

Is There Any Association between Spina bifida occulta and Primary Vesicoureteral Reflux?

Mehrzad Mehdizadeh^{1,2}, MD; Azadeh Roohi², MD; Mohsen Hemami³, MD,
and Seyed-Taher Esfahani^{*2,4}, MD

1. Department of Radiology; Tehran University of Medical Sciences, Tehran, IR Iran
2. Children's Medical Center, Pediatrics Center of Excellence, Tehran, IR Iran
3. Department of Epidemiology and Biostatistics, Tehran University of Medical Sciences, Tehran, IR Iran
4. Department of Pediatrics, Tehran University Of Medical Sciences, Tehran, IR Iran

Received: Jul 26, 2009; Final Revision: Dec 08, 2009; Accepted: May 10, 2010

Abstract

Objective: Spina bifida occulta (SBO) has been largely considered a benign entity without clinical significance; however, there has been dispute among various authorities, and some believe that the lesion may be linked with various neurologic conditions like urologic dysfunctions. Vesicoureteral reflux (VUR) and lower urinary tract dysfunction are closely related. We examined whether the existence of SBO is related to the prevalence and severity of VUR in children.

Methods: We investigated 359 children, 2-14 years old, referred to radiology department for obtaining voiding cystourethrogram after the first attack of febrile urinary tract infection. After treatment of infection, with written order of responsible physicians all underwent a standard voiding cystourethrogram to detect VUR and other lower urinary tract anomalies. The patients were divided into two groups: group 1 patients who had not SBO and group 2 patients with SBO in postvoiding or KUB films. In each group the presence and severity of VUR was determined in relation to the location of SBO.

Findings: Out of 359 children, 228 (63.5%) had normal spine and 131 (36.5%) had SBO. Fifty four (23.7%) out of 228 children with normal spine had VUR and 40 (30.5%) out of 131 children with SBO had VUR. The prevalence of VUR in children without SBO and children with SBO was not statistically different. Also we compared the severity of VUR between the two groups and there was no significant difference or trend between presence of SBO and severity of reflux (Chi² for trend). VUR was more common in children with SBO in L5-S1 (38.3%). There was no significant relation between location of SBO and prevalence of VUR.

Conclusion: Location of SBO and prevalence of VUR are not related.

Iranian Journal of Pediatrics, Volume 20 (Number 3), September 2010, Pages: 348-352

Key Words: Spina Bifida Occulta; Vesico-ureteral reflux; Voiding dysfunction; Spinal anomalies

* Corresponding Author;

Address: Division of Nephrology, Children's Medical center, Pediatrics Center of Excellence, Dr Gharib St, Tehran, IR Iran

E-mail: esfahani@tums.ac.ir

© 2010 by Pediatrics Center of Excellence, Children's Medical Center, Tehran University of Medical Sciences, All rights reserved.

Introduction

Spina bifida is a disorder in which two sides of vertebral arches, usually in lower vertebrae, fail to fuse during development resulting in an open vertebral canal. There are two types of spina bifida:

- 1) The commonest type is spina bifida occulta (SBO), in which there is a defect in the vertebral arch of L₅ or S₁, and results in failure of posterior arch to fuse in the midline.
- 2) The more severe form of spina bifida vera cystica involves complete failure of fusion of posterior arch at the lumbosacral region with a large outpouching of the meninges. This may contain cerebrospinal fluid (meningocele) or a portion of spinal cord (myelomeningocele).

These abnormalities may result in a variety of neurologic defects including impaired walking and bladder function^[1,2].

The reported frequency of occurrence of SBO varies widely, depending largely on the age group included in a particular study^[3,4]. Although it is true that this lesion is present at birth, some cases seem to vanish with increasing bone maturity. It should be kept in mind that in normal conditions, the two halves of the posterior arch fuse between the ages of 5 and 8 years. Studies with children have indicated that an apparent SBO defect that persists past the age of 8 years may fill with new bone as a part of the aging process, ossifying, in fact a radiologically undetectable connective tissue matrix in the posterior arch. To obtain a true sense of the prevalence of this lesion, one must examine its frequency of occurrence in relation to chronologic age. In <18 year age groups, the incidence may be as high as 50%. The true prevalence of this anomaly, excluding cases that later disappear, has been estimated at approximately 17% of examined spines^[3,5].

Spina bifida vera is associated with variety of neurologic dysfunctions in lower urinary tract, but SBO often is asymptomatic and is considered of no clinical significance^[6,7]. However, it has been implicated in the etiology of urinary voiding problems^[8-10]. Vesicoureteral reflux

(VUR) and lower urinary tract dysfunction are closely related^[11-14]. Bladder dysfunction may have an etiologic role in development of VUR or may aggravate it^[11,12]. Treatment of bladder dysfunction increases spontaneous resolution of VUR^[13]. Primary VUR generally improves with time^[14]. So both conditions are more common in infancy and may resolve with increasing age and both may be related to the voiding dysfunction. Herein we studied if there is a relation between SBO and primary VUR in children.

Subjects and Methods

We investigated 359 children after the first attack of febrile urinary tract infection that had been referred to radiology department for obtaining voiding cystourethrography (VCUG). After treatment of infection, with written order of their physicians and permission of parents all underwent a standard VCUG to detect VUR and other lower urinary tract anomalies.

Reflux grading was done according to the international classification of the reflux^[15]. In each patient presence or absence of SBO, and if SBO was present, its location was determined with KUB or post-voiding films. SBO was diagnosed by hypoplasia or aplasia of the spinous process, producing a radiolucent defect on anteroposterior KUB or postvoiding films. Children with neurologic bladder or with other spinal anomalies were excluded from the study.

SPSS software version 16.0.2 (SPSS Inc., Chicago, IL), was used for data analysis. To compare frequency of variables between groups Chi-square and for detecting trend, a trend Chi square were used. *P* value lower than 0.05 was considered significant.

This study was approved by the ethics committee of the Tehran University of Medical Sciences. All radiographs were taken with written order of responsible physicians. The nature and purpose of performing VCUG was explained and verbal consent taken from parents before radiography.

Table 1: Prevalence of different grades of vesicoureteral reflux in patients with and without spina bifida occulta Figures in the parentheses are percentages

| Reflux grade | Right kidney | | Left kidney | |
|--------------|---------------------|------------------|---------------------|------------------|
| | without SB n=228 | with SB n=131 | without SB n=228 | with SB n=131 |
| No reflux | 201(88.1) | 104(79.3) | 185(81.1) | 110(48.2) |
| Grade I | 6(2.6) | 8(6.1) | 3(1.3) | 2(0.9) |
| Grade II | 8(3.5) | 7(5.3) | 25(11.0) | 11(4.8) |
| Grade III | 9(3.9) | 7(5.3) | 8(3.5) | 3(1.3) |
| Grade IV | 1(0.4) | 3(2.3) | 2(0.9) | 1(0.4) |
| Grade V | 3(1.3) | 2(1.5) | 5(2.2) | 4(1.8) |

Findings

Three-hundred fifty nine children (128 boys and 231 girls), 2-14 years old (mean age 75 ± 37 months), were evaluated. Of 359 children 228 (63.5%) had normal spines and 131 (36.5 %) had SBO. The location of SBO was: L5 6 (0.045%) cases, L5-S1, 47 cases (35.9%), S1 78 (59.5 %) cases. Of 228 children with normal spines 174 (76.3%) had no VUR and 54 (23.7%) had VUR. Of 131 children with SBO, 91 (69.5%) had no VUR and 40 (30.5%) had VUR.

The prevalence of VUR between children without SBO and children with SBO was not significant ($P=0.15$). Among children with SBO in L5, 16.7%, in L5-S1 38.3% and in S1 26.6 had VUR. There was no significant relation between location of SBO and prevalence of VUR ($P=0.4$).

We also evaluated if there was a relation between presence of SBO and severity of VUR. The VUR grades in both kidneys are shown in Table 1. There was no significant difference or trend between presence of SBO and severity of reflux (Chi^2 for trend).

Discussion

SBO is a common anomaly^[3-5]. Fidas et al reviewed plain radiographs of the urinary tract

on 2707 normal adult volunteers. This condition was twice as common in males as in females and was commoner in young adults^[3]. The commonest defects were either in S1 alone or involved S1 and S2 together. Isolated defects of L5 or S2 were rare. The incidence of SBO decreases with age, possibly as a result of new bone formation or calcification^[4,5]. According to Sutow and Pryde the incidence of SBO diminishes in male from 22% in the seventh year to 4% in adults, and in females from 9% in the seventh year to 1% in adults^[4].

The spina bifida vera cystica is well known and has undisputed neurologic sequelae, but most individuals with SBO are asymptomatic and lack neurologic signs and the condition is considered of no clinical significance^[6,7]. However, SBO is linked with a variety of functional disorders of lower urinary tract in children^[8-10].

Sakakibara et al performed uro-neurologic assessment of patients with spina bifida cystica and occulta. Their study showed that SBO presents with a wide spectrum of urodynamic abnormalities including upper and lower neuron types of bladder and urethral dysfunction. In some of their SBO patients urinary symptoms like nocturnal enuresis were the sole initial complaints and had no other neurological abnormalities^[8]. In contrary, Samuel and Boddy evaluated 158 children presenting with lower urinary tract and bowel dysfunction for

associated spinal abnormalities. They concluded that SBO in these children is probably a coincidental finding and its significance in this cohort is not established^[16]. Also, Nejat and co-workers showed that there is no significant association between SBO and functional bowel and urinary problems^[17]. So, there are conflicting reports regarding the relation between SBO and functional disorders of urinary tract. Probable role of SBO as a finding associated with spinal cord dysfunction remains unclear.

Miyazato et al reported that the location of SBO predicts the outcome of treatment for primary nocturnal enuresis in children, in their study patients with L5 and L5-L51 spina bifida were less responsive to treatment compared to patients with sacral SBO^[9]. In contrary, Kumar et al studied incidence of SBO in nocturnal enuresis cases and compared outcome of enuresis in children with or without SBO. They found that outcome of patients with SBO is not different from those without SBO^[18]. Ritchey et al reviewed retrospectively 456 patients with diurnal enuresis to determine the relationship between spina bifida occulta and attainment of continence. They compared urodynamic findings in children with or without SBO. Findings on urodynamic studies were similar in both groups. Also, the outcome for enuretic children with SBO was comparable to those with normal spines^[19].

VUR and lower urinary tract dysfunction are closely related. There are reports showing that lower urinary tract dysfunction is a factor in pathogenesis of VUR^[11,13] and treatment of voiding dysfunction prompts resolution of VUR^[13]. Because both SBO and VUR may be related to voiding disorders, we proposed that there may be a relation between these two disorders, but our study showed that SBO or its location has no relation with prevalence or severity of VUR.

The SBO is subject of dispute among various authorities, with one faction adamantly refusing to accept the premise that the lesion may be linked with various neurologic conditions and the opposing side insisting that a plethora of abnormalities may be associated with the condition^[5].

Conclusion

In our study, the prevalence of VUR between children without SBO and children with (isolated) SBO was not significant (23.7% vs. 30.5%). In addition, there is not any association between presence and location of (isolated) SBO and severity of primary VUR.

Acknowledgment

This research was approved by Pediatrics Department Research Committee in Tehran University of Medical Sciences.

Conflict of Interest: None

References

1. Drake RL, Vogle W, Mitchell AWM. Anatomy for student, 1st ed. Philadelphia; Churchill Livingstone. 2005; Pp:35b.
2. Gleeson JG, Dobyns WB, Plawner L, Ashwal S. Congenital structural defects. In: Swaiman KF, Ashwal S, Ferriero DM (eds): Pediatric Neurology, Principles and Practice, 4th ed. Philadelphia; Mosby-Elsevier. 2006; Pp: 463-90.
3. Fidas A, MacDonald HL, Elton RA, et al. Prevalence and patterns of spina bifida occulta in 2707 normal adults. Clin Radiol. 1987; 38(5):537-42.
4. Sutow WW, Pryde AW. Incidence of spina bifida occulta in relation to age. Am J Dis Child 1956; 91:211-7.
5. Gregerson DM. Clinical consequences of spina bifida occulta, J Manipulative Physiol Ther 1997;20(8):546-50.
6. Silverman FN, Slovis TL. Congenital malformations, in Kuhn JP, Slovis TL, Haller JO (eds): Coffey's Pediatric Diagnostic Imaging, 10th ed. Mosby, Philadelphia, 2004; Pp:654-67.
7. Kinsman SL, Johnston MV. Congenital anomalies of the central nervous system. In: Kliegman RM, Behrman RE, Jenson HB, Stanton BF (eds): Nelson Textbook of Pediatrics, 18th ed. Philadelphia; Saunders-Elsevier. 2007; Pp: 2443-56.
8. Sakakibara R, Hattori T, Uchiyama T, et al. Uroneurological assessment of spina bifida

- cystica and occulta. *Neurol Urodyn.* 2003; 22(4):228-34.
9. Miyazato M, Sugaya K, Nishijima S, et al. Location of spina bifida occulta and ultrasonographic bladder abnormalities predict the outcome of treatment for primary nocturnal enuresis in children. *Int J Urol.* 2007;14(1):33-8.
 10. Fidas A, MacDonald HL, Elton RA, et al. Neurological defects of the voiding reflex arcs in chronic urinary retention and their relation to spina bifida occulta. *Br J Urol.* 1989;63(1): 16-20.
 11. Sillén U. Vesicoureteral reflux in infants. *Pediatr Nephrol.* 1999; 13(4):355-61.
 12. Yeung CK, Godley ML, Dhillon HK, et al. Urodynamic patterns in infants with normal lower urinary tracts or primary vesico-ureteric reflux. *Br J Urol.* 1998; 81(3):461-7.
 13. Sillén U. Bladder dysfunction in children with vesico-ureteric reflux. *Acta Paediatr Suppl.* 1999;88(431):40-7.
 14. Mattoo TK, Mathews R. Vesicoureteral reflux and renal scarring, in Avner ED, Harmon WE, Niaudet P, Yoshikawa N (eds): *Pediatric Nephrology*, 6th ed. Springer, Berlin Heidelberg. 2009; Pp:1311-36.
 15. International Reflux Study Committee. Medical versus surgical treatment of primary vesicoureteral reflux. *Pediatrics.* 1981;67(3): 392-400.
 16. Samuel M, Boddy SA. Is spina bifida occulta associated with lower urinary tract dysfunction in children? *J Urol.* 2004;171(6Pt 2):2664-6.
 17. Nejat F, Radmanesh F, Ansari S, et al. Spina bifida occulta: is it a predictor of underlying spinal cord abnormality in patients with lower urinary tract dysfunction? *J Neurosurg Pediatr.* 2008; 1(2):114-7.
 18. Kumar P, Aneja S, Kumar R, et al. Spina bifida occulta in functional enuresis. *Indian J Pediatr.* 2005;72(3):223-5.
 19. Ritchey ML, Sinha A, DiPietro MA, et al. Significance of spina bifida occulta in children with diurnal enuresis. *J Urol.* 1994;152(2Pt 2): 815-8.