Neurological classification in myelomeningocele as a spine deformity predictor

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In myelomeningocele, spinal deformities are responsible for major disability. Our aim was to check the predictive power for future spine deformity of a neurological classification applied at 5 years of age. We classified patients into four groups according to their neurological examination made at the age of 5 years. Groups were defined as follows: group I, L5 or below (meaning that all the patients in this group have a paralysis that at least leaves the L5 segment intact); group II, L3–L4; group III, L1–L2; group IV, T12 and above (meaning that all the patients in this group have a paralysis that reaches T11 or above). One hundred and sixty-three patients were included. The results showed that group I is a predictor for the absence of spinal deformity. Group III or IV is a predictor for spinal deformity. Group IV is a predictor of kyphosis. It was previously known that the higher the neurological level, the higher the rate of spinal deformity at maturity, but no work had given physicians a guideline to assess the spinal prognosis in myelomeningocele patients. Our work showed, on the basis of this classification made at the age of 5 years, that future spinal disorders may be expected in some patients, while no spinal deformity may be expected in some others. Thus, an appropriate therapeutic strategy and follow-up can be planned.
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Introduction

Spinal deformity and hip dislocation are common features in patients with myelomeningocele (MMC). Spinal deformities are mainly scoliosis, but also kyphosis and lumbar hyperlordosis. In the literature, the prevalence of scoliosis ranges from 60 to 90% [1,2]. The reported prevalence of kyphosis varies from 1% [3] to 46% [4] and the prevalence of lumbar hyperlordosis in patients with MMC was 1.5% in the survey conducted by Piggott [5]. These deformities develop after birth, during growth (developmental deformities) or are congenital (in the case of vertebral malformations such as hemivertebra) [1], but it is known that developmental factors can increase a congenital curve [5]. The functional prognosis in patients with MMC has been shown to be based on age and neurological level, but also on spinal deformity [6,7]. Spinal deformities (and especially scoliosis) and the neurological level are known to be linked, and it has been shown that the higher the neurological level, the higher the rate of spine deformity at maturity [5,8,9]. Nevertheless, the first two works among the three previously mentioned focused only on scoliosis, and the latter [9] only on kyphosis. None of them gives a comprehensive view of the global spinal deformity in both frontal and sagittal plans. Additionally, no work in the literature gives a clear guideline that would be usable for physicians to assess the prognosis for future spinal deformity in children with MMC.

Our aim was to check the predictive power for future spinal deformity (scoliosis, kyphosis and lumbar hyperlordosis) of a four-step neurological level classification based on the neurological examination made at the age of 5 years (from year five to year six), knowing, since the work of McDonald in 1986 [10], that the stability of muscle testing measurements in MMC over time is reached at the age of 5 years. In addition, the predictive power for hip dislocation occurrence of this classification was also checked.

Materials and methods

All of the charts and radiographs from the 210 patients with a diagnosis of MMC seen in our institution (Hôpital d'Enfants de la Timone, Marseille) or in the two paediatric rehabilitation centres of Hyères city were reviewed. The criteria for inclusion in this study included a diagnosis of MMC, an age at the first consultation below 5 years and an age at the last review of over 15 years, a serial documentation of the clinical neurological examination (motor power assessed with standard muscle-testing), a serial documentation of the spine geometry in both anteroposterior and lateral radiographs. Patients for whom a single radiograph or clinical evaluation was available (often children from other centres who had been referred to our institution for a second opinion)

were excluded. Patients with congenital deformity of the spine were also excluded. Eventually, 163 patients were included. There were 86 females (53%) and 77 males (47%).

The bone defect level was defined as the highest level at which the laminar arch is intact. The neurological level was assessed at 5 years and was defined as the lowest level at which the patient has a normal muscular strength on the worst side, using a manual muscle test according to Kendall [11] and based on the examination reported at 5 years. Muscles were tested against gravity and active resistance applied by the examiner. The result was recorded for each muscular group using a percentage: 0% was defined as no contraction felt; 10% was defined as active contraction without any motion; 50% was defined as a motion against gravity; 100% was defined as a motion against resistance.

Scoliosis was defined as a more than 20° curve, determined with the Cobb technique [12] on the later radiographic examination or on the preoperative radiographic examination in cases of surgery. Kyphosis was defined as a generalized curve exceeding 50° on the later radiographic examination or on the preoperative radiographic examination in cases of surgery. Lumbar hyperlordosis was defined as a curve over 60° on the later radiographic examination or on the preoperative radiographic examination in cases of surgery.

Hip dislocation was defined as complete loss of congruity between the femoral and acetabular surfaces. When an operation had been performed in order to keep the joint in place, the hip was considered as dislocated. Hip subluxations without any surgery were classified as normal.

The following data were collected: date of birth, sex, bone defect level (assessed on plain radiographs), neurological motor level (based on the neurological examination at 5 years), spine deformities (scoliosis, kyphosis or lumbar hyperlordosis) and surgery undergone, presence of retethering cord, presence of diastematomyelia, presence of syrinx and its clinical expression (progressive upper limb motor power loss and spasticity) and presence of hip dislocation. The presence of Arnold–Chiari malformations was not checked.

Based on the neurological examination at 5 years, we classified all the neurological deficiencies into four groups: group I, L5 or below (meaning that all patients in this group have a paralysis that at least leaves the L5 segment intact); group II, L3–L4; group III, L1–L2; group IV, T12 and above (meaning that all the patients in this group have a paralysis that reaches T11 or upper thoracic segments).

The current opinion in the literature is that the higher the neurological level, the higher the rate of spinal deformity at maturity [5,8,9]. But the limit above which spinal deformity may be expected is still unknown, and the limit below which no spinal deformity may be expected is also unknown. The determination of these limits was conducted as follows. A statistical analysis, in which the neurological classification made at 5 years was considered as a test, was performed in order to check three hypotheses.

(1) Is group IV a powerful predictor for the development of spinal deformity (scoliosis, kyphosis or lumbar hyperlordosis)?
(2) Is group III or IV a powerful predictor for the development of spinal deformity (scoliosis, kyphosis or lumbar hyperlordosis)?
(3) Is group II, III or IV a powerful predictor for the development of spinal deformity (scoliosis, kyphosis or lumbar hyperlordosis)?

Concerning kyphosis, the same method was applied and three additional hypotheses were checked.

(4) Is group IV a powerful predictor for the development of kyphosis?
(5) Is group III or IV a powerful predictor for the development of kyphosis?
(6) Is group II, III or IV a powerful predictor for the development of kyphosis?

Finally, two additional hypotheses were checked.

(7) Is group I a powerful predictor for the absence of spine deformity (scoliosis, kyphosis or lumbar hyperlordosis)?
(8) Is group II and III a powerful predictor for hip dislocation occurrence?

Hypothesis 1

A four-box grid comparing the presence or absence of group IV patients with the presence or absence of spinal deformity (scoliosis, kyphosis or lumbar hyperlordosis) was constructed as shown in Table 1. A Fisher test (because the theoretical population was small) was performed, comparing the presence or the absence of group IV patients with the presence or the absence of spinal deformity. In addition, sensitivity, specificity and positive predictive value were calculated. We considered a positive predictive value and specificity of more than 80% to indicate that this test is a powerful predictor [13].

Hypothesis 2

In the same way, a four-box grid comparing the presence or absence of group III or IV patients with the presence or absence of spinal deformity (scoliosis, kyphosis or lumbar hyperlordosis) was constructed as shown in Table 2. A Pearson χ^2 test was performed, comparing the presence or the absence of group III or IV patients with the presence or the absence of spinal deformity. In addition, sensitivity, specificity and positive predictive value were

Table 1 Hypothesis 1: is group IV a powerful predictor for spinal deformity (scoliosis, kyphosis or hyperlordosis)?

	Deformity (scoliosis, kyphosis or hyperlordosis)	No deformity
Group IV patients	9	0
Group I, II or III patients	59	95

Table 2 Hypothesis 2: is group III or IV a powerful predictor for spinal deformity (scoliosis, kyphosis or hyperlordosis)?

	Deformity (scoliosis, kyphosis or hyperlordosis)	No deformity
Group III or IV patients	28	6
Group I or II patients	40	89

Table 3 Hypothesis 3: is group II, III or IV a powerful predictor for spinal deformity (scoliosis, kyphosis or hyperlordosis)?

	Deformity (scoliosis, kyphosis or hyperlordosis)	No deformity
Group II, III or IV patients	62	32
Group I patients	6	63

Table 4 Hypothesis 4: is group IV a powerful predictor for kyphosis?

	Kyphosis	No kyphosis
Group IV patients	8	1
Group I, II or III patients	9	145

Table 5 Hypothesis 5: is group III or IV a powerful predictor for kyphosis?

	Kyphosis	No kyphosis
Group III or IV patients	13	21
Group I or II patients	4	125

Table 6 Hypothesis 6: is group II, III or IV a powerful predictor for kyphosis?

	Kyphosis	No kyphosis
Group II, III or IV patients	15	79
Group I patients	2	67

calculated. We considered a positive predictive value and specificity of more than 80% to indicate that this test is a powerful predictor [13].

Hypothesis 3

In the same way, a four-box grid comparing the presence or absence of group II, III or IV patients with the presence or absence of spinal deformity (scoliosis, kyphosis or lumbar hyperlordosis) was constructed as shown in Table 3. A Pearson χ^2 test was performed, comparing the presence or the absence of group II, III or IV patients with the presence or the absence of spinal deformity. In addition, sensitivity, specificity and positive predictive value were calculated. We considered a positive predictive value and specificity of more than 80% to indicate that this test is a powerful predictor [13].

Hypothesis 4

Another four-box grid comparing the presence or absence of group IV patients with the presence or absence of kyphosis was constructed as shown in Table 4. A Fisher test (because the theoretical population was small) was performed, comparing the presence or absence of group IV patients with the presence or absence of kyphosis. In addition, sensitivity, specificity and positive predictive value were calculated. We considered a positive predictive value and specificity of more than 80% to indicate that this test is a powerful predictor [13].

Hypothesis 5

A four-box grid comparing the presence or absence of group III or IV patients with the presence or absence of kyphosis was constructed as shown in Table 5. A Pearson χ^2 test was performed, comparing the presence or the absence of group III or IV patients with the presence or the absence of kyphosis. In addition, sensitivity, specificity and positive predictive value were calculated. We considered a positive predictive value and specificity of more than 80% to indicate that this test is a powerful predictor [13].

Hypothesis 6

For the sixth hypothesis, a four-box grid comparing the presence or absence of group II, III or IV patients with the presence or absence of kyphosis was constructed as shown in Table 6. A Pearson χ^2 test was performed, comparing the presence or the absence of group II, III or IV patients with the presence or the absence of kyphosis. In addition, sensitivity, specificity and positive predictive value were calculated. We considered a positive predictive value and specificity of more than 80% to indicate that this test is a powerful predictor [13].

Hypothesis 7

A four-box grid comparing the presence or absence of group I patients with the presence or absence of spinal deformity (scoliosis, kyphosis or lumbar hyperlordosis) was constructed as shown in Table 7. A Pearson χ^2 test was performed, comparing the presence or the absence of group I patients with the presence or the absence of spinal deformity. In addition, sensitivity, specificity and positive predictive value were calculated. We considered a positive predictive value and specificity of more than 80% to indicate that this test is a powerful predictor [13].

Hypothesis 8

A four-box grid comparing the presence or absence of group II and III patients with the presence or absence of hip dislocation was constructed as shown in Table 8. A Pearson χ^2 test was performed, comparing the presence or the absence of group II or III patients with the presence or the absence of hip dislocation. In addition, sensitivity, specificity and positive predictive value were calculated. We considered a positive predictive value and specificity of more than 80% to indicate that this test is a powerful predictor [13].

Results

Table 9 shows the overall results. The bony level matched the neurological level within one grade in 86 cases (53%). In 59 cases (36%) the neurological level was lower than the bony level. In 18 cases (11%) the neurological level was higher than the bony level.

Intracanal abnormalities were found in 32 patients. Eleven cases of tethered cords were found. In each case, only one cord release was performed. Thirty-one syrinx were noted but none was clinically symptomatic. Two diastematomyelia were noted. There was no evidence of syrinx-related scoliosis in our survey.

Twenty-eight patients with scoliosis (50%) underwent surgery. None from group I, 12 from group II (39% cases among patients with scoliosis from this group), 12 from group III (71% cases among patients with scoliosis from this group) and four from group IV (80% cases among patients with scoliosis from this group). Eleven patients with kyphosis underwent surgery (64.7%), none from group I or II, four from group III (80% cases among patients with kyphosis from this group) and seven from group IV (88% cases among patients with kyphosis from this group). No patient with lumbar hyperlordosis underwent surgery.

Table 7 Hypothesis 7: is group I a powerful predictor for the absence of spinal deformity?

	No deformity	Deformity (scoliosis, kyphosis or hyperlordosis)
Group I patients	63	6
Group II, III or IV patients	32	62

Table 8 Hypothesis 8: is group II or III a powerful predictor for hip dislocation occurrence?

	Hip dislocation	No hip dislocation
Group II or III patients	52	33
Group I or IV patients	9	69

Hypothesis 1

The Fisher test showed the number of spinal deformities (scoliosis, kyphosis or lumbar hyperlordosis) to be statistically different between group IV and group I, II or III patients ($P < 0.01$). The positive predictive value was 100%, the specificity was 100% and the sensitivity was 13% (the prevalence of spine deformity was 0.42). Group IV is considered as a powerful predictor for spinal deformity (scoliosis, kyphosis or lumbar hyperlordosis).

Hypothesis 2

The Pearson χ^2 test showed that the number of spinal deformities (scoliosis, kyphosis or lumbar hyperlordosis) is statistically different between group III or IV and group I or II patients ($P < 0.01$). The positive predictive value was 83%, the specificity was 94% and the sensitivity was 41% (the prevalence of spine deformity was 0.42). Group III or IV is considered as a powerful predictor for spinal deformity (scoliosis, kyphosis or lumbar hyperlordosis).

Hypothesis 3

The Pearson χ^2 test showed the number of spinal deformities (scoliosis, kyphosis or lumbar hyperlordosis) to be statistically different between group II, III or IV and group I patients ($P < 0.01$). The positive predictive value was 66%, the specificity was 66% and the sensitivity was 91% (the prevalence of spine deformity was 0.42). Group II, III or IV is not considered as a powerful predictor for spinal deformity (scoliosis, kyphosis or lumbar hyperlordosis).

Hypothesis 4

The Fisher test showed that the number of kyphosis cases is statistically different between group IV and group I, II or III patients ($P < 0.01$). The positive predictive value was 84%, the specificity was 99% and the sensitivity was 47% (the prevalence of spine deformity was 0.10). Group IV is considered as a powerful predictor for kyphosis.

Hypothesis 5

The Pearson χ^2 test showed that the number of kyphosis cases is statistically different between group III or IV and group I or II patients ($P < 0.01$). The positive predictive value was 38%, the specificity was 86% and the sensitivity was 76% (the prevalence of kyphosis was 0.10). Group III or IV is not considered as a powerful predictor of kyphosis.

Table 9 Overall results

	Number of patients	Spine deformity	Scoliosis	Kyphosis	Hyperlordosis	Hip dislocation
Group I	69	6	3	2	1	9
Group II	60	34	31	2	3	35
Group III	25	19	17	5	0	17
Group IV	9	9	5	8	0	0

Hypothesis 6

The Pearson χ^2 test showed the number of kyphosis cases to be statistically different between group II, III or IV and group I patients ($P < 0.01$). The positive predictive value was 15%, the specificity was 46% and the sensitivity was 88% (the prevalence of kyphosis was 0.10). Group II, III or IV is not considered as a powerful predictor for kyphosis.

Hypothesis 7

The Pearson χ^2 test showed that the number of spine deformity cases is statistically different between group I and group II, III or IV patients ($P < 0.01$). The positive predictive value was 84%, the specificity was 91% and the sensitivity was 66% (the prevalence of kyphosis was 0.42). Group I is considered as a powerful predictor for the absence of spinal deformity.

Hypothesis 8

The Pearson χ^2 test showed the number of hip dislocations to be statistically different between group II or III and group I or IV patients ($P < 0.01$). The positive predictive value was 62%, the specificity was 68% and the sensitivity was 85% (the prevalence of hip dislocation was 0.10). Group II or III is not considered as a powerful predictor for hip dislocation.

Discussion

Spinal deformity in MMC patients may have a dramatic impact on the patients' quality of life. It appears that ambulation and spine deformity are strongly linked. In 1988, a survey pointed out that three major factors have an impact on ambulation in MMC: age, neurological level and scoliosis [6,7]. Actually, spinal deformity in MMC represents a complex, and possibly changing clinical picture that is influenced by numerous variables: spinal cord tethering (changing motor exams) requiring releases, obesity, mental status. Any of these can have an effect on whether spinal deformity was present on a radiograph, and a distinction must be made between transient deformities and structural curves which are known to worsen the patients' quality of life. As recommended by Trivedi [8], scoliosis in MMC should realistically be reserved for curves exceeding 20°. In the same way we chose to define kyphosis as a generalized curve exceeding 50°, and lumbar hyperlordosis as curves over 60° in order to limit the number of transient and reversible deformities.

The neurological level can be accurately assessed quite early in a child's life (at about 5 years of age) [10], but motor examination may worsen over time (and therefore the group in which the patient was initially placed may change) due to symptomatic tethering, contractures of lower extremities, obesity or changes in ambulatory status. In order to control this bias, we based our neurological classification on the earliest reliable reported neurological examination which is made at 5 years (from year five to year six) in our survey, regardless of possible later worsening of the motor power level.

The bony defect level can be accurately assessed long before the patient is 5 years old, but it is known that the bony defect level does not perfectly match the neurological one (only in 53% of cases in our survey did the bony level and neurological level match within one grade). So, since physical muscle testing is feasible from the age of 5 years [10] it is more relevant to base our prognosis on this. Therefore, a global prognosis in MMC patients could be assessed early, matching the neurological level at 5 years and the probability for future spinal deformity, and an adequate therapeutic strategy and follow-up could be planned.

In previous works, some authors focused on predictor factors of scoliosis, regardless of kyphosis in spite of the fact that kyphosis is known to raise specific problems. Trivedi in 2002 [8], reviewing 141 patients, pointed out that the clinical motor level, the ambulatory status and the bony level were all found to be predictive factors for scoliosis development in MMC patients but did not comment on the predictive factor for kyphosis development. Our work not only strengthens Trivedi's [8] opinion about the risk of scoliosis occurrence but some additional knowledge is brought about the risk of kyphosis and lumbar hyperlordosis occurrence. Other works focused on the kyphosis progression rate. Carstens in 1996 [9] established that the progression of the kyphosis in MMC patients depended on the level of paralysis. But Carstens [9] did comment on the risk of kyphosis occurrence among MMC patients, and among recent works, none focused on the predictor factors for kyphosis onset. It has been shown that the higher the neurological level, the higher the rate of spinal deformity at maturity [5,8,9], but no work has provided a guideline that would be usable for physicians to assess the prognosis for future spinal deformity in children with MMC.

Our aim was to establish a neurological motor level classification that could be used as a predictive rule for spine deformities development (not only scoliosis, but also kyphosis or even lumbar hyperlordosis).

The predictive rule is defined as follows: group I, low probability for spine deformity (scoliosis, kyphosis or lumbar hyperlordosis); group II, medium probability for spine deformity (scoliosis, kyphosis or lumbar hyperlordosis); groups III and IV, high probability for spine deformity (scoliosis, kyphosis or lumbar hyperlordosis); group IV, high probability for kyphosis.

Concerning the risk of hip dislocation occurrence, contradicting opinions are reported in the literature. Sharrard in 1964 [14] reviewing 183 patients, suggested that patients with a neurological level above T12 (who

would have been classified as group IV patients according to our criteria) would never suffer from hip dislocation whereas patients with lower neurological levels are prone to develop hip dislocations during their life because of muscle imbalance. But since the work of Beeker in 1986 [15], it has been suggested that hip deformity and dislocation are not as common as first assumed by Sharrard [14], and that the development of these abnormalities cannot be predicted on the basis of the level of lesions [15]. Broughton in 1993 [16] reviewing 802 patients pointed out that hip dislocation in patients over 11 years was distributed as follows: group IV patients (according to our criteria) were dislocated in 59% of cases, group III patients were dislocated in 75% of cases, group II patients were dislocated in 33–50% of cases and group I patients in 2–20% of cases. In our own work, no hip dislocation in group IV patients was found, but because of the small size of this group (only nine cases in our work) it is impossible to give any conclusion about group IV hip dislocation rate. In groups II and III, dislocation rates (group III, 68%, group II, 58%) were close to the rates reported by Broughton [3] and lower than the rates reported by Sharrard [14]. Concerning the dislocation rate in group I patients, it is probably actually higher than suggested by Sharrard [14], but probably lower than reported by Broughton [16]. In our survey, group I patients presented a dislocated hip in 13% cases. The statistical analysis showed that our four-step motor-power classification is not a powerful predictor for hip dislocation, strengthening the opinion of Beeker [15] that the development of hip abnormalities cannot be predicted on the basis of the neurological level of lesions.

In conclusion, as far as the neurological level can be assessed quite early in the MMC children's life [10], future spinal disorders can be expected in these patients, and an appropriate therapeutic strategy and follow-up can be planned. Concerning hip dislocation, no prediction can be done simply based on the neurological level of lesions.

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