Memoirs of the Scientific Sections of the Romanian Academy
Tome XXXIX, 2016

ANTHROPOLOGY

SPINA BIFIDA OCCULTA IN MEDIEVAL AND POSTMEDIEVAL TIMES IN EASTERN ROMANIA

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This paper provides bioarchaeological evidence of *Spina Bifida Occulta* (SBO) in human skeletons discovered in medieval and post-medieval sites of Eastern Romania and evaluates its prevalence and patterns by examining its relationship to age, sex, type of sites (urban, elite and rural), and its morphological patterns.

The skeletons found in burial and reburial tombs from five necropolises of the 14th–19th centuries, discovered in Eastern Romania, were analysed to determine individuals' age and sex, pathologies and anomalies. Sacral SBO was identified in 11 subjects, for an overall prevalence of 4%; sacral SBO seems to be more common in endogam groups, as it is assumed to be that of the Princely Court. Apparently, this defect is more frequent in men (9 cases were males), its prevalence decreasing with age (from an overall value of 5.47% in young adults to 2.85% in old ones), but no statistically valid association could be demonstrated between the presence/absence of SBO and sex or age. Morphologically, in 8 cases, the sacral SBO corresponded to a more than 50% opening in the posterior arch.

Keywords: bioarchaeology, sacral *spina bifida occulta*, frequency, morphology.

1. INTRODUCTION

Bioarchaeological studies on human skeleton remains reveal that most of the developmental anomalies of the skeleton in ancient populations are located in the spine, affecting frequently the lumbosacral region [16]. Paleopathological analyses indicate *spina bifida* as a common congenital defect, characterized by an incomplete fusion of the posterior midline of the vertebral arch, which leaves the spinal cord relatively unprotected [15]. Kumar and Tubbs [14] recommended the simple assigning of the name *spina bifida* only to the anomalies noticed on the middle line of the spine, no matter the location level. In the absence of meningocele or meningomyelocele, whichever the size of the spine lesion, the anomaly is not an open *spina bifida*, its mentioning as *spina bifida occulta* being compulsory.

Mild forms of *spina bifida* are easily recognised on skeletons and are well represented in paleoanthropological collections. Sacral *spina bifida occulta* was

repeatedly reported as a congenital anomaly, representing a valuable instrument for detecting specific features in a population, such as the degree of kinship. A high prevalence of sacral *spina bifida* is interpreted as a meaningful indicator of biological isolation and endogamy [14]. In the international literature, most paleoanthropological studies regarding *spina bifida* focused on the sacrum, even if 60% of the complications affecting human physiology are associated to *spina bifida* at a level of L3-S1 and only 10% at a level of S2-S5 [14].

The reported frequency of *spina bifida occulta* ranges greatly among researchers and populations; it varies also across gender, ethnicity and geographic location. However, its actual frequency seems to range from 1 to 5 cases per 1,000 live births [6]. The prevalence of *spina bifida occulta* decreases with age, probably because the subjects affected with such a disease died earlier or, possibly, as a result of new bone formation or calcification [18]; males appear to be more frequently affected by developmental delay defects in the sacrum than females [1]. The reported incidence of *spina bifida* in actual populations is higher in Hispanics than in Caucasians or African Americans [23].

This study contributes to better understanding the prevalence of *spina bifida occulta* in medieval and post-medieval times in Eastern Romania, and its results may enrich the forensic anthropology with data on the differences of this abnormality within old populations.

2. MATERIALS AND METHOD

The paleoanthropological assemblages subjected to the study were exhumed from five necropolises discovered in Eastern Romania (Table 1).

Table 1
Paleoanthropological assemblages subjected to the study

Necropolis, chronology	**Type of settlement**	**Location**	**Number of skeletons**	**Excavation year/ references**
"Virgin Mary" Catholic Church (15^{th}–19^{th} centuries)	Urban	Iaşi (Iaşi County)	89	1995 / [9–10]
"Saint Nicholas-Ciurchi" Church (16^{th}–18^{th} centuries),	Urban	Iaşi (Iaşi County)	680	2007 / [24]
Banu Church (16^{th}–19^{th} centuries)	Urban	Iaşi (Iaşi County)	67	2011 / [9–10]
Princely Court (17^{th} century)	Elite	Iaşi (Iaşi County)	111	2008 / [7, 8, 11]
Dărmăneşti(14^{th}–15^{th} centuries)	Rural	Dărmăneşti (Neamţ County)	32	2012 / [12]

The preserved skeletal fragments were restored, marked and subjected to morphoscopic analysis, to determine individuals' age and sex, pathologies and

anomalies. There are some limitations to this study, caused by the precarious state of preservation of some skeletons, as well as by the absence of certain bone fragments.

Paleoanthropological examination of all skeletons revealed the presence of *spina bifida oculta* predominantly in the sacral region. The status of skeleton conservation is variable in the five necropolises and, consequently, the rate of adult sacrum preservation is different (Table 3), namely very high in elite and rural necropolises – about 85% sacra of all adult skeletons at the Princely Court of Iaşi (17th century) and 82% at Dărmăneşti, and very low in the three urban necropolises – about 26% sacra, on the average. This low proportion of adult sacra can be caused by the re-inhumation of skeletons – a practice that has been identified in the urban necropolises of Iaşi, but not in the elite and rural necropolises.

The present study divided the assemblages of human skeletons into groups, according to age and sex criteria. Only the adult subjects, aged 18–x years, have been analysed. For adult subjects over 18 years, age was assessed based on dental wear, but also with different skeletal age indices, according to the methods recommended in literature [2, 17, 22, 28]. Sex was determined from skeletal morphometry, using standard methods [2, 3, 17, 22, 27].

We were particularly interested in identifying the cases of *spina bifida*, however, prior to offering a diagnosis of this defect in neural arch fusion, the skeletons were thoroughly examined, for evidencing the presence of possible associated skeletal abnormalities (*i.e.* sacralisation and lumbarisation, Wormian bones). Estimation of kinship degrees based on the presence of *spina bifida occulta* could be done only when such assertions can be supported by supplementary proofs [14].

The frequency of *spina bifida occulta* observed in each sample is based on the number of identified sacra; its prevalence was expressed percentually in each sample, on also calculating the standard error (SE) of this ratio. To test the associations between the presence/absence of SBO and sex or age, the statistical Fisher's Exact Test (the probability value being calculated with two sides), recommended for the analysis of small samples, was applied [29].

The cases of *spina bifida occulta* have been classified into five categories, considering to the longitudinal extent of the sacral canal opening [25]: type I – the sacral canal is completely open starting from the top of the S1 spine, continuing up to sacral hiatus at tip of sacrum; type II – the sacral canal is open below S1 down to S5; type III – open sacral canal below S2 down to S5; type IV – the sacral canal is open below S3 to S5; type V – the sacral canal is open between S1-S2 and also between S3-S5.

Table 2
Distribution of skeletons according to age and sex

Necropolis		Total number of skeletons	Age				Sex of adults (18–x years)			
			Young (0–18 years)		Adult (18–x years)		Male		Female	
			Nr.	%	Nr.	%	Nr.	%	Nr.	%
"Virgin Mary" Catholic Church (15th–19th centuries)		89	18	20.22	71	79.78	38	53.52	33	46.48
"Saint Nicholas-Ciurchi" Church (16th–18th centuries)		680	161	23.68	519	76.32	234	45.09	285	54.91
Banu Church (16th–19th centuries)		67	19	28.36	48	71.64	28	58.33	20	41.67
Total urban		836	198	23.68	638	76.32	300	47.02	338	52.98
Princely Court (17th century)	Elite	111	4	3.60	107	96.40	78	72.90	29	27.10
Dărmăneşti (14th–15th centuries)	Rural	32	15	46.88	17	53.12	9	52.94	8	47.06
Total		**979**	**217**	**22.17**	**762**	**77.83**	**387**	**50.79**	**375**	**49.21**

3. RESULTS AND DISCUSSION

Age and sex analysis of the skeletal assemblages exhumed from the five necropolises of the 14th–19th centuries found in Eastern Romania showed that the frequency of adults with ages over 18 years varies from one type of settlement to another. In the city of Iasi (urban necropoles), the percentage of adult individuals is, on the average, about 76%, whereas in the rural site of Dărmăneşti this frequency is much lower, reaching only 53%. The percentage of adults is extremely high (96%) at the Princely Court of Iaşi (elite settlement), where no young, 0–14 year-old individuals were identified. These differences, evidenced in Table 2, may be related to the living conditions that affect the mortality of young individuals.

The ratio between the number of male and female skeletons is, with the exception of two necropolises (Table 2), slightly higher for men, with an average of 1.2 considering both urban and rural sites. In the necropolis of "Saint Nicholas-Ciurchi" Church (16th–18th centuries), the ratio between sexes is slightly higher for women – 1.2, while at the Princely Court of Iaşi (17th century) this ratio is obviously balanced in favor of men – 2.7.

3.1. GENERAL PREVALENCE

In the five analyzed necropolises, 11 cases of *spina bifida occulta* were identified, the general prevalence being of 4% *vs* the total preserved adult sacra (275). The prevalence rates according to sex and age were calculated using as denominator adult individuals with complete sacral spine. If we compare the overall prevalence of *spina bifida occulta* in medieval and postmedieval populations of

Eastern Romania with the results of paleopathological studies concerning other sites from Europe, we observe a similarity with the medieval Hungarian population – 4.2% [13], but not with other samples – *e.g*. Roman Britain, with 51% [26], and the 18th–19th century population of London, with 15.2% [21]. Nevertheless, in our samples, the frequency of *spina bifida occulta* could be actually higher, if considering that it had been determined only on the sacral region.

As Tables 3–4 show, the percentages of cases with *spina bifida occulta* vary from one necropolis to another, as well as according to age and sex. In order to estimate for a more accurate variability of the *spina bifida occulta* prevalence, the cases of the first three urban necropolises were totalized, as they belong to the same city. The prevalence of *spina bifida occulta* is higher in the elite sample (6.59%) than in the urban collection (2.35%). The high percentage recorded in the rural necropolis (7.1%) could be an overestimation, due to the small size of the sample. An increased frequency of *spina bifida occulta* can indicate an endogamous group, if considering the role of the hereditary factor in inducing this defect, as also suggested by epidemiologic or paleoanthropological studies. Eubanks and Cheruvu [4], studying a large osteological sample of 3,100 specimens, representing a population living in Ohio State of USA one century ago, found out that *spina bifida occulta* was about half more common in whites than in blacks. Zemirline *et al*. [30], analyzing a skeletal sample of 30 individuals excavated from a medieval necropolis of southwest Brittany, found out a higher frequency of *spina bifida occulta* (about 10%). The authors also explained the contribution of genetic factors: the Celtic origin of the Britton people, who migrated from Great Britain in the High Middle Ages, given that the actual Britton population presents a higher frequency of spinal dysraphism, like the Celtic people [20]. In the case of our elite sample representing the Princely Court (17th century), we suspect familial relationships, supported with additional evidence of metric characters (*e.g*., a relatively tall stature, on the average, of 169 cm at males and 165 cm at females) and nonmetric data (*e.g.*, Wormian bones in 25 subjects and methopic suture in 7 subjects) [7].

3.2. RELATIONSHIP WITH SEX AND AGE

Among the 11 subjects with *spina bifida occulta* only two are females, one discovered in an urban necropolis and one at the Princely Court (Table 3). However, the results of Fisher's Exact Test, applied to each sample and also to total subjects, point to no statistically valid association between the presence/ absence of *spina bifida occulta* and sex (Table 3). Furthermore, there are different opinions in the literature of the field: some authors discovered this defect more common in male than in female skeletons [4, 13], but others found the same sex ratio in *in vivo* radiological surveys or even more common in females than in males [19].

Analysis of the prevalence of *spina bifida occulta* with respect to adult age groups suggests that this defect is present through-out adult life. The group of young adults (18-29 years) seems to be affected in a higher proportion (14.28%) at the Princely Court. As an exception, this result can support the previously formulated hypothesis regarding the familial relationships in this group. The next group, of 30–49 years, is affected in all three types of samples (urban, elite and rural) in proportions ranging between 1.81% at the Princely Court and 20% at Dărmăneşti. The oldest adults (50–x years) also show this defect, but only in the urban collection (1.69%) and in the elite sample (12.5%). Several authors [5, 14, 20] observed that individuals with *spina bifida occulta* tend to die young, or/and there is a tendency for degenerative calcification filling in the spinal defect, so that an overall prevalent tendency of an age-related decrease might be manifested (*i.e.*, in our study, from 5.47% in the group of 18–29 years to 3.78 at 30–49 years and to 2.85% in the group of over 50 years).

For testing the association between the presence/absence of SBO and age, we used the categories of 18–49 years and 50–x years. Similarly with previous examinations, the results of Fisher's Exact Test, also applied to each sample and to all subjects, point to no statistically valid association (Table 4).

3.3. MORPHOLOGY

Considering the morphology of the lumbo-sacral region, a variety of defects may be described among the 11 analysed subjects (Table 5). Three subjects show transitional abnormalities at the lumbo-sacral junctions besides *spina bifida occulta*: one 40 year-old female, from the necropolis of "Virgin Mary" Catholic Church (15^{th}–19^{th} centuries), evidenced total sacralization of the last lumbar vertebra (L5) (Fig. 1); two males from the necropolis of the Princely Court (17^{th} century) – a 40–45 year-old one, with partial sacralization of L5 (fusion of the vertebral arch and incompletely of the vertebral body), and a 25–30 year-old one with partial lumbarization of the first sacral vertebra S1 (fusion of the vertebral arch).

Five subjects with *spina bifida occulta* of V type were found. In four cases, the defect is located at the levels of S1 and S3-S5 (subjects M5 – Fig. 1; R113F; MXIV – Fig. 4; MIV); in only one case, at the S2 and S3-S5 levels (subject MXIII – Fig. 5a, b). Subject M5, a female of 40 years previously mentioned with sacralization of L5, shows also an extension of *spina bifida occulta* into L5 (Fig. 1). Two specimens indicate type I of sacral *spina bifida occulta*, affecting all five vertebrae. Both subjects belong to the Princely Court necropolis – one male of 40–45 years previously mentioned as having partial sacralization of L5 (in this case, L5 is also affected by *spina bifida occulta*) (Fig. 2a, b), and an older male, of 55–60 years (Fig. 3). Two specimens show type IV of *spina bifida occulta* located on S4-S5 (subjects G8 M25 and M18), one has type II on S2-S5 (subject M126A – Fig. 6), and one – type III on S3-S5 (subject M45).

Table 3

Prevalence (Prev. %) of *Spina Bifida Occulta*: presence (+) of defects according to sex, in skeletons with sacral spine (SE = standard error of *Spina Bifida Occulta* ratio in the sample). Fisher's Test for the association between the presence/absence of SBO and sex

Necropolis	Preserved sacrum		Male adults				Female adults				Total adults			
	Nr.	%*	N	+	Prev. %	SE	N	+	Prev. %	SE	N	+	Prev. %	SE
"Virgin Mary" Catholic Church (15^{th}–19^{th} centuries)	28	39.43	18	–	–	–	10	1	10.0	9.48	28	1	3.57	3.50
"Saint Nicholas-Ciurchi" Church (16^{th}–18^{th} centuries)	129	24.85	62	2	3.22	2.24	67	–	–	–	129	2	1.55	1.08
Banu Church (16^{th}–19^{th} centuries)	13	27.08	8	1	12.50	11.69	5	–	–	–	13	1	7.69	7.38
Total urban	170	26.64	88	3	3.41	1.93	82	1	1.22	1.21	170	4	2.35	1.16
	Fisher's Test = 0.6215; not statistically significant													
Princely Court (17^{th} century) **Elite**	91	85.04	66	5	7.57	3.25	25	1	4.0	3.91	91	6	6.59	2.60
	Fisher's Test = 1.0000; not statistically significant													
Dărmăneşti (14^{th}–15^{th} centuries) **Rural**	14	82.35	7	1	14.28	13.22	7	–	–	–	14	1	7.14	6.88
	Fisher's Test = 1.0000; not statistically significant													
Total	**275**	**36.08**	**161**	**9**	**5.59**	**1.81**	**114**	**2**	**1.75**	**1.22**	**275**	**11**	**4.00**	**1.18**
	Fisher's Test = 0.1299; not statistically significant													

* Relative to the total number of adult skeletons

Table 4

Prevalence (Prev. %) of *Spina Bifida Occulta*: presence (+) of defects according to age, in skeletons with sacral spine (SE = standard error of *Spina Bifida Occulta* ratio in the sample). Fisher's Test for the association between the presence/absence of SBO and age

Necropolis	18–29 years				30–49 years				50–x years			
	N	+	Prev. %	SE	N	+	Prev. %	SE	N	+	Prev. %	SE
"Virgin Mary" Catholic Church (15^{th}–19^{th} centuries)	6	–	–	–	12	1	8.33	7.96	10	–	–	–
"Saint Nicholas-Ciurchi" Church (16^{th}–18^{th} centuries)	31	–	–	–	56	2	3.57	2.47	42	–	–	–
Banu Church (16^{th}–19^{th} centuries)	2	–	–	–	4	–	–	–	7	1	14.28	13.22
Total urban	39	–	–	–	72	3	4.16	2.35	59	1	1.69	1.67
	18–49 years						**50–x years**					
	Fisher's Test = 1.0000; not statistically significant											
Princely Court (17^{th} century) **Elite**	28	4	14.28	6.61	55	1	1.81	1.79	8	1	12.5	11.69
	18–49 years						**50–x years**					
	Fisher's Test = 0.4337; not statistically significant											
Dărmăneşti(14^{th}–15^{th} centuries) **Rural**	6	–	–	–	5	1	20.0	17.88	3	–	–	–
	18–49 years						**50–x years**					
	Fisher's Test = 1.0000; not statistically significant											
Total	**73**	**4**	**5.47**	**2.66**	**132**	**5**	**3.78**	**1.65**	**70**	**2**	**2.85**	**1.98**
	18–49 years						**50–x years**					
	Fisher's Test = 0.7350; not statistically significant											

Table 5
Subjects showing *Spina Bifida Occulta* (SBO)

Necropolis	Subject	Sex and age	Type: segments affected by SBO	Other skeletal abnormalities
"Virgin Mary" Catholic Church (15th–19th centuries)	Subject M5	Female, 40 years	V: L5-S1; S3-S5	Sacralization of L5.
"Saint Nicholas-Ciurchi" Church (16th–18th centuries)	Subject R113F	Male, 30–35 years	V: S1; S3-S5	–
	Subject M126A	Male, 40–45 years	II: S2-S5	–
Banu Church (16th–19th centuries)	Subject M45	Male, 50–55 years	III: S3-S5	Four Wormian bones on the lambdoid suture and other two on the squamosal suture
Princely Court (17th century)	Subject G8 M25	Female, 18–20 years	IV: S4-S5	–
	Subject MXIV	Male, 18–19 years	V: S1; S3-S5	–
	Subject MXIII	Male, 25–30 years	V: S2; S3-S5	Incomplete central anterior lumbarization (separation) of the first sacral vertebra (S1)
	Subject MIV	Male, 25–30 years	V: S1; S3-S5	Nine Wormian bones on the lambdoidal suture
	Subject G10 M33-A	Male, 40–45 years	I: L5+S1-S5	Sacralization (fusion) of L5
	Subject MXI	Male, 55–60 years	I: S1-S5	–
Dărmăneşti(14th–15th centuries)	Subject M18	Male, 40–45 years	IV: S4-S5	–

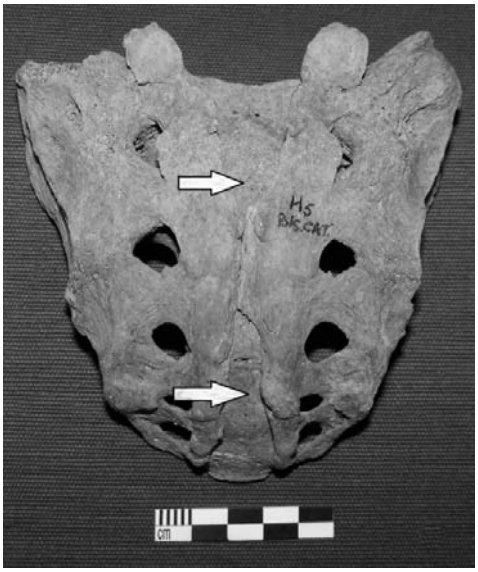
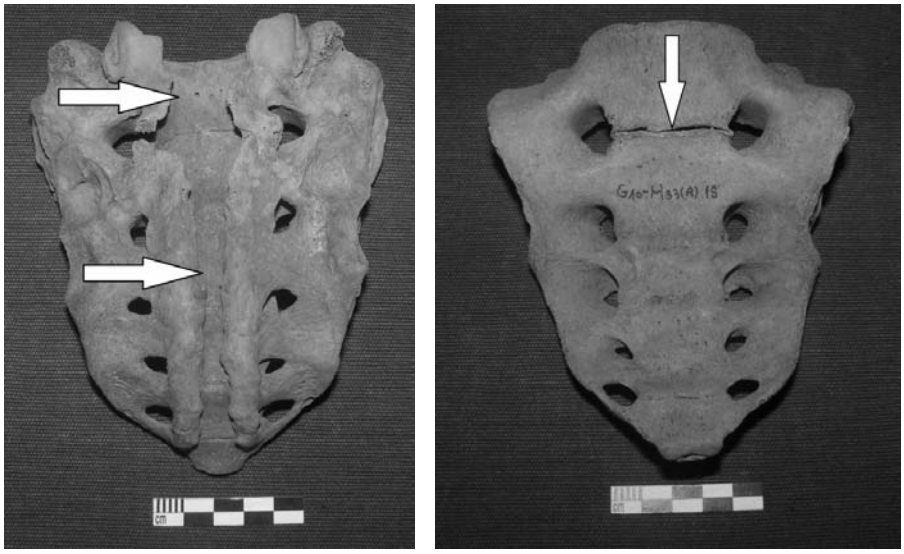


Fig. 1. Subject M5 – ♀, 40 years: posterior view of sacrum – *spina bifida occulta* in the L5-S1 and S3-S5 segments, associated with the sacralization of L5.



a b

Fig. 2. Subject G10 M33-A – ♂, 40–45 years: a. posterior view of sacrum – *spina bifida occulta* in the L5-S5 segment; b. anterior view of sacrum – sacralization of L5.

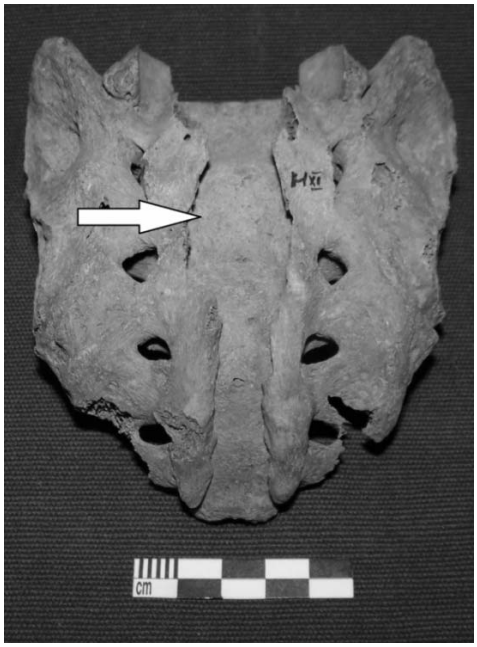


Fig. 3. Subject MXI – ♂, 55–60 years: posterior view of sacrum – total sacral *spina bifida occulta* (S1-S5).

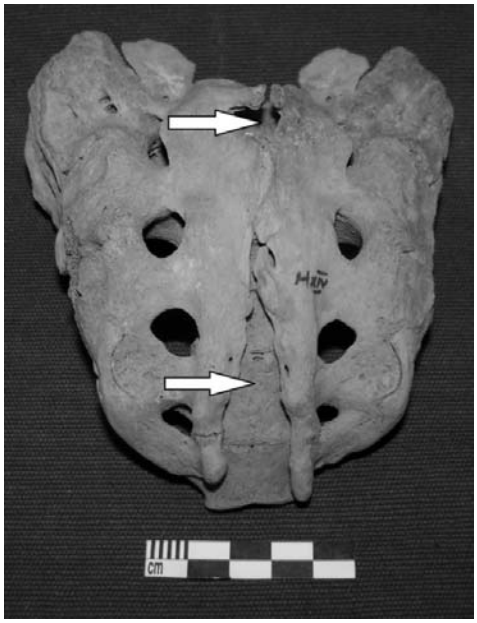
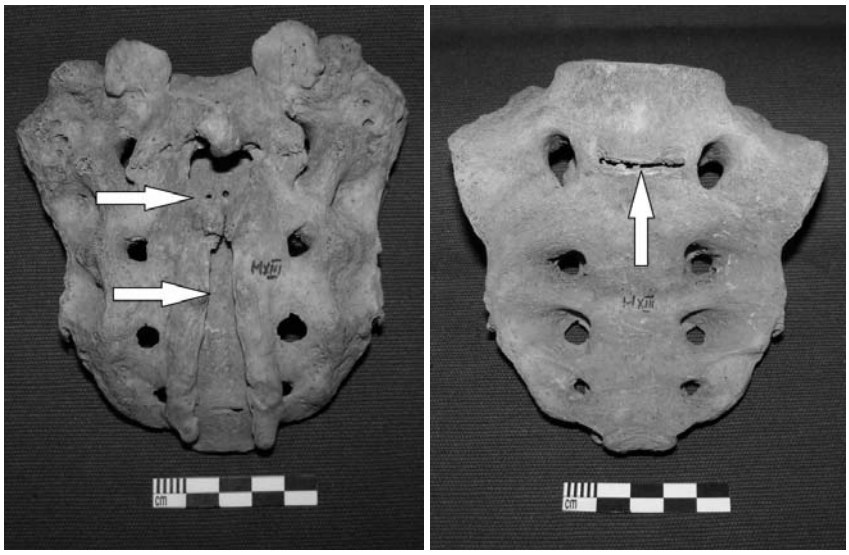


Fig. 4. Subject MXIV – ♂, 18–19 years: posterior view of sacrum – sacral *spina bifida occulta* in the S1 and S3-S5 segments.



a b

Fig. 5. Subject MXIII – ♂, 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 lumbarization of the first sacral vertebra (S1).

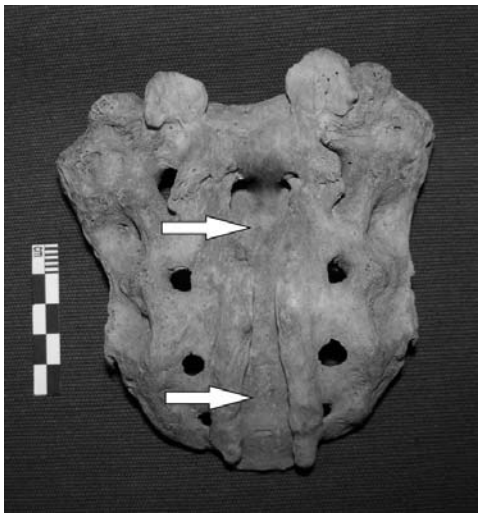


Fig. 6. Subject M126A – ♂, 40–45 years: posterior view of sacrum:
sacral spina bifida occulta in the S2-S5. segment.

4. CONCLUSIONS

This article describes in bioarchaeological terms the frequency and skeletal manifestation of sacral *spina bifida occulta* in medieval and post-medieval populations (14^{h}–19^{th} centuries) from Eastern Romania. No cases of cervical, thoracic or lumbar interlaminar dehiscences were discovered.

Sacral *spina bifida occulta* has a relatively low overall prevalence, of 4%, however some variability has been observed in frequency, according to the type of assemblages: higher in the elite sample (6.59%) and even in the rural necropolis (7.1%) than in urban collection (2.35%). The elevated prevalence of sacral *spina bifida occulta* in the mentioned groups may indicate the influence of the genetic factor. In the case of the Princely Court (17^{th} century), we suspect familial relationships to be involved.

Analysis of sex and age patterning indicates a higher prevalence of sacral *spina bifida occulta* in men than in women and a low frequency in old subjects, however the statistical evaluation of associations between the presence/absence of this abnormality and sex or age proved not significance.

Considering the morphology of the lumbo-sacral region, a variety of skeletal manifestation of sacral *spina bifida occulta* may be described: type V – five specimens; types I and IV – two specimens each; types II and III – one specimen each. Three subjects show additionally transitional abnormalities at the lumbo-sacral junctions besides *spina bifida occulta*.

Acknowledgements. We thank Dr. Stela Cheptea (senior archaeologist at the Centre for European History and Civilization of Iaşi, Romania) and Daniel Garvăn (archaeologist at the Museum of History and Archaeology, Piatra Neamţ, Romania) for the osteological materials made available for the present anthropological study.

Authors' contributions: Vasilica-Monica Groza (principal author) – 40%; Angela Simalcsik (second author) – 20%, Luminița Bejenaru (principal and corresponding author) – 40%

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Received February 12, 2016