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Occult spinal dysraphism and its association with hip dysplasia in females

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Abstract We examined the pelvic radiographs of two groups of patients (more than 12 years of age) from six medical centres. Hip dysplasia was considered to be present if Shenton's line was broken and more than one third of the femoral head was revealed to be uncovered in an antero-posterior radiograph of the pelvis. Patients with hip dysplasia due to teratological or neurological causes were excluded. There were 291 patients with treated or untreated hip dysplasia in the dysplastic group. The control group of 415 individuals was collected from consecutive outpatients (with a pre-set standardised female/male ratio) for whom an antero-posterior radiograph of the pelvis had been made in one of two medical centres and which did not disclose any abnormality of the hip joints. The aim of the study was to assess the co-existence of hip dysplasia and occult spinal dysraphism.

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Radiographs of all patients were examined, and any partial or complete defect of the posterior vertebral arch was recorded. In the dysplastic group, a defect was recorded in 23% (67/291) radiographs and in the control group in 12% (48/415). In both groups, L5 and S1 were the most commonly recorded sites with a defect. In the dysplastic group, a defect was recorded in 56/190 females and in the control group in 30/302 females. In males, there was no significant difference between the recorded findings in the two groups. In females with hip dysplasia, occult spinal dysraphism seems to be fairly common.

Résumé Nous avons examiné les radiographies pelviennes de deux groupes des malades âgés de plus de 12 ans de six centres médicaux. Nous avons défini la dysplasie de la hanche par la rupture de la ligne de Shenton et le défaut de couverture de plus d'un tiers de la tête fémorale sur une radiographie du bassin de face. La dysplasie de la hanche dû aux causes tératologique ou neurologiques a été exclue. Nous avons inclus 291 malades avec dysplasie de la hanche traitée ou non traitée dans le groupe dysplasique. Le groupe témoin de 415 individus a été rassemblé parmi les malades vu consécutivement en consultation (avec une proportion homme/femme standardisée) qui avait une radiographie du bassin sans anomalie des hanc. Le but de l'étude était d'apprécier la coexistence de dysplasie de la hanche avec un dysraphisme vertébral occulte. Les radiographies pelviennes de tous les malades ont été examinées et tout défaut partiel ou complet de l'arc vertébral postérieur a été enregistré. Dans le groupe dysplasique un défaut a été enregistré dans 23% (67/291) des radiographies, et dans le groupe témoin dans 12% (48/415). Dans les deux groupes L5 et S1 étaient les deux étages le plus fréquemment concerné par le défaut. Dans le groupe dysplasique un défaut a été enregistré dans 56/190 femmes et dans le groupe témoin dans 30/302 femmes. Chez les hommes il n'y avait aucune différence notable entre les conclusions enregistrées dans les deux groupes. Chez les femmes avec dysplasie de la hanche, le dysraphisme vertébral a une prévalence certaine.

Introduction

Spina bifida occulta (SBO) is a defect of the vertebral arch due to failure of fusion. It is one of the most common congenital anomalies but is seldom of clinical significance. It is considered a normal variation in the majority of individuals and is often noted as an incidental finding on spine radiographs. It is rarely accompanied by intraspinal anomalies. The incidence of SBO is estimated to be as high as 20% in the general population. The fifth lumbar and first sacral vertebrae are the most commonly involved sites [2, 3, 4, 5].

Developmental dysplasia of the hip (DDH) is accompanied by several well-known risk factors and other musculoskeletal deformities [3, 5, 6]. In a previous study, it was reported that half of the patients who had DDH and who then failed to respond to primary surgical treatment also had an occult spina bifida [7].

Patients and methods

This was a multi-centre study and included the assessment of an antero-posterior pelvic radiograph of 706 individuals older than 11 years. Fusion of the vertebral arches usually occurs during the first 3–5 years of life but is sometimes delayed until adolescence. This could lead to a higher incidence of SBO in the spine radiographs of infants and young children than in adolescents and adults [4, 5].

A retrospective study of the medical records of treated and of untreated patients with DDH was made at six different medical centres. Shenton's line was broken in all the initial radiographs, and more than one third of the femoral head was not covered by the bony acetabulum. Patients who had undergone a complete clinical and radiological examination were included in the study, with the exception of those with teratologic or neuromuscular hip dislocations. Two hundred and ninety one patients (246 females and 45 males) met these criteria and formed the DDH group, and their radiographs were assessed. The control group was collected from patients who were seen in the orthopaedic outpatient clinics of two different medical centres and for whom an antero-posterior radiograph of the pelvis was available. The female/male ratio in the control group was selected as 4:1, which is the same as the female/male ratio in developmental dysplasia of the hip [3, 5, 6]. The first 332 consecutive females and 83 males who had no bilateral clinical or radiological hip joint abnormalities formed the control group. The first two authors assessed the radiographs of the control group, and any partial or complete defect of the vertebral arch was recorded (Fig. 1).

Statistical analysis of the data was made using a PC program (SPSS 9.0 for Windows, SPSS Inc., Chicago, IL, USA). The chi-square test was used for comparison of the ratios between the two groups, and the *t*-test for independent samples for comparison of the means between the two groups. A *p* value of less than 0.05 was considered significant.

Results

The mean age was 30 ± 17 years in the DDH group and 38 ± 17 years in the control group ($p < 0.001$). Of the patients in the DDH group, 67/291 (23%) had a defect of the vertebral arch. In the control group, a defect was recorded in 48/415 patients (12%) ($p < 0.001$). In both groups, the most commonly involved vertebrae were L5 and S1 (Table 1).

Fig. 1 Antero-posterior pelvic radiograph of a 29-year-old female patient operated on at 14 months of age for bilateral developmental dysplasia of the hip (DDH). The radiographic result was considered satisfactory on both sides. An evident spina bifida occulta (SBO) can be seen at S1 (arrow)

Table 1 Localisation of spina bifida occulta (SBO) in the control and developmental dysplasia of the hip (DDH) groups

Involved vertebra	Control group	DDH group	Total
L5 alone	6	10	16
S1 alone	38	42	80
S2 alone	0	2	2
S3 alone	1	0	1
L4 and L5	1	5	6
L5 and S1	2	5	7
S1 and S2	0	2	2
L4 and L5 and S1	0	1	1
Total	48	67	115

In females, 56/246 patients (23%) in the DDH group had an SBO, while in the control group there were only 30/302 (9%) ($p < 0.001$). In males, the rate of SBO was similar in the two groups (24% vs 22%).

Discussion

The number of examined females in both groups is sufficient to allow statistical conclusions to be made from the data. However, the total number of examined males could have been higher, which might be considered a shortcoming of the present study. The infrequent occurrence of DDH in males is the main reason for this. Although the mean ages of the two groups differed, this difference was considered insignificant. Obtaining similar ratios between females and males in the two groups was considered more important, as this allowed reliable statistical conclusions to be made.

There are three primary ossification centres in each vertebra (one in the centrum and one in each half of the vertebral arch), and ossification becomes evident in the

vertebral arches by the end of the eighth gestational week. The arches first unite in the lumbar region, and union then progresses cranially [4]. The hip joint begins to develop by the fourth week of gestation and is completely differentiated by the end of the eighth week [1, 4], when the fetus has an almost completely differentiated musculoskeletal system. A number of possible genetic, developmental, environmental and mechanical factors may lead to DDH by affecting the development of the joint during gestation [1]. It is possible that the significant coexistence of DDH and SBO observed in this present study may be a possible partial explanation for the appearance of DDH. In addition, development of the hip joint and fusion of the vertebral arches may be impaired at the same time during the gestational period by genetic, mechanical or other factors. However, results of the present study reveal that this possible coincidental impairment occurs only in females. It was also noted that the incidence of SBO was nearly twice as high in the DDH group than in the control group. After careful analysis, it was evident that this significant difference was mainly related to the number of female patients with DDH. Another interesting finding is that the female patients with DDH had nearly two and a half times the incidence of SBO when compared to females without any hip joint abnormality. This difference was not present in males. Thus it can be said that SBO is more frequently seen in healthy males than in healthy females, but that the risk of a coexisting DDH and SBO seems to be significantly higher in females. Another theory may also explain the observed significant coexistence of DDH and SBO as, although there are no significant positive neurological findings in the majority of individuals with SBO, a minor neurological abnormality may lead to hip dysplasia in some individuals with SBO. Wilkinson and Sedgwick [7] have previously reported that more than half the patients with co-existing DDH and SBO had abnormal somato-sensory-evoked potentials. This associa-

tion seems to support our second theory. In the present study, however, clinical examination of the lower limbs failed to reveal any neurological abnormalities.

The findings of our study may be useful in clinical practice. For instance, the finding of a posterior vertebral arch defect during fetal ultrasonography may make it possible to warn the mother of a possible risk of an associated DDH, although her child may be born without any intraspinal anomaly at the lumbosacral area. Such infants could be considered as "at risk babies" after birth and may require ultrasonographic screening of their hips.

We can conclude from a study of the individuals in the present study that SBO may be a congenital musculoskeletal anomaly that can coexist with DDH in females. Thus, it may be considered as a risk factor for DDH in females. However, further detailed embryological, genetic and neurological studies are needed to clarify further this possible relationship.

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