

Benefits from using an ankle-foot orthosis in children with myelomeningocele

Benefícios do uso da órtese tornozelo-pé nas crianças com mielomeningocele

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ABSTRACT

Myelomeningocele (MMC) or *spina bifida* is a defect of the neural tube in which the spinal cord, its envelopes (meninges), and vertebral arches develop abnormally in the beginning of gestation, and owing to this failure of closure there appear a series of congenital defects and associated comorbidities, impairing in several aspects the functioning of the life of children with MMC. The congenital clubfoot has been found the most common orthopaedic anomaly in patients with MMC. The ankle-foot orthosis (AFO) is an orthopaedic device commonly used by these children to minimize the sequelae caused by this anomaly. **Objective:** Identify the functional benefits brought about by the use of the AFO to children with MMC, as reported by their guardians. **Method:** Descriptive, transversal study. Convenience sample consisting of 25 guardians of children with MMC who were using/had used an AFO. **Results:** Eighty percent of the guardians have reported at least one benefit brought about by the use of the AFO, among them: improvement in foot position (68%), foot growth (40%), improvement in foot balance (32%), and balance sitting position (15%). **Conclusion:** The use of the AFO by children with MMC can provide several benefits reported by their guardians.

Keywords: Ankle-foot orthosis (AFO), myelomeningocele (MMC), benefits, child, functioning, prevention.

RESUMO

A mielomeningocele (MMC) ou espinha bífida é um defeito do tubo neural no qual a medula espinal, seus envoltórios (meninges) e os arcos vertebrais desenvolvem-se anormalmente no início da gestação e, como consequências, temos uma série de defeitos congênitos e comorbidades associadas prejudicando a funcionalidade em diversos aspectos da vida das crianças com MMC. O pé torto congênito foi apontado como a anomalia ortopédica mais comum nos pacientes com MMC e a órtese tornozelo-pé (OTP) é um aparelho ortopédico que pode ser usado nessas crianças para amenizar as sequelas geradas por essa anomalia. **Objetivo:** Identificar os benefícios funcionais trazidos pelo uso da OTP em crianças com MMC relatados pelos seus responsáveis. **Método:** Estudo descritivo, transversal. Amostra de conveniência composta por responsáveis de 25 crianças com MMC que usavam/usaram a OTP que responderam a uma entrevista estruturada que contemplava as variáveis estudadas. **Resultados:** Oitenta por cento dos responsáveis relataram pelo menos um benefício causado pelo uso da OTP, dentre eles: melhorou a postura do pé (68%), o pé cresceu (40%), melhorou o equilíbrio em pé (32%), melhorou o equilíbrio sentado (15%). **Conclusão:** O uso da OTP em crianças com MMC pode proporcionar benefícios identificados pelos responsáveis.

Palavras-chave: Órtese tornozelo-pé (OTP), mielomeningocele (MMC), benefícios, criança, funcionalidade, prevenção.

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INTRODUCTION

Myelomeningocele (MMC) or *spina bifida cistica* is one of the most complex congenital malformations compatible with life. Children born with this malformation may have lesions in the spinal cord as well as in the brain.¹ Its global incidence varies from 0.1 to 10 cases in each 1.000 live newborns. The highest incidence is associated with regions of low socioeconomic development, as shown in the congenital defect chart published by the World Health Organization in 2003.² According to these data, the lowest incidences of *spina bifida cistica* in 1.000 live newborns occur in France/Paris (0.077) and England/Wales (0.095), and the highest incidences occur in Mexico (1.525) and Venezuela (1.196).² Brazil was mentioned in this study as occupying the fourth highest place in the incidence of *spina bifida cistica* among the forty-one countries researched, with a 1.139 rate in each 1.000 live newborns.²

Considering that the survival and quality of life of such patients are subject to neurological, urological and orthopaedic complications, as well as general ones, it is important to stimulate prevention as well as diagnosis and early treatment of this incapacitating disease, aiming at reducing its impact on society.³

The use of conservative methods of treatment during the first year of life can frequently prevent deformities caused by wrong posture or by muscle action non-opposed by its antagonists. These deformities develop with the child's growth and locomotion.⁴

Congenital clubfoot has been found the most common orthopaedic anomaly in patients with MMC in a reference hospital in Rio de Janeiro.⁵ The introduction of the orthosis during the first year, for weight load, and the attempts on the part of the child to stand have been considered *criteria* for a gait and orthostatic posture training programme to be started.⁶

Orthoses can be described as a system of forces that can act over a given part of the human body, aiming at various ends.⁷ The most evident indications for ankle orthosis are: secondary pain and instability, rigidity and deformities. For these problems, to ensure outside support, the essential type of orthosis is the traditional AFO (ankle-foot orthosis).⁷ AFOs are made of high temperature thermoplastics, and polypropylene is the most frequently used material in the making of such orthosis.⁷

The AFO should be used by the infant who already presents contractures, with the aim of lengthening

its soft tissues. It can be used on the anterior aspect when the goal is the correction of the calcaneus valgus, or on the posterior one, when it is to maintain the foot in the plantigrade position, to improve posture. If the AFO is used during sleep as well as part of the day, and if it is associated with passive movement, the acquired deformities of the foot tend to be less severe.⁴

The actual care for these children requires not only the adaptation of the family to the new routine, but also the learning of specific caring, such as vesical catheterism, administration of continuous use medication, prevention of skin lesions, use of orthosis, among others.³

The objective of this study is to identify the benefits of the use of an AFO accrued to children with MMC, recognised by their parents' reports.

MATERIALS AND METHODS

This study has obtained the approval of the Ethics Committees of the participating Institutions (0063.1.008.231-10), and complies with the rules of Resolution 196/96 of the National Health Council, and all the parents of the infants studied have signed an informed and free consent term (IFCT).

This is a descriptive and transversal study. The data were obtained by means of structured interviews with the parents of the children with MMC during a 13-month period, by the filling out of a questionnaire devised by the researcher. The data collected have been stored in a data bank (Excel) and a descriptive analysis was made with frequency distribution and central and dispersion trend measurements. The sample has been constituted according to the following inclusion *criteria*: children from 6 months to 3 years of age with MMC, who were using/had used an AFO, whose guardians had agreed to participate in the research and to sign the IFCT. There have been no exclusion *criteria*.

The variables studied have been: the child's gender, level of the myelodysplasia, the child's age, the child's age at the moment of prescription of the AFO, the professional who has prescribed the AFO, the reason for its prescription, the recognition of any benefits, and the kind of the benefit(s) brought about by the use of the AFO.

The data were collected at Fernandes Figueira Institute (IFF – Fiocruz) because it is a highly-re-

garded maternity hospital that cares for women with risk pregnancy and/or with foot uses presenting congenital anomalies. The data were also collected at Association of the Friends of Handicapped Children (AFHC) because it is a highly-regarded Rehabilitation Centre in an under-privileged neighbourhood of Rio de Janeiro that cares for children with MMC who live there.

RESULTS

The parents of twenty-five children with MMC were found eligible for the study. Twenty-five parents of children from six and thirty-three months of age were interviewed (mean: 20.4; DP \pm 7.7). The most frequent myelodysplasia was at the higher lumbar level (7/28%), followed by lower lumbar and sacral (6/24% each), thoracic (3/12%). In three children it was not possible to ascertain the level or the lesion. Distribution by gender: 14 female and 11 male. The most frequent moment of the AFO prescription was during the second semester of life (14/56%), followed by the first semester (10/40%) and the third semester (1/4%).

Table 1 provides the distribution of the categorical variables analysed.

Table 1. Distribution of the reasons of the AFO prescription, which professional prescribed it and the perception of benefits

Variable	Category	n	%
Professional who prescribed the AFO	Physician	15	60
	Physiotherapist	10	40
Reason for prescription	Prevent deformities	10	40
	Correct deformities	10	40
	Stand/walk	1	4
	Don't know	4	16
Perception of a benefit*	Yes	20	90.9
	No	2	9.1
Improvement in foot posture	Yes	17	85
	No	3	15
Improvement in balance for sitting	Yes	3	15
	No	17	85
Improvement in standing balance	Yes	8	40
	No	12	60
Growth of feet	Yes	10	50
	No	10	50

* Three did not know how to answer.

DISCUSSION

According to the literature, MMC can affect both genders in a similar way.⁸ Our sample revealed a slight predominance of the female gender, in agreement with some studies,^{9,10} but at variance with others.^{11,12} As to the location of the lesion, some studies^{5,6,8,13-15} have shown that the majority were found in lower lumbar and sacral levels. Our study has evidenced a higher incidence of the higher lumbar level (28%) and the majority was only affected at the lumbar level (52%). The differences above reported may have occurred owing only to the type of sample and the number of participants involved. According to the studies analysing the use of the AFO, the *criteria* for its prescription was contingent to the deambulation prognosis, that is, patients with lesion at lower lumbar or sacral levels would be prescribed an AFO, since they would show a better prognosis for deambulation.^{8,16,17}

One review of literature showed that, besides the levels of the lesions, orthopaedic alterations were found in 40% of the articles analysed.¹⁸ The authors suggest that orthopaedic changes have as much importance as the levels of the lesions in the deambulatory prognosis in MMC.¹⁸ In the present study, the referral to the use of an AFO seemed not to occur exclusively due to the deambulation prognosis, since 40% of the children with MMC presented their lesion in higher lumbar/thoracic levels and yet/also used AFOs.

In children with MMC, the neurological level of the lesion can determine functional deficits characterised by the wrong skeletal alignment which, if disregarded, may turn deambulation problematic and later cause pain the joints. For this reason, functional deficits require orthotic treatment, to improve alignment and protect lower members from pathological forces that may limit mobility.¹⁹ Independently of the level of the lesion the AFO may be prescribed to patients with MMC for different reasons. In the group presenting thoracic or higher lumbar lesions, the AFO may be prescribed to prevent or correct deformities; in the group with lower lumbar or sacral lesions, the AFO may be prescribed to improve orthostatic posture and gait, and need a good foot alignment.²⁰ Children with a higher level lesion (thoracic) have worse prognosis for gait, but they need correct foot alignment for the use of a wheelchair.

Children with MMC who present any postural foot alteration need an AFO or corrective surgery followed by the use of orthosis.²¹

The capacity a child with MMC should have of learning the world and of developing its individuality is frequently impaired by its difficulty in actively assuming orthostatic postures (standing or sitting) and in locomotion. It is enough to watch how a twelve-month-old healthy baby explores its environment to realize how poor in experiences must be the world of the handicapped child; measures can be taken to avoid this dearth of experience.⁴ Individuals with MMC present a delay in their sensory-motor development, and one of the reasons for this is the alteration in the strength of lower members.²² For this reason they are referred to a follow-up in the areas of rehabilitation, including physiotherapy, with the aim of the improving muscular strength and the preventing contractions.²² Further objectives of the prescription of orthoses for the feet are: support load, reduce pressures, alleviate pain and improve mobility.²³ The objectives of the prescription of the AFO in this study have been: prevent deformity of feet (40%), correct deformity of feet (40%) and assume orthostatic stance/deambulate (4%); 16% of the parents had no knowledge of the reason for the AFO prescription. Our results point to objectives similar to those described in the literature, with the proviso that both prevention and correction of deformities have prevailed.

The introduction of the orthosis during the first year, for weight load, and the child's attempts to stand have been considered enough reason to begin a training programme for gait and orthostatic posture.⁵ There has been concern, in our study, for the early use of the AFO, before the first year of life was complete, since the majority of the prescriptions were made during the first or second semesters of life.

We have not found studies that had exclusively researched the benefits of the use of an AFO by patients with MMC. There are studies about deambulation orthoses in which the AFO is coupled to a long tutor aiming at aligning the feet during gait.²⁴⁻²⁷

A child with MMC may show a delay in the development of head and thorax control. It will take longer to balance itself in the sitting position whenever its head control is precarious, and it is slow in learning the postural adjustments specific to a task

and its respective context, since the palsy of the lower members reduces the efficacy of the postural adjustments in the sitting position, which demand bracing of feet or thighs.⁴

It is known that for the deambulation of a child with MMC to occur, previous orthostatism training is necessary, and so that this training may occur with no lesions, the adequate alignment of the feet bearing the weight of the child, that is, plantigrade feet, is of paramount importance.^{24,28,29} The benefits reported in the present study confirm the data related in the literature, that AFOs may provide greater postural stability, thus affording their users the benefits of improved balance and posture.³⁰

As far as we have been able to ascertain, this is the first study to consider the satisfaction of the parents of children with MMC with the use of the AFO.

The majority of parents (80%) reported having noticed at least one benefit resulting from the use of the AFO, such as: improvement in foot position (68%), growth of foot (40%), improvement of balance in orthostatic posture (32%), improvement of balance in sitting (32%).

In some researches, the importance of the family's satisfaction regarding their relatives' quality of life is emphasised.³¹⁻³³ The needs of the children/families encompass physical, psychosocial and financial issues. One of the reasons for dissatisfaction is the inconvenience in obtaining service in the area of health.³¹

A disabling condition, such as MMC, may cause impairment to self care and mobility, and cause social isolation and low self esteem. This impairment directly affects the quality of life of children with MMC and of their families.³⁰ A study affirms that the concept of quality of life represents a step forward in the perspective of enlargement the concepts good health/ill health, and that the presence of the stigma linked to this condition causes dissatisfaction in young people, and interferes in their place in society as well as in their expression of happiness, social recognition and self esteem, that is, in their quality of life.³³

As a possible limitation of the present study, it is important to mention the high quality of the professionals who work in the two highly-regarded health care centres where the collection of data was conducted. The expertise of these professionals may have been the reason for their early prescriptions of the

AFO, which may have biased the positive/good results of this research.

CONCLUSION

The use of the AFO by children with MMC may afford benefits recognised by their parents, and such a finding should be taken in consideration by the health professionals, for the prescription and the optimisation of the use of AFOs. The positive reports encourage the families of the children with MMC to try and improve the quality of their lives.

This study could assume the role of catalytic of new researches that may allow the further examination of the issues here raised, such as the early use of AFOs by children with MMC and its benefits in the quality of life of these children. As a suggestion for future studies is the early intervention field, since the health professionals feel the need for more evidence in this area.

REFERENCES

- Liptak GS, Dosa MP. Mielomeningocele. Upstate Medical University, Syracuse, NY, USA, 2010.
- Bizzi JWW, Machado A. Mielomeningocele: conceitos básicos e avanços recentes. *J Bras Neurocirurg*. 2012;23(2):138-51.
- Medeiros DRM, Teixeira LL, Saraiva LLO, Costa DCSC, Nascimento LSGC. Plano terapêutico multidisciplinar para crianças com mielomeningocele em um hospital universitário no interior do Rio Grande do Norte. *R Bras Ci Saúde*. 2011;15(2):219-22.
- Shepherd R. *Fisioterapia em Pediatria*. 1. ed. São Paulo: Santos, 1998. p. 244-53.
- Sá MRC, Orsini M, Abelheira L, Sobler MP. Perfil de crianças com mielomeningocele em hospital de referência – Rio de Janeiro. *Rev Bras Neurol*. 2010;46(4):7-11.
- Bartonek A. Motor development toward ambulation in preschool children with myelomeningocele--a prospective study. *Pediatr Phys Ther*. 2010 Spring;22(1):52-60.
- Osqueteau TC, Eckardt LR. Ortesis para pie y tobillo. *Reumatología*. 1999;15(1):51-5.
- Umphred DA. *Fisioterapia neurológica*. 2. ed. Lilia Breteniz Ribeiro. São Paulo: Manole, 1994. p. 397-400.
- Castro SC, Moraes DCG, Martins THP, Comácio SM. Estudo retrospectivo de casos de disrafismo espinhal no Hospital de Clínicas da Universidade Federal de Uberlândia (HCUFU), no período de 1992-2002. *Biosci J*. 2010;26(4):653-60.
- Brandão AD, Fujisawa DS, Cardoso JR. Características de crianças com mielomeningocele: implicações para a fisioterapia. *Fisioter Mov*. 2009;22(1):69-75.
- Llopis ID, Muñoz MB, Agulló EM, et al. Ambulation in patients with myelomeningocele: a study of 1500 patients. *Paraplegia*. 2009;31:28-32.
- Baldisserotto CM, Kondo LH, Chamliam TR. Perfil epidemiológico dos pacientes com mielomeningocele do centro de reabilitação Lar Escola São Francisco. *Med. Rehabil*. 2010;29(3):76-9.
- Ulsenheimer MMM, Antoniuk SA, Santos LHC, et al. Mielomeningocele: a Brazilian University Hospital experience. *Arq. Neuro-Psiquiatr*. 2004;62(4):963-8.
- Filgueiras MG, Dytz JL. Avaliação do perfil de recém-nascidos portadores de defeitos do tubo neural. *Brasília Med*. 2006;43:17-24.
- Neto LM, Dirani M, Alimena LJM. O perfil da mielomeningocele em um hospital pediátrico. *Rev Bras Ortop*. 1999;34(4):295-8.
- Kao PC, Ferris DP. Motor adaptation during dorsiflexion-assisted walking a powered orthosis. *Gait Posture*. 2009;29(2):230-6.
- Wolf SI, Alimusaj M, Rettig O, Doderlein L. Dynamic assist by carbon fiber spring afos for patients with myelomeningocele. *Gait Posture*. 2008;28:175-7.
- Ramos FS, Macedo LK, Scarlato A, Herrera G. Fatores que influenciam o prognóstico deambulatório nos diferentes níveis de lesão da mielomeningocele. *Rev Neurociênc*. 2005;13(2):80-6.
- Malas BS. What variables influence the ability of an AFO to improve function and when are they indicated? *Clin Orthop Relat Res*. 2011;469(5):1308-14.
- Thomas SE, Mazur JM, Child ME. Quantitative evaluation of AFO use with myelomeningocele children. *Z Kinderchir*. 1989;44 Suppl 1:38-40.
- Westcott MA, Dynes MC, Remer EM, Donaldson JS, Dias LS. Congenital and acquired orthopedic abnormalities in patients with myelomeningocele. *Radiographics*. 1992;12(6):1155-73.
- Collange LA, Franco RC, Esteves RN, Zanon-Collange N. Desempenho funcional de crianças com mielomeningocele. *Fisioter Pesqui*. 2008;15(1):58-63.
- Seitzberg A, Lind M, Bierring-Sorensen F. Ambulation in adults with myelomeningocele. Is it possible to predict the level of ambulation in early life? *Childs Nerv Syst*. 2008;24:231-7.
- Thomas SS, Buckon CE, Melchionni J, Magnusson M, Aiona MD. Longitudinal Assessment of Oxygen Cost and Velocity in Children with Myelomeningocele: Comparison of the Hip-Knee-Ankle-Foot Orthosis and the Reciprocating Gait Orthosis. *J Pediatr Orthop*. 2001;21(6):798-803.
- Gerritsma-Bleeker CLE, Heeg M, Vos-Niël H. Ambulation with the reciprocating-gait orthosis. Experience in 15 children with myelomeningocele or paraplegia. *Acta Orthop Scand*. 1997;68(5):470-3.
- Malagon V, Carrillo G, Diaz A, et al. Los trastornos músculo-esqueléticos em el mielomeningocele. *Rev Colomb Ortop Traumatol*. 1988;2(2):55-71.
- Yngve DA, Douglas R, Roberts JM. The reciprocating gait orthosis in myelomeningocele. *J Pediatr Orthop*. 1984;4(3):304-10.
- Crabtree CA, Higginson JS. Modeling neuromuscular effects of ankle foot orthoses (AFOs) in computer simulations of gait. *Gait Posture*. 2009;29(1):65-70.
- Santos CMT, Pereira CU, Santos EAS, Monteiro JTS. Reabilitação na Mielomeningocele. *Revista Brasileira de Medicina*. 2007;64(11):518-20.
- Ring H, Treger I, Gruendlinger L, Hausdorff M. Neuroprosthesis for footdrop compared with an ankle-foot orthosis: effects on postural control during walking. *J Stroke Cerebrovasc Dis*. 2009;18(1):41-7.
- Cipriano MAB, Queiroz MVO. Cuidado com a criança portadora de mielomeningocele: vivência da família. *Rev Rene*. 2008;9(4):72-81.
- Medeiros DRM, Teixeira LL, Saraiva LLO, Costa DCSC, Nascimento LSGC. Plano terapêutico multidisciplinar para crianças com mielomeningocele em um hospital universitário no interior do Rio Grande do Norte. *Rev Bras Ci Saúde*. 2011;15(2):219-22.
- Soares AHR, Moreira MCN, Monteiro LMC, Pohl HG. A qualidade de vida de jovens portadores de espinha bífida do Children's National Medical Center- Washington, DC. *Ciênc. Saúde Coletiva*. 2006;11(3):13-21.