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COMPARISON BETWEEN TWO METHODS OF MEASURING PELVIC OBLIQUITY IN CEREBRAL PALSY AND MYELOMENINGOCELE

COMPARAÇÃO ENTRE DOIS MÉTODOS DE MEDIDA DE OBLIQUIDADE PÉLVICA EM PARALISIA CEREBRAL E MIELOMENINGOCELE

COMPARACIÓN ENTRE DOS MÉTODOS DE MEDICIÓN DE LA OBLICUIDAD PÉLVICA EN PARÁLISIS CEREBRAL Y MIELOMENINGOCELE

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ABSTRACT

Objective: To evaluate the intra- and inter-observer reproducibility of the evaluation of the pelvic obliquity (PO) in patients with neuromuscular deformities via the method that uses the iliac crests and the method that uses the upper endplate of S1 and to determine whether there is a relationship between the methods. Methods: The digitized panoramic radiographies of thirty patients with cerebral paralysis or myelomeningocelein outpatient monitoring were evaluated by four examiners: two experienced spinal surgeons and two fellows. Two radiographs were excluded because analysis was impossible. All exams were obtained in accordance with the periodic monitoring protocol in the sitting position, using digitized film and a film-focus distance of 110 cm. Results: High intra- and inter-observer agreement was observed both for method that uses the iliac crests and the method that evaluates the S1 endplate. However, no significant relationship between the two methods was observed. Conclusions: The methods evaluated had good reproducibility and agreement among the observers. It was confirmed that, on account of the existent linear relation, it is possible to estimate the value of the iliac crest method knowing the value obtained by the S1 plateau multiplied by 0.76. There was no agreement between the iliac crest and S1 plateau PO evaluation methods. ***Level of evidence IV; Retrospective cross-sectional study.***

Keywords: Illium; Sacrum; Neuromuscular Diseases; Cerebral Palsy; Meningomyelocele.

RESUMO

Objetivo: Estimar a reprodutibilidade intra e interobservadores da avaliação da obliquidade pélvica (OP) nos pacientes com deformidades neuromusculares pelo método que usa as cristas ilíacas e pelo método que usa o platô superior de S1e avaliar se há relação entre os métodos. Métodos: Trinta radiografias panorâmicas digitalizadas de pacientes com paralisia cerebral e mielomeningocele acompanhados em ambulatório foram avaliadas por quatro examinadores, sendo dois cirurgiões de coluna experientes e doisfellows. Foram excluídas duas radiografias que impossibilitavam a análise. Todos os exames foram obtidos conforme protocolo de acompanhamento periódico, na posição sentada, em filme digitalizado e distância foco-filme de 110 cm. Resultados: Observou-se alta concordância intra e interobservadores tanto do método que usa as cristas ilíacas quanto do método que avalia o platô de S1. No entanto, não foi observada relação significativa entre os dois métodos. Conclusões: Os métodos avaliados apresentam boa reprodutibilidade e concordância entre os observadores. Verificou-se que, por conta da relação linear existente, é possível estimar o valor do método das cristas ilíacas conhecendo o valor obtido pelo método do platô de S1 multiplicado por 0,76. Não houve concordância entre o método de avaliação da OP pelas cristas ilíacas em comparação com o método de avaliação pelo platô de S1. ***Nível de evidência IV; Estudo transversal retrospectivo.***

Descritores: *Crista Ilíaca; Sacro; Doenças Neuromusculares; Paralisia Cerebral;Meningomielocele.*

RESUMEN

Objetivo: Estimar la reproducibilidad intra e interobservador de la evaluación de la oblicuidad pélvica (OP) en pacientes con deformidades neuromusculares por el método que utiliza las crestas ilíacas y por el método que utiliza la meseta superior de S1 y evaluar si existe una relación entre los métodos. Métodos: Treinta radiografías panorámicas digitales de pacientes con parálisis cerebral y mielomeningocele acompañadosen clínica ambulatoria fueron evaluados por cuatro examinadores, dos cirujanosde columna de vasta experiencia y dos fellows. Se excluyeron dos radiografías porque imposibilitaban el análisis. Todos los exámenes se obtuvieron según el protocolo de acompañamiento periódico, en posición sentada, sobre película digitalizada y distancia foco-película de 110 cm. Resultados: Se observó una alta concordancia intra e interobservador tanto en el método que utiliza las crestas ilíacas como en el que evalúa la meseta de S1. Sin

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embargo no se observó ninguna relación significativa entre los dos métodos.Conclusiones: Los métodos evaluados presentaron buena reproducibilidad y concordancia entre los observadores. Se observó que, debido a la relación lineal existente, es posible estimar el valor del método de las crestas ilíacas conociendo el valor obtenido por el método demeseta de S1 multiplicado por 0,76. No hubo concordancia entre el método de evaluación de la OP por las crestas ilíacas en comparación conel método de evaluación por meseta de S1. ***Nivel de evidencia IV; Estudio transversal retrospectivo.***

Descriptores: *Ilion; Sacro; Enfermedades Neuromusculares; Parálisis Cerebral; Mielomeningocele.*

INTRODUCTION

Neuromuscular diseases are responsible for major spinal deformities, causing postural and muscular imbalance throughout the individual's development, resulting in pronounced long- and short-radius curves associated or not with compensatory curves, kyphotic deformities, and the occurrence of pelvic obliquity.[1,2]

Neuromuscular scolioses are classified according to the SRS (Scoliosis Research Society) as neuropathic and myopathic, depending on the causal factor. Neuropathic scolioses are subdivided into upper motor neuron lesions, represented by cerebral palsy, lower motor neuron lesions, and mixed lesions, represented by myelomeningocele.

Pelvic obliquity (PO) is defined by the SRS as the angulation of the pelvis in relation to the horizontal in the frontal plane. Dubousset et al. defined PO as any imbalance between the spine and the pelvic structures, both in relation to the frontal and horizontal planes.[3] This imbalance can be caused by lower limbs of different lengths, hip contractures, spinal deformities, or a combination of these.[1,2,4] For this reason, the adequate measurement of PO using radiographs is an important tool in therapeutic orientation and in the planning of the surgical correction.

Myelomeningocele and cerebral palsy patients with a high PO angle value may present subluxation of the sacroiliac joints.[1,5] One of the most used measurement methods, that of horizontal pelvic obliquity, does not consider this possibility and its possible influence on the value obtained. Thus, the S1 vertebra and its endplate would be useful parameters in measuring pelvic obliquity in patients with neuromuscular scoliosis and muscle contractures.[4,6]

The objective of the present study is to compare horizontal pelvic obliquity measured using the iliac crests with a new method using the anatomical reference of the S1 plateau, testing intra- and interobserver variability.

METHODS

The project was duly submitted to Plataforma Brasil (27060314.2.0000.0085) and approved by the Institutional Review Board.

In the second half of 2014, an initial retrospective sample of 30 outpatients with neuromuscular scoliosis from a tertiary hospital was divided into two groups: the first made up of patients with cerebral palsy and the second of patients with myelomeningocele. The cerebral palsy patients were classified according to the GMFCS (Gross Motor Function Classification System),[7] which quantifies the level of involvement. The patients with myelomeningocele were stratified according to the topographical classification of the lesion as thoracic, high lumbar, low lumbar, or sacral.

Pelvic obliquity was measured and analyzed in 30 radiographical examinations, two of which were excluded for poor visualization of the anatomical markers due to distended intestinal loops (one of them from a patient with thoracic myelomeningocele and the other with quadriplegic spastic cerebral palsy).

Anteroposterior and lateral panoramic radiographs were obtained in accordance with the periodic monitoring protocol, in the sitting position, on 35 cm x 43 cm digital film, at a film-focus distance of 110cm with the table in the horizontal and vertical positions, using a Shimadzu FLEXIVISION X-ray machine with a multipurpose remote-controlled table.

PO was measured using the horizontal pelvic obliquity method. The angle formed by the intersection of a line passing through the vertebral center of T1 to the vertebral center of S1 with a line adjacent to the upper edge of the iliac crests was measured (Figure 1A)[8] and then,the angle formed by the same vertical lane and the line tangent to the S1 endplate was measured (Figure 1B).

In cases where demarcation of the S1 endplate is difficult, the sacral intersulci line was used, which generally has the best anatomical correlation with the sacral surface line.[9]

The physicians responsible for the measurements underwent training/calibration of the two techniques before performing the actual measurements. All the measurements were taken by four orthopedic physicians: two physicians *working on postgraduate specializations in spinal surgery* and two experienced spinal surgeons. After an interval of 21 days, the same examiners performed new measurements of the same examinations in order to obtain intra-observer agreement.

For better characterization of the PO measurement methods in two different populations, all analyses were conducted separately between the group of patients with cerebral palsy and the group of patients with myelomeningocele.

The normality of the PO values obtained from each method was analyzed using Q-Q graphs and visually assessed using histograms. After confirming the normality of the distribution, outliers, defined as PO values with intra-evaluator variation greater than a 99% degree of acceptance (mean of the difference between the first and second evaluationswith a standard deviation of ±2.576) were excluded. After this exclusion, the Kolmogorov-Smirnov test was applied to assess the normality of the final sample.

The reliability of the two PO calculation methods was evaluated by calculating the intraclass correlation coefficient (ICC) and its 95% confidence interval. For the inter-observer variability, different ICCs were calculated for the first- and twenty-first-day evaluations. For the intra-observervariability, an ICC was calculated for each evaluator. All the ICCs were calculated based on an absolute agreement model.

The standard error of measurement (SEM)of both PO calculation methods was estimated as the square root of the mean squared error of the analysis of variance produced during the calculation of the ICCs as a measure of the repeatability of the measurements.

The agreement between the methods was calculated by the ICC with the respective confidence intervalof 95% for the first evaluation of the main evaluator and the repeatability was extracted to estimate the error between the methods. Using a Bland-Altman plot, possible trends between the measurements of the methods were assessed.

A linear regression model was also created to estimate the value of

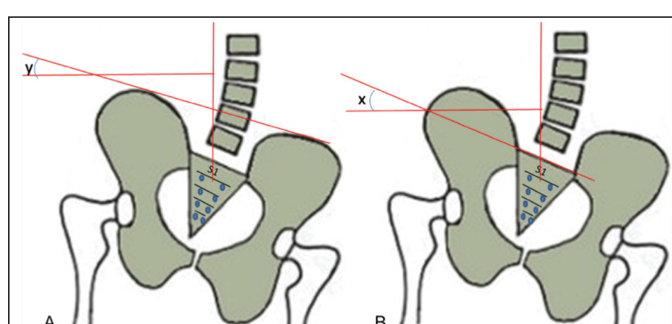

Figure 1. Horizontal pelvic obliquity method. A – measured with reference to the iliac crests, B – measured with reference to the S1 endplate.

the iliac crest method as a function of the S1 endplate method. Thus,we evaluated whether it is possible to obtain the value ofone method by knowing the value of the other, even with no agreement between them.

All analyses were conducted in SPSS version 20.0 (IBM Statistics).

RESULTS

Table 1 shows that the measurements taken by the same method present quite similar mean values for all evaluations and also among the evaluators. Between the methods it was observed that the mean angle obtained by the S1 endplate method was always greater than the mean angle obtained by the iliac crest method.

Table 2 shows that the intra-observer reproducibility of both methods was quite high (ICC > 0.9). Only the last evaluatorhad lower interclass correlation values, but these did not compromise the reproducibility of the PO angle values. The repeatability measure shows that, mainly for the first two evaluators, the error of the S1 endplate method was greater,at 4^o to 6^o, than that of the iliac crest method, at 2.5^o to 3^o.

Table 3 shows that inter-observer reproducibility for each PO measurement method was as high as intra-evaluator reproducibility (Table 2), that the interclass correlation coefficients were always greater than 0.8, and that the estimated errors (repeatability) were similar to the intra-evaluator onesobtained. The only exception occurred among the less experienced evaluators in the second series of angle measurements for the iliac crest method, but the ICC was still greater than 0.7 (ICC = 0.703).

Figure 2 suggests the randomness of the differencesin angles between the evaluation methods but we can see that the values of the angles obtained using the S1 endplate method are always higher than those obtained by the iliac crest method, as the values in the graphare concentrated in the negative part and the mean difference between the angles was -9.5^o. There was no agreement between the methods (ICC = 0.723; CI95%: -0.072 to 0.923), as the interclass correlation coefficient presented a confidence interval that passes through zero.

Figure 3 shows that there is a linear relationship between the angles evaluated by the two methods. Despite the absence of agreement in the results, it is possible to estimate the value of one of the methods from the known value of the other. Using the angle evaluated by the S1 endplate method to estimate the angle obtained by the iliac crest method, the model accounts for 84.6% of the variability of the crest angles when the endplate angle is known, and the expected crest angle can be obtained by multiplying the endplate angle by 0.76. For example, an angle of 34^o given by the endplate method would have a mean crest-method angle of $0.76 \times 34^o = 25.8^o$.

DISCUSSION

Theoretically, the sacrum acts as cornerstone inverted as a mediator between the lower limbs and the pelvis, paired with the spine.[5] The rotation of the ilium inevitably causes an inclination of the sacrum, predisposing to an imbalance of the sacral base. The result is compensatory curvature of the spine.[10]

Pelvic asymmetry can also be caused by a dysfunction of the sacroiliac joint,[10] being a confounding factor when quantifying pelvic obliquity, especially in neuromuscular scolioses. Because of this

Table 2. Results of the intra-evaluator agreement measurements for each PO angle measurement method.

Evaluator	Method	ICC	CI 95%,		Repeatability
			Lower	Upper	
Evaluator 01	Iliac Crests	0.942	0.867	0.974	2.94
	S1 Endplate	0.915	0.827	0.960	4.09
Evaluator 02	Iliac Crests	0.964	0.925	0.983	2.41
	S1 Endplate	0.851	0.705	0.928	5.91
Evaluator 03	Iliac Crests	0.982	0.698	0.995	1.17
	S1 Endplate	0.969	0.687	0.991	1.63
Evaluator 04	Iliac Crests	0.745	0.489	0.878	5.97
	S1 Endplate	0.902	0.801	0.953	4.68

Table 3. Results of the inter-evaluator agreement measurements for each PO angle measurement method.

Evaluators	Evaluation	Method	ICC	CI 95%		Repeatability
				Lower	Upper	
More experienced	First	Iliac Crests	0,936	0.867	0.970	3.20
		S1 Endplate	0,807	0.625	0.906	6.19
	Second	Iliac Crests	0,959	0.910	0.981	2.51
		S1 Endplate	0,902	0.798	0.953	4.78
Any less experienced	First	Iliac Crests	0,989	0.977	0.995	1.46
	Second	S1 Endplate	0,890	0.777	0.948	4.76
	First	Iliac Crests	0,703	0.311	0.970	6.11
	Second	S1 Endplate	0,915	0.825	0.960	3.95

Table 1. Description of the PO angles evaluated by 4 evaluators and by the two methods and the results of the normality ofthe data distribution test.

Evaluator	Evaluation	Method	Mean	SD	N	Kolmogorv-Smirnov Z	P
Evaluator 01	First	Iliac Crests	27.9	12.5	28	1.03	0.238
		S1 Endplate	37.4	13.2	28	0.61	0.848
	Second	Iliac Crests	29.7	13.5	28	0.72	0.676
		S1 Endplate	38.5	14.9	28	0.57	0.898
Evaluator 02	First	Iliac Crests	28.4	12.5	28	0.91	0.378
		S1 Endplate	38.3	14.6	28	0.39	0.998
	Second	Iliac Crests	28.4	12.7	28	0.96	0.318
		S1 Endplate	40.5	16.4	28	0.71	0.695
Evaluator 03	First	Iliac Crests	26.9	14.2	28	0.81	0.526
		S1 Endplate	37.3	13.7	28	0.53	0.938
	Second	Iliac Crests	29.1	14.2	28	1.02	0.248
		S1 Endplate	39.8	13.7	28	0.58	0.886
Evaluator 04	First	Iliac Crests	27.1	13.7	28	0.85	0.472
		S1 Endplate	39.2	15.6	28	0.65	0.787
	Second	Iliac Crests	23.1	11.4	28	0.91	0.382
		S1 Endplate	38.1	14.2	28	0.77	0.601

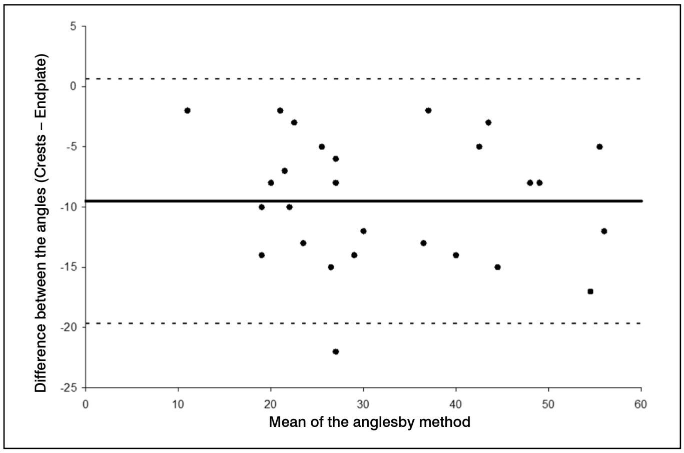

Figure 2. Brand-Altman plot showing the differences between the methods for the first measurement by the main study evaluator.

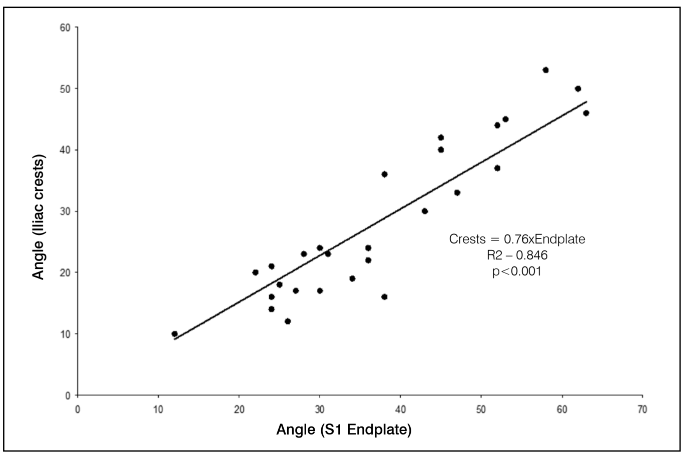

Figure 3. Dispersion diagramof the PO angle evaluated by each method and the results of the linear regression model of the angle evaluated by the crest method as a function of the angle evaluated by the endplate.

peculiarity, the S1 vertebra and its endplate would be a good parameter for measuring pelvic obliquity in patients with neuromuscular scoliosis and muscle contractures.[4,6]

One of the difficulties in demarcating the true S1 endplate occurs with the increase in sacral angulation and the consequent radiographic superpositioning of the posterior arches in relation to the sacral surface(which are normally visualized below the S1 endplate when the sacral angulation is less than 20˚to 25º).[11] In these cases, the line of the sacral intersulci can be used as a method correlating with the sacral surface.[4,6,9]

Both methods are reproducible, as observed by the mean values obtained for each method by the same examiner and among all the examiners.

Identifying the iliac crests in the radiographical exams is generally easier than identifying the S1 endplate. This fact explains in part why the errors of measurement will be slightly greater in the second method. Even so, both the methods have high intra-observer agreement.

Because the execution of each one of the methods is based on fixed parameters inradiographic projections, it is believed that the greatest difficulty is precisely the identification of these parameters. The high quality of radiographic exams, especially digital ones, facilitates the identification and measurement of the PO and contributed to the strong inter-observer agreement obtained.

The measurements taken using the iliac crest method also yielded lower absolute values than those obtained by evaluating the S1 endplate. On average, the value of the first method corresponds to 76% of the second (PO by the iliac crest method = 0.76 x the s1 endplate method)**,** obtained through the linear relationship between the measured angles.

The S1 endplate method value was always higher, reaching a mean of 9.5º for the main evaluator of this study. When we compared the two methods using the values of this evaluator, there was no agreement in relation to the values obtained.

The anatomical relationship between the iliac crests and the S1 plateau is limited. However, rotational and angular changes in the pelvis that cause a dissociation of this relationship can be explained, for example, due to possible changes in the mobile segment that separates the two structures, that is, the sacroiliac joints. In this case, anatomical radiological studies of the joints may provide additional information.

Differences can also be based on technical difficulties at the time of measurement.Changes in sacral inclination can make it difficult to establish the S1 endplate in anteroposterior radiographs. However, even considering this difficulty, there was high intra- and inter-observer agreement in the evaluation using the S1 endplate.

CONCLUSION

High degrees of inter-observer and intra-observer agreement were observed in the measurement of pelvic obliquity using both the iliac crest and S1 plateau methods.

We observed that, due to the existing linear relationship, it is possible to estimate the value of the iliac crest methodif we know the value obtained by the S1 endplate method, multiplying it by 0.76 (PO by the iliac crest method = 0.76 x the s1 endplate method).

There was no agreement between the iliac crest and S1 endplate PO evaluation methods.

All authors declare no potential conflict of interest related to this article.

CONTRIBUTIONS OF THE AUTHORS: MVBM, SNM, and RR: data collection, radiographical measurements; MVMB, SNM, RR, and PTMC: data collection, radiographical measurements, writing of the article. PTMC: review and study concept. AJRR and MINR: radiographical measurement, review, and study concept.

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