SPINA BIFIDA OCCULTA IN MEDIEVAL AND POST-MEDIEVAL SKELETONS FROM IASI CITY, IN NORTH-EAST ROMANIA

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Abstract. Using macroscopic examinations, this paper provides diagnoses of spina bifida occulta (occult spinal dysraphism) in skeletons from the medieval and post-medieval sites of Iasi City in North-East Romania. As a congenital disorder consisting of an incomplete fusion of the posterior neural arch, spina bifida occulta appears mostly in the lumbosacral region, affecting the sacrum. Palaeopatological analysis of the osteological lesions could reveal factors of stress leading to spinal dysraphism: deficiency in the maternal nutritional state, exposure to teratogenic factors and genetic predisposition. The osteological material available for analysis consists of 947 skeletons found in burial and reburial tombs from four necropolises of XVth–XIXth centuries, discovered between 1995 and 2011. Within these populations (18–x years), sacral spina bifida occulta (sacral occult spinal dysraphism) was identified in 10 subjects (eight males aged 18-60 years and two females aged 18-20 years and 40 years, respectively). The incidence of sacral spina bifida occulta is 3.83% in the total population for which the sacrum was preserved (261 sacra) and by gender we recorded 5.19% in the male group (154 sacra) and 1.86% in the female group (107 sacra).

Keywords: spina bifida occulta, paleopathology, medieval and post-medieval, Iasi city, Romania.

Introduction
This study concerns the medieval and post-medieval human populations living in the Iasi city (Romania). The study is mainly focus on the osteological lesions associated with the congenital anomaly named spina bifida occulta.

Congenital anomalies or malformations are produced by pathological changes in the normal development of the embryo during intrauterine life (Aufderheide & Rodriguez-Martin, 1998); some of them can be identified in the new-born babies, but most defects remain undetected until childhood or even adolescence (Marcskik et al., 2002).
Congenital anomalies were recorded from the most ancient times, on the Babylonian clay plates from the Royal Library of Nineveh, assembled by the Syrian king Assurbanipal (700 BC). Traces of congenital malformations in the primitive cultures have been found on some bones of the affected people, specifically on a sacrum and on a femur discovered on the territory of France and dated from the Neolithic period (Savona-Ventura, 2007). Bone anomalies were found in the cranial segment, in the spine and in the appendicular skeleton. The highest incidence of the developmental anomalies was recorded in the spine, followed by the cranial segment and the appendicular skeleton, with lower incidences. Reports on the incidence of congenital malformations in prehistoric populations are very rare, because most of the affected children died during birth or shortly after that (Barnes, 1994).

Developmental anomalies appear due to genetic influences or an environmental stimulus which manifested itself during a critical stage of development, when the developing structures are vulnerable. Most of the developmental anomalies of the skeleton in historical populations are located in the spine, affecting most frequently the lumbosacral region (Masnicová & Beňuš, 2003); thus, spina bifida is the most common congenital defect, characterized by an incomplete fusion of the posterior midline of the osseous tissues, which leaves the spinal cord relatively unprotected (Marcšik et al., 2002). Spina bifida can also be defined as a developmental defect resulting in aplasia or hypoplasia of one or both parts of the neural arch and/or spinous process (Masnicová & Beňuš, 2003). Spina bifida occurs within the first month of pregnancy as a result of a defect in the process of neurulation in which the vertebral column and spinal cord are supposed to fully form and close (Fishman, 2003).

In the XIXth century, specifically in 1886, was published one of the most important monographs on spina bifida, by Friederich von Recklinghausen, who included in his work two remarkable illustrations that clearly outline both the internal and the external pathology of spina bifida. Recklinghausen also observed that some patients with spina bifida survived into adulthood (Marcšik et al., 2002). Spina bifida is the expression of spinal dysraphism (Zemirline et al., 2012), which includes a spectrum of congenital disorders caused by incomplete or abnormal closure of the neural tube during early embryogenesis (Vesna & Nirmala, 1998).

Based on physical observation, cases of spinal dysraphism can be grouped in two categories: serious anomalies such as spina bifida aperta or cystica (posterior protrusion of neural tissue through a bony vertebral defect that results in non-skin covered lesions with exposed neural tissue, such as the meningomyelocele, an open neural tube defect) and mild, asymptomatic lesions, such as spina bifida occulta (occult spinal dysraphism) (Armstrong, 1993; Byrd et al., 1991). All these malformations can induce a variety of neurological defects, such as impaired walking and problems in the bladder function (Gleeson et al., 2006). Hydrocephalus is another effect of spina bifida, which appears due to the excess cerebral spinal fluid accumulated in the brain. It appears in approximately 11-90% of the people with spina bifida, occurring most often in those with a higher lesion level (Dicianno et al., 2008) and those with myelomeningocele (Fishman, 2003). Genetic studies show that occulta and cystic forms of spina bifida are different expressions of the same dominant gene (McKusick, 1998).

Paleoanthropological studies on spina bifida were mainly conducted on the sacrum, although 60% of the physiological effects on the organism are associated with
spina bifida of the L3-S1 and only 10% involve the S2-S5 segments (Kumar & Shane Tubbs, 2011). The reported incidence of spina bifida in the lumbosacral region (L5 and S1) is up to 25% (Barnes, 1994), usually involving only one or two vertebrae, but occasionally several vertebrae can be affected, particularly in the sacrum. Spina bifida occulta of the sacrum is considered the most common type of spinal abnormality (Senoglu et al., 2008).

According to the report on the clinical significance of spina bifida occulta (occult spinal dysraphism), this disorder ranges from mild, asymptomatic lesions to severe defects such as the meningomyelocoele or the neurologic deficiencies (Albrecht et al., 2007).

Many authors published data on the incidence of spina bifida occulta, obtaining different results: the reported frequency ranged greatly among researchers and populations. Spina bifida varies across gender, ethnicity and geographic location. However, its frequency ranges from 1 to 5 cases per 1000 live births (Fishman, 2003). The prevalence of spina bifida occulta decreases with age, possibly as a result of new bone formation or calcification (Mehdizadeh et al., 2010) and males appear to be more frequently affected by developmental delay defects in the sacrum than females (Aufderheide & Rodriguez-Martin, 1998). The reported incidence of spina bifida is higher in Hispanics than it is in Caucasian or African Americans (Shaer et al., 2007).

The presence of spina bifida is linked to vitamin, folic acid and zinc deficiency among mothers. It is argued that vitamin deficiencies, acting together, may interfere with the closure of the neural tube or that such deficiencies may allow some unknown teratogenic factors (agents causing malformations) to have a certain influence (Lovett & Gatrell, 1988). The origin of spina bifida is multifactorial. This anomaly involves a genetic predisposition and environmental factors which act like triggers; however, the precise cause remains unknown (Windham & Bjerkedal, 1982).

The sacrum usually consists of five segments, but in some cases the number of segments can change with either the addition of the sixth lumbar vertebra (lumbarization) or the sacralization of the last lumbar vertebra or the first coccygeal vertebra (White & Folkens, 2005). The clinical significance of the lumbosacral transitional vertebra (lumbarization) has made the subject of many debates and the incidence of this anomaly varies greatly, ranging from 4% to 24% (Delport et al., 2006). Sacralization was described by O’Connell in 1951 and it represents the extension of the sacrum to the lower region of the pelvis, whereas the spinous process of the last lumbar vertebra can be united to the iliac crests or it can be located immediately below them (Prakash et al., 2011). In both lumbarization and sacralization, the defect is complete or incomplete, unilateral or bilateral, symmetrical or asymmetrical (Barnes, 1994).

Paleoanthropological studies in Romania have already addressed preliminary questions related to the demography and pathology, including diagnosis of occult spinal dysraphism, characterizing the medieval and post-medieval populations living in the Iasi city of Romania (Simalcsik et al., 2011; Groza et al., 2012).

Iasi city is located in the North-East of Romania, in the Moldavia region; it was the capital of the Principality of Moldavia from 1564 to 1859. Since XVth century, the historical evolution of the city took place in the conditions of permanent disasters: was often assaulted and burned by Tatars, Turks, Polish or Russians, was hit by starvation periods and plaque. The city had a slow development, being limited in surface and with a small but diverse population (Romanians, Armenians, Hungarians, Polish, Germans or Russians). There are various references to the life of the inhabitants; they were dealers,
craftsmen or farmers (Caprosu & Zahariuc, 1999). A characterization of peripheral slums of Iasi is offered by N.A. Bogdan (1997-2004) who describes these suburbs considered countryside as having small houses, built out of ordinary materials, with unpaved street, with poor people, which worked a lot with their arms, with carting agriculture and cattle and poultry breeding. The description also underlines the insanitary conditions of the slums.

Material and Methods

The osteological material subjected to the study (947 skeletons) was unearthed from four necropolises discovered in the Iasi city area and dated between the second half of the XVth century and the first half of the XIXth century. The archaeological excavations conducted in the Iasi city area (Romania) between 1995 and 2011 by a group of scientists directed by Mrs. Stela Cheptea, archaeologist from the Centre for European History and Civilization in Iasi (Romania).

The first sample, the osteological series exhumed in 1995 from the necropolis of the “Virgin Mary” Catholic Church (XVth-XIXth centuries), are badly preserved and consists of 89 skeletons found in burial and reburial tombs. The second sample, from the necropolis discovered in 2007 on the premises of the “St. Nicholas-Ciurchi” Church (XVIth-XVIIIth centuries), offered for study 680 inhumed and re-inhumed skeletons, most of them also ill-preserved. The third sample, the osteological series excavated in 2008 from the necropolis located on eastern side of the former Princely Court (XVIIth century), consists of 111 inhumation skeletons, is in a satisfying state of preservation. The fourth sample, from the necropolis of XVIth-XIXth centuries discovered in 2011 on the premises of the Banu Church pursuant to the edifice reinforcement and rehabilitation works, contains 67 inhumed and re-inhumed skeletons.

The bone fragments were restored, marked and subjected to a bio-morphoscopic analysis to determine the individual age and gender, the pathologies and the anomalies associated. The precarious state of preservation for some of the skeletons, as well as the absence of bone fragments made it very difficult for us to conduct the anthropological and paleopathological analysis.

The age and sex determination for the subjects over 18 years was accomplished using the methods and procedures recommended by Bruzek (2002), Mays (1998), Brothwell (1981), Schmitt (2005), Walrath et al. (2004). In the case of subadults, the age was estimated based on the primary and permanent teeth stages of development (Ubelaker, 1979; Schaefer et al., 2009), as well as on the analysis of the level of ossification between the long bones epiphyses and diaphyses and their repartition in the corresponding age categories (Maresh, 1955; Fazekas & Kosa, 1978; Jeanty, 1983; Scheuer & Black, 2000).

The size was calculated based on the dimensional scales suggested by Manouvrier (1982), Bach (1965), Breitinger (1938), Trotter & Gleser (1952, 1958).

We were particularly interested in identifying the cases of spina bifida occulta (occult spinal dysraphism), tracing to that effect the abnormalities in the midline of the spine, regardless of their position. In this study we took into account only the subjects aged 18-x years. We macroscopically established for each subject if spina bifida occulta was present or absent in the sacrum, since this particular anomaly can be easily recognized in a skeleton (Waldron, 2009). Where we identified this abnormality, we also marked its location. In order to accurately evaluate the prevalence of this lesion, one must analyze its incidence in relation to the chronological age. If hydrocephalus was absent, the anomaly is
classified as spina bifida occulta or occult spinal dysraphism. The presence of spina bifida occulta in the segments S3, S4 and S5 should come under the normal range of variation of the sacral hiatus. However, all cases of spinal dysraphism must be recorded regardless of their incidence in the spine (Kumar & Shane Tubbs, 2011). As regards total sacral spina bifida occulta, the posterior lamellae of all sacral vertebrae are completely unfused (Senoglu et al., 2008). Besides spina bifida occulta, we also identified other anomalies, such as sacralization and lumbarization. Complete sacralization consists of a total union between the spinal process of the last lumbar vertebra (L5) and the sacrum. Incomplete sacralization shows a well-defined joint line between the process and the sacrum. If the first sacral segment (S1) is separated from the sacrum, the defect is called lumbarization (Aufderheide & Rodriguez-Martin, 1998; Barnes, 1994).

Results and Discussion
Analyzing by age and gender the entire osteological material (947 skeletons) exhumed from the four necropolises of the XVth-XIXth centuries found in the Iasi city area, the mortality rate for children aged 0-14 years is approximately 17%. The death rate recorded for adolescence is about 8%, for adults (20-30 years) is 12%, and 57% for mature people (30-60 years); the senile people (60-x years) recorded about 6%. The ratio between the number of deceased males and the number of deceased females is 1.06 – there are a higher number of male skeletons compared to the female skeletons (406/383).

In the total number of subjects aged 18-x years (744 skeletons), we identified 10 cases of sacral spina bifida occulta (8 males aged 18-60 years and 2 females aged 18-20 years and 40 years, respectively). The incidence of the sacral spina bifida occulta is 3.83% in the total number of subjects for whom we retrieved the sacrum (261 sacra) and by gender we recorded 5.19% in the male group (154 sacra) and 1.86% in the female group (107 sacra).

First sample
From the total of 89 human skeletons unearthed from the necropolis of the “Virgin Mary” Catholic Church (XVth-XIXth centuries), the mortality rate for the subjects under 20 years is approximately 24%, of which 20.23% (18 subjects) is associated with children aged 0-14 years. In the subjects past the age of 20, the maximal death frequency is recorded in the maturity stage (approximately 57% - 51 subjects: 28♂ and 23♀), whereas in the adulthood stage the mortality rate is 9% (eight subjects - 3♂ and 5♀). In the age interval 60-x years we recorded a mortality rate of 10.11% (nine subjects: 5♂ and 4♀). In the segment 18-x years (71 subjects), the sacrum is present in only 28 of the subjects.

From the total 28 sacra, just one female sacrum (subject M5) belonging to a subject aged approximately 40 in the moment of death, presents spina bifida occulta (occult spinal dysraphism). Thus, the incidence of spina bifida occulta in this population is 3.57% if we consider the total number of subjects in which the sacrum is present and 10% if we refer to the female group.

Subject M5. The cranial segment of the female subject in question is badly preserved and presents a high degree of fragmentation. A fragment of the calvaria – the parietal-temporal area; a small bone fragment is also missing from the left side of the occipital. The facial massif lacks the palate area, the left
cheek bone and the left half of the mandible. The postcranial segment lacks the following components: the thoracic vertebrae T2, T3, T4, T5 and T12, the lumbar vertebrae L3 and L4, the tibiae and the fibulae, the left calcaneus and the astragals.

The stature calculated by the long bones is 1673 mm on average, which falls under the tall category.

The sacrum of the subject M5 has a posterior interlaminary dehiscence in the L5-S1 and S3-S5 segments (spina bifida occulta) – the posterior laminae of the vertebrae didn’t fuse in these segments. Besides spina bifida occulta, in this sacrum we can also observe the sacralization of the last lumbar vertebra – L5 (Fig. 1).

Second sample
The second osteological series, unearthed from the necropolis of the “St. Nicholas-Ciurchi” Church (XVI\textsuperscript{th}-XVIII\textsuperscript{th} centuries) contains 680 skeletons: 179 subadults, (infans I, infans II and juvenis) and 501 subjects (224♂ and 278♀) past the age of 20 years (adultus, maturus, senilis). The mortality rate in the subadult segment, meaning subjects aged 0-20 years is considerably high (10.29% for infans I, 7.65% for infans II și 8.38% for juvenis - 34♂; 23♀), which means that approximately one quarter of the population (26.32%) didn’t reach adulthood. In the age interval 20-x years, the highest death rate is recorded for the subjects in the maturus category (55.44% - 176♂; 201♀), whereas the rate for the adultus is 12.35% - 28♂; 56♀, and for the subjects past the age of 60 (senilis) we recorded a mere 5.88% (21♂; 19♀).

The sex ratio (the ratio between the number of males and females) considered for the entire population indicates a slightly higher prevalence of the female skeletons compared to the male skeletons (299 females and 259 males).

In the total number of subjects aged 18-x years (519 individuals), the sacrum is present in 129 cases (62♂ and 67♀).

Only two sacra out of 129 present spina bifida occulta (occult spinal dysraphism); both sacra belong to male subjects aged 30-45 years. Sacral occult spinal dysraphism (sacral spina bifida occulta) represents 1.55% of the total number of subjects in which the sacrum was preserved and 3.22% of the male subjects.

Subject R113F. The first case of sacral spina bifida occulta was identified in a male subject (subject R113F) aged 30-35 years and 1645 mm tall (medium-sized). This well preserved skeleton was exhumed from a tomb which also contained re-inhumations. The sacrum of the subject has two dehiscences: the first one is located in the S1 segment, whereas the second is in the S3-S5 segment (Fig. 2).

In this case, the two interlaminary sacral dehiscences are associated with other pathologies: the arthritis of the clavicles in the acromioclavicular joint area and osteophytes on the radial tuberosity; the right coxal bone – with osteophytes on the iliac crest; the thoracic vertebrae T6-T12 and the lumbar vertebrae L1-L3 have osteophytes on the edges of the vertebral bodies and Schmorl nodules.

Subject M126A. The second skeleton with sacral spina bifida occulta also belonged to a male subject (subject M126A), aged 40-45 years. This skeleton is incomplete, since in the cranial segment we only have the frontal bone. In the postcranial skeleton lacks the right astragal, fragments of the shoulder blades, the left clavicle, humeri, the left ulna and the left radius.
The sacrum has a posterior interlaminary dehiscence (sacral spina bifida occulta) in the S₂-S₅ segment (Fig. 3).

**Figure 1.** Subject M5 - ♀, 40 years: posterior view of sacrum - spina bifida occulta in the L₅-S₁ and S₃-S₅ segments, associated with the sacralization of L₅.

**Figure 2.** Subject R133F - ♂, 30-35 years: posterior view of sacrum - sacral spina bifida occulta in the S₁ and S₃-S₅ segments.

**Third sample**

In the osteological series unearthed from the eastern side of the Princely Court (XVIIᵗʰ century) we identified 111 skeletons, of which 15 subadults (juvenis) (10♂ and 5♀ - 13.51%), 21 adults (12♂ and 9♀ - 18.92%), 67 mature people (56♂ and 17♀ - 65.77%) and two senile males (1.80%) – the number of male skeletons was significantly higher compared to the female skeletons (80/31). No death was recorded in the age interval 0-14 years.

The sacrum is present in 66 males and 25 females (91 subjects) of the total series, which included 106 subjects (18-x years). We observed that six sacra in the total of 90 bones present spina bifida occulta – five sacra belonged to males aged 18-60 years and a sacrum belonged to a female aged 18-20 years. Spina bifida occulta recorded an incidence of 6.59% in the total number of subjects, affecting 6.57% of the males and 4% of the females.

**Subject M XIV.** The first case of sacral spina bifida occulta was identified in a male subject (subject M XIV), aged 18-19 years. The well preserved and almost integral skeleton was exhumed from an individual tomb.

The subject’s height is 1657 mm (medium-sized). The tibiae and the humeri have bone excrescences at the muscle insertions. The sacrum presents occult spinal dysraphism categorized as posterior spina bifida occulta with a dehiscence located in the S₁ and S₃-S₅ segments (Fig. 4).

The dehiscence located in the S₁ segment represents 20% of the total types of closed spinal dysraphism. This form of dysraphism doesn’t present associated clinical
symptoms and the sacral hiatus identified in the S3-S5 segment is frequently reported in the scholarly literature (Tortori-Donati et al., 2000).

Subject G8 M25. Another case of spina bifida occulta was noted in a female subject (subject G8 M25) aged 18-20 years, who was buried in the same tomb with two other mature subjects aged 40-50 years. From the cranial skeleton of this female we only retrieved the frontal bone and a fragment of the occipital bone. The incomplete sacrum presents a posterior interlaminary dehiscence in the S4-S5 segment (sacral spina bifida occulta) – the posterior laminae of the sacral vertebrae failed to fuse in this segment (Fig. 5).

Subject M XIII. The male subject (subject M XIII) aged 25-30 years in the moment of death has an incomplete postcranial skeleton; however, the bone fragments are well preserved (the calculated stature is 1671 mm – over-medium sized). The sacrum presents sacral spina bifida occulta in the S2 and S3-S5 segments (Fig. 6a) associated with the incomplete central anterior lumbarization (separation) of the first sacral vertebra S1 from the rest of the sacrum’s body (Fig. 6b).
Figure 6. Subject M XIII – ♂, 25-30 years: a. posterior view of sacrum - sacral spina bifida occulta in the S2 and S3-S5 segments; b. anterior view of sacrum – incomplete central lumbarization of the first sacral vertebra (S1).

Subject M IV. The cranial segment of the subject M IV belonging to an adult male aged 25-30 years presents a dip near the coronal suture caused by a blow; it also has nine Wormian bones on the lambdoid suture. The postcranial segment lacks the tibiae and the fibulae, a fragment of the left femur and the right calcaneus. The sacrum has two dehiscences: the first one is located in the S1 segment and the second is in the S3-S5 segment (Fig. 7).

Figure 7. Subject M IV - ♂, 25-30 years: posterior view of sacrum – sacral spina bifida occulta in the S1 and S3-S5 segments.

Subject G10 M33-A. Another case of spina bifida occulta was identified in a male mature subject (aged 40-45 years), marked as G10 M33-A. He was buried next to three other subjects: a male aged 40-45 years and two females aged 20-55 years. The skull is absent and the postcranial skeleton lacks the femurs, the tibiae and the fibulae, the left radius and the left ulna. The subject’s stature is 1796 mm (falls under the tall category). In this case, there is a total spina bifida occulta from the L5 segment to the S1-S5 segment (Fig. 8a), associated with the central sacralization (fusion) of the last lumbar vertebra (L5) to the body of the sacrum (Fig. 8b).
Subject M XI. The last case of sacral spina bifida occulta, in the third series, was identified in a mature male (55-60 years) marked as M XI, with a stature of 1667 mm (medium-sized). The subject has in the cranial segment, specifically on the right parietal, a hole caused by an ante-mortem blow. The thoracic vertebrae (T8-T11) of the subject have marginal osteophytes. We observed a total spina bifida occulta S1-S5 (posterior interlaminary dehiscence) and a channel formed between the laminae (Fig. 9).

Fourth sample
The osteological material (67 skeletons of XVIth-XIXth centuries) unearthed from
the necropolis of the Banu Church is mostly derived from reinhumation tombs. We found 18 skeletons of children (0-14 years: approximately 27%), three teenagers (2♂ and 1♀ - approximately 4%), two adults (2♂ - 2.99%), 40 mature people (22♂ and 18♀ - 59.70%) and four senile people (3♂ and 1♀ - 5.97%).

In this osteological series, we retrieved 13 sacra (8♂ and 5♀) from the total of 48 subjects, aged 18-x years. Among these 13 bones there was a single case of spina bifida occulta in a male subject (subject M45) aged 50-55 years. The incidence of sacral spina bifida occulta (sacral occult spinal dysraphism) is 7.69% in the total number of subjects for which the sacrum is present and 12.50% in the male group.

**Subject M45.** The skeleton exhumed from an individual tomb is incomplete. The cranial segment lacks the base, a fragment of the occipital bone, the left mastoid process and the left side of the facial skeleton. The skull has four Wormian bones on the lambdoidal suture and two Wormian bones on the parietal-temporal suture. The postcranial skeleton lacks the following components: fragments of the coxal bones, the right tibia and the right fibula, the radius and the left ulna.

Apart from the sacrum, all the other elements of the vertebral column are represented only by fragments of vertebral bodies.

The sacrum of the subject M45 shows signs of spina bifida occulta (Fig. 10) with a dehiscence located in the S3-S5 segment.

**Conclusions**

This research assesses the frequency of sacral spina bifida occulta (occult spinal dysraphism) in medieval and post-medieval populations inhabiting the city of Iasi (XVth-XIXth). The human remains subjected to analysis (947 skeletons) were exhumed from four necropolises discovered in the Iasi city area, between 1995 and 2011.

We have found no case of cervical, thoracic or lumbar interlaminar dehiscences.

The osteological data generated in this research suggest that the frequency of sacral spina bifida occulta is relatively low. Thus, the incidence of sacral spina bifida occulta is 3.83% in the total population for which the sacrum was preserved (261 sacra) and by gender we recorded 5.19% in the male group (154 sacra) and 1.86% in the female group (107 sacra).

This paper makes a contribution to our understanding of health during the medieval and post-medieval times. Spina bifida occulta (occult spinal dysraphism), as abnormality defined by an incomplete fusion of the osseous tissues in the midline, has a multifactorial origin involving both a genetic predisposition and environmental factors which act like triggers. The association between the two categories of factors explains why spina bifida doesn’t “run” in the family like other genetic disorders.

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