset of paralysis and the death. However, although motoneuron lesions in adults also cause contractures due to muscle imbalance, it was not possible that it would lead to a change in the femoral neck anteversion angle from 8-15 degrees to nearly 60 degrees.

During the period of growth any alteration in child's ambulation, absence of weight-bearing bone stimulation or presence of neuromuscular imbalance caused by a particular pattern of abnormal or spastic muscle group's balance may contribute to further delay or even prevent correction of FNA (Renshaw and Deluca 2006; Sauser et al. 1986; Shefelbine and Carter 2004). Furthermore, it has been suggested that in some cases the same factors could lead to even increasing FNA from the neonatal value (Renshaw and Deluca 2006; William and Warner 2003a).

Therefore, childhood neuromuscular lesion remained the most likely explanation in our case. The lesions of lower motoneurons (flaccid type of paralysis), such as poliomyelitis which is usually

asymmetric and with the known bone deformities in its chronic phase, also were candidates for the diagnosis in this skeleton. Given that paralysis due to poliomyelitis usually occurs before the age of five (William and Warner 2003b; World Health Organization), the growth of the proximal femur is frequently abnormal, leading to coxa valga, persistent femoral anteversion and other deformities (Morrissy and Weinstein 2006; William and Warner 2003b). Osseous growth potential in early childhood makes children more vulnerable to the secondary deformities; thus, the worst deformities develop in young children as well as in those with severe muscle imbalance (Morrissy and Weinstein 2006). Hence, the effect of poliomyelitis on the skeleton is thought to depend on the period of life during which the infection was contracted. If it happens during childhood when the skeleton is still developing, the bones in the paralyzed limbs will be shorter and more gracile than those in the unaffected limb, and anteversion will be increased. On the other hand, if the disease is contracted after skeletal maturity, the bones of the paralyzed limb or limbs will be of the same length as those on the normal side; however, they will be more gracile due to disuse atrophy and will highly unlikely express such a high anteversion angle. In the paleopathological literature the childhood poliomyelitic bone changes on the skeletal remains are ascertained exclusively on the basis of observing limb length discrepancy (Waldron 2009), which was not found in our case. Like in other neuromuscular disorders, coxa valga, i.e., increased femoral neck-shaft angle can be found in an affected femur, which was opposite to the findings from our study. The lack of degenerative joint disease in the individual from Leskovac in spite of her advanced age could speak in favour of poliomyelitis, as poliomyelitis is thought to be associated with very low incidence of osteoarthritis (Glyn et al. 1966). However, the lack of other characteristic features on the skeleton (Table 2) in our case rebutted against poliomyelitis as a diagnostic possibility.

Differential diagnosis should also include myelomeningocele – a complex of congenital malformations of the central nervous system commonly described as spina bifida (Noonan 2006; William and Warner 2003b). Sometimes, this disorder can lead to increased FNA angle (Noonan 2006). This neuromuscular disorder is depicted as hernia protrusion of the spinal cord and its meninges through a

defect in the vertebral canal with consequent variable neurological defects depending on the location and severity of the lesion (William and Warner 2003b). According to Noonan (2006) myelomeningocele usually occurs in the low thoracic and lumbosacral regions (affecting function of the lower extremity) while the cervical region is rarely affected. Given the poor preservation of the vertebrae and the absence of other signs (bilaterality and shallow accetabulum) in our specimen, this diagnosis could not be established.

Overall, in our case, the most likely diagnostic possibility was cerebral palsy – a syndrome resulting from a non-progressive lesion of the developing brain. Although commonly misunderstood, the lesion is actually non-progressive since the cause is acute, but clinical manifestations change as the brain matures. Cerebral palsy can be caused by numerous prenatal, perinatal or postnatal factors. Thus, aetiology is quite diverse: from vascular reasons and birth head trauma, to infections and other causes. Furthermore, even nowadays the accurate aetiology could be identified in only 50% of cases in clinical settings (Berker and Yalçin 2005). Although cerebral palsy is usually diagnosed as any nonprogressive central nervous system injury which occurred below the age of two (Berker and Yalçin 2005; Renshaw and Deluca 2006), aetiological agents could also afflict the central motoneurons chronologically after the defined age producing the same clinical manifestations during childhood. As this differentiation does not have any impact on further prognosis, most authors refer to both lesions as cerebral palsy (Renshaw and Deluca 2006). Deformity and displacement of the hip are, after the equinovarus foot deformity, the second most common orthopaedic problem in children with cerebral palsy presenting an increase in the severity and incidence of hip pathology with the severity of the palsy (Murray and Robb 2006). In cerebral palsy, FNA angle could be normal at birth (Shefelbine and Carter 2004), but would not decrease with growth (Beals 1969; Bobroff et al. 1999; Fabry et al. 1973) and remains high at adult age (55 degrees on average: Lewis et al. 1964). Namely, changes to bone morphology in spastic palsy occur according to Wolff's law and bone functional adaptation principle since the muscle forces acting on bones govern bone remodelling and development (Cibulka 2004). In clinical settings, the diagnosis is straight forward since it takes into consideration the

history of prenatal, perinatal and early postnatal life which is usually easily obtained from the parents of these patients (e.g., birth trauma). Other clinical features of cerebral palsy in its various forms further make this condition easily distinguishable from primary bone and joint diseases. However, considering the lack of data from an archaeological context, diagnosing cerebral palsy in this case is quite challenging.

Given that there are two cardinal and two usual signs on dry bones (Table 2), the morphological features observed on this adult female skeleton were compatible with diagnosis of cerebral palsy. Tachdjian and Minear (1956) already reported that the degree of coxa valga (increased neck-shaft angle) in cerebral palsy is proportional to the loss of muscle power. Normal neck-shaft angle in this case suggested that the woman did not lose the muscle power excessively and that she continued to be active. In cases with neurological deficits, due to disuse of the paralyzed limbs, it can be expected that the bones of the affected extremity are more gracile and even shorter (if the paralysis appeared at an early age), which was not the case in this woman confirming that the individual was active. In addition, Renshaw and Deluca (2006) concluded that FNA values in the patients with cerebral palsy are higher in ambulatory than in non-walkers and observed that it does not change significantly after the age of six.

Comprehensive analysis of diagnostic possibilities led us to the most likely explanation of the increased femoral torsion in our case. Moreover, our study could also serve as diagnostic guidelines for determining the causes of increased femoral neck anteversion in further research in archaeological populations.

## Conclusion

Analysis of the femoral neck anteversion angle in a medieval population from Leskovac revealed a case of unilaterally increased femoral neck anteversion indicative of asymmetric form of cerebral palsy. The differential diagnosis of increased femoral neck anteversion was under-studied in previous archaeological and anthropological literature. Systematic recording of the femoral neck anteversion could reveal the traces of various orthopaedic and neuromuscular disorders in past human populations.

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