STUDY OF SPINA BIFIDA OCCULTA BASED ON AGE, SEX AND LOCALIZATION

Sipos Tamas-Csaba¹, Denes Lorand¹, Brinzaniuc Klara¹, Sipos Remus Sebastian¹, Raduly Gergo¹, Pap Zsuzsanna¹

¹ University of Medicine, Pharmacy, Sciences and Technology of Tîrgu-Mureş

Ráduly Gergő

Department of Anatomy and Embryology, University of Medicine, Pharmacy, Science and Technology of TîrguMureş,
38 Gheorghe Marinescu Street, 540139 Tîrgu Mureş, Romania
email: raduly_gergo@yahoo.com
phone: +40 747714387

ABSTRACT

Introduction: Spina bifida is a spine malformation that appears as an incomplete closure of the spine during development. Most frequently it involves lumbosacral vertebrae. There are two types of spina bifida: spina bifida aperta and spina bifida occulta. In most of the cases spina bifida occulta is asymptomatic, and it is identified by accident.

Objectives: the aim of this study was to correlate localization of spina bifida occulta with age and sex of the patients.

Materials and methods: Between July 2017 and January 2019 there have been 108 patients diagnosed with spina bifida occulta at the Radiology Department of Dora Medicals Tîrgu Mureş. The diagnoses were based on thoracolumbar spine x-ray, pelvic x-ray and chest x-ray studies requested by specialist physicians. Radiologic diagnosis was followed by data processing and statistical analysis.

Results: In 81% of the cases the lesion was localized to vertebra S1, and in 15% to vertebra L5; we identified other five cases of rare localizations: C7- T1- T2, T1, T2, S2. This lesion was diagnosed most frequently at ages from 11 to 15 years (44.4%). Spina bifida localized to L5 was more frequent in males (11/16, 68.7%). Cases localized to S1 were more frequent in females (60/88, 68.1%). All spina bifida cases in females localized to vertebra L5 were diagnosed before 20 years of age.

Conclusions: Our results partially correspond to those reported in other published studies.

Keywords: spina bifida, malformation, diagnosis, localization

INTRODUCTION

Spina bifida is one of the most complex congenital malformations of the spine, which is characterized by incomplete closure of the spine (bifida). Fusion of the vertebral arches is incomplete due to deficient development of the neural tube during the embryonic period, in the first four weeks following fecundation. Most frequent localization is lumbar or sacral (1,2).

Etiology of spina bifida is complex; there are genetic factors, environmental factors or medicines / drugs used by the mother during pregnancy or a combination thereof (3). According to several studies, the incidence

of neural tube defects decreases drastically as a result of the beneficial effects of folic acid administered during pregnancy (4,5).

There are two major categories of spina bifida: spina bifida aperta (open) and occulta (closed) (6). These two are further subdivided based on the presence of a hernia sac, its contents and whether there is skin coverage or not.

Spina bifida aperta or spina bifida cystica has three clinical forms: meningocele, myelomeningocele and myeloschisis. These subtypes are characterized by a hernia sac that is not covered by skin. Myeloschisis is in fact the defective closure of the neural tube, so the spinal cord will be split. It is the most severe form of spina

Table 1. Clinical data of patients with spina bifida occulta according to localization

	S1		L5		C7-T2	T1	T2	D2	TOTAL
Ages (years)	Female	Male	Female	Male	Male	Female	Male	Male	
1-10	6	2	1	1	0	0	0	0	10
11-15	29	11	1	7	0	0	0	0	48
16-20	6	4	3	2	0	0	0	1	16
21-50	10	8	0	0	1	1	1	0	21
51-80	9	3	0	1	0	0	0	0	13
Total	60	28	5	11	1	1	1	1	108

bifida (6). Meningocele is the rarest subtype, and it is characterized by herniation of the meninges, without involvement of the spinal cord. It may also be a variant of spina bifida occulta, if it is covered by skin. Myelomeningocele is a subtype of spina bifida aperta, where due to incomplete closure of the neural tube during embryonic development a spinal cord segment protrudes into the hernia sac from the spinal canal (7).

The other category is spina bifida occulta or closed spina bifida, where the hernia lesion is covered by the skin. There are several subtypes, like lipomyelomeningocele and lipomeningocele. In case of lipomyelomeningocele the hernia sac has a meningeal and neural component, as well as adipose tissue. If the hernia sac contains only meninges and adipose tissue, it is a lipomeningocele, a less severe variant of the disease (7).

Another variant of spina bifida occulta is without herniation. This is the most frequent subtype, and since it is asymptomatic it is discovered usually by accident. This is considered spina bifida as is, and the other two subtypes (lipomyelomeningocele and lipomeningocele) are more severe variants of spina bifida occulta.

OBJECTIVES

This retrospective study presents the incidence of spina bifida occulta according to age, sex and localization, diagnosed using radiological studies.

MATERIALS AND METHODS

Between July 2017 and January 2019 there have been a total of 108 patients diagnosed

with spina bifida occulta at the Radiology Department of SC. Dora Medicals Tîrgu Mureş. The patients presented for AP radiographic studies of the cervical and thoracolumbar spine; AP radiographic studies of the pelvis; PA radiographic studies of the chest; they had spine pain, scoliosis.

Radiographic studies were performed using a Siemens Multix Select DX equipment. Following reassessment of the images in order to determine location of spina bifida, we centralized the clinical data (sex, age) of patients. Statistical analysis of the data was performed using Graph Pad In Stat 3 software, version 3.06 (GraphPad Software Inc, San Diego, USA). Significant association was considered in case of $p < 0.05$, with a 95% confidence interval.

RESULTS

During the 19 months considered for the study, a total of 109 patients presented at the Radiology Department for radiographic studies, which led to the diagnosis of spina bifida occulta. Of the 109 cases 60.6% were females and 39.4% were males (Table 1).

The most frequent localization was lumbosacral. In 81% of the cases the lesion was localized to the S1 vertebra, and in 15% of the cases localization was the L5 vertebra. In some cases we identified rare localizations of spina bifida, i.e. C7-T1-T2, T1, T2 and S2.

According to age group distribution the lesion was most frequently diagnosed by radiological studies in young patients aged 11 to 15 years (44.4%). Sex ratio was approximately similar in all age groups (females:males = 1 -

2.33) ($p=0.83$). The fewest cases were diagnosed before the age of 10 years, also considering the risk of irradiation at young ages (Table 1, Figure 1).

Figure 1. Sex of the patients according to age

Spina bifida localized to L5 was more frequent in males (11/16, 68.7%) in all age groups, except the 16-20 years age group, where the ratio between sexes was 1.5 with female predominance. Most cases were diagnosed between 11-15 years (8/16, 50%) ($p=0.25$) (Figure 2).

Figure 2. Spina bifida occulta localized to L5 according to age and sex

Spina bifida localized to S1 was more frequent in females (60/88, 68.1%), with most of the cases diagnosed also between 11-15 years (40/88, 45.4%) (Table 1, Figure 3). There was no statistically significant correlation for this localization between the age and sex of the patients ($p=0.66$). Sex ratio was variable between 1.25 and 3 (females:males = 3 between 1-10 years and over 51 years; females:males = 1.25 between 21 and 50 years) (Figure 3).

Figure 3. Spina bifida occulta localized to S1 according to age and sex

Comparing sex ratios according to localization it is notable that the ratio of cases localized to S1 and diagnosed in women has a decreasing tendency between 1 and 20 years of age, followed by an increasing tendency between 21 and 80 years.

All cases localized to L5 and identified in women were diagnosed before 20 years of age, with a higher incidence between 16 and 20 years. The ratio of cases localized to S1 in males was higher between 21 and 50 years of age, contrasting with cases localized to L5 (Figure 4).

Figure 4. Spina bifida occulta according to age, sex and localization

Our cases included a number of rare localizations: C7- T1- T2, T1 (Figure 5), T2 (Figure 6) and S2 (Figure 7).

Figure 5. Spina bifida occulta localized to T1

Figure 6. Spina bifida occulta localized to T2

Figure 7. Spina bifida occulta localized to S2

DISCUSSION

In most of the cases spina bifida occulta is asymptomatic and it is identified by accident or even post-mortem, without causing major problems in day to day life of the population. Certain specialties, like neurology, urology, radiology, epidemiology or even neurosurgery, are especially concerned with some aspects of this disease (age, sex, localization, symptoms) for diagnosis, therapy or possible complications.

In most of the cases spina bifida occulta involves the lumbosacral spine; this finding is supported by our study, considering that most of the lesions were localized to L5 and S1 (8). Our study enrolled 108 patients, 38.9% (42) males and 61.6% (66) females aged from 3 years to 65 years. Nicolici D. et al. studied 95 patients with spina bifida occulta (65.1%) aged from 3 years to 18 years, with a similar sex ratio, 44.2 % (42) males and 55.8% (53) females (9). Eubanks J.D. et al. studied the incidence of spina bifida occulta on 2866 human skeletons, and observed this lesion in 12.4% (355) of the cases on the lumbosacral spine. In contrast to our observations, of the 355 skeletons 42 were females and 313 males (10). Between the years 1967 and 1977 A. Fidas et al. studied 2707 radiographs to identify spina bifida occulta with different localizations. They reported a total of 643 cases of spina bifida, i.e. 23% of the randomly examined population, with 414 males and 229 females (11).

Table 2. Frequency of spina bifida occulta according to sex

Article	Female	Male	Total	Female:male
Our study	66	42	108	1.57
Nicolici D. et al. (2012)(9)	53	42	95	1.26
Eubanks J.D. et al. (2009)(10)	42	313	355	0.13
A. Fidas et al. (1987)(11)	229	414	643	0.55
Albrecht T.L. et al. (2007)(12)	19	34	53	0.55

According to published data, sex ratio varies between 0.13 and 1.57 with female predominance (Table 2). In our study spina bifida occulta was localized most frequently to vertebra

S1 (88/106). Of the 88 cases 60 were females (68.18%). In the A. Fidas et al. study, spina bifida localized to S1 was more frequent in males (Table 2). Shabana et al. demonstrated a prevalence of 5.5% for grade 1 spina bifida occulta (S1-S2), this area being the most affected, against 4.5% for grade IV (open sacral canal) (3).

According to our results, in contrast with S1 localization, spina bifida localized to vertebra L5 was more frequent in males (68.7%). According to certain studies, spina bifida localized to L5 is more rare compared to that with sacral localization and it is more frequent in males. The Fidas study lists spina bifida localized to S2, a total of 23 cases, 9 females and 14 males (11).

In rare cases thoracic spina bifida is described. Manenti G. et al. described spina bifida occulta in a 26 years old male that was localized to vertebra T1 (8). Our cases included a number of rare localizations: C7- T1- T2; T1; T2 and S2.

CONCLUSIONS

There are very few published studies that analyze clinico-pathological data of spina bifida patients. In our patient pool, spina bifida occulta was more frequently localized to vertebra S1, occurred especially in females, and it was diagnosed by radiology studies especially at young ages from 11 to 15 years.

REFERENCES

1. Copp AJ, Adzick NS, Chitty LS, Fletcher JM, Holmbeck GN, Shaw GM. Spina bifida. *Nat Rev Dis Primers*. 2015 Apr 30;1:15007.
2. Fletcher JM, Brei TJ. Introduction: Spina bifida--a multidisciplinary perspective. *Dev Disabil Res Rev*. 2010;16(1):1-5.
3. Ali S, Azeemi AA, Shoukat S. The prevalence of spina bifida occulta in Pakistani population: A study of dry human sacra. *ANAESTH, PAIN & INTENSIVE CARE*. 2014;18(2):157-61.
4. Au KS, Ashley-Koch A, Northrup H. Epidemiologic and genetic aspects of spina bifida and other neural tube defects. *Dev Disabil Res Rev*. 2010;16(1):6-15.
5. Atta CA, Fiest KM, Frolkis AD, Jette N, Pringsheim T, St Germaine-Smith C, et al. Global Birth Prevalence of Spina Bifida by Folic Acid Fortification Status: A Systematic Review and Meta-Analysis. *Am J Public Health*. 2016 Jan;106(1):e24-34.
6. Enciulescu C. *Embriologie specială*, Vol. II: University Press Tg. Mureș; 2011. p 135-136.
7. Mohd-Zin SW, Marwan AI, Abou Chaar MK, Ahmad-Annuar A, Abdul-Aziz NM. Spina Bifida: Pathogenesis, Mechanisms, and Genes in Mice and Humans. *Scientifica (Cairo)*. 2017;2017:5364827.
8. Manenti G, Iundusi R, Picchi E, Marsico S, D'Onofrio A, Rossi G, et al. Anatomical variation: T1 spina bifida occulta. Radiological findings. *Radiol Case Rep*. 2017 Mar;12(1):207-9.
9. Nikolic D, Petronic I, Cvjeticanin S, Brdar R, Cirovic D, Bizic M, et al. Gender and morphogenetic variability of patients with spina bifida occulta and spina bifida aperta: prospective population-genetic study. *Hippokratia*. 2012 Jan;16(1):35-9.
10. Eubanks JD, Cheruvu VK. Prevalence of sacral spina bifida occulta and its relationship to age, sex, race, and the sacral table angle: an anatomic, osteologic study of three thousand one hundred specimens. *Spine (Phila Pa 1976)*. 2009 Jul 1;34(15):1539-43.
11. Fidas A, MacDonald HL, Elton RA, Wild SR, Chisholm GD, Scott R. Prevalence and patterns of spina bifida occulta in 2707 normal adults. *Clin Radiol*. 1987 Sep;38(5):537-42.
12. Albrecht TL, Scutter SD, Henneberg M. Radiographic method to assess the prevalence of sacral spina bifida occulta. *Clin Anat*. 2007 Mar;20(2):170-4.