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Bone mineral density and body composition in a myelomeningocele children population: effects of walking ability and sport activity

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Abstract. – Myelomeningocele causes serious locomotor disability, osteoporosis and pathologic fractures. The aim of this study was to investigate the relationship between body composition, bone mineral density, walking ability and sport activity in myelomeningocele children.

60 patients aged between 5 and 14 yrs with myelomeningocele (22 ambulatory and 38 non-ambulatory), were studied. Fat mass and fat-free-mass were calculated by anthropometry. The bone mineral density at lumbar and femoral neck were evaluated.

Bone mineral density at the lumbar and femoral neck was lower than in the normal population. In the non-ambulaty group, bone mineral density was ~1 SD lower than in the ambulatory one (*p* <0.01). Fat mass was greater than expected but without significantly differences between walking group (mean 26%) and wheel-chair users (25%). Patients practised sport activity had a better bone mineral density and body fat compared with other patients with the same disability.

Patients with myelomeningocele have decreased bone mineral density and are at higher risk of pathologic bone fractures. All subjects showed an excess of fat as percentage of body weight and are shorter than normal children. The measurement of bone mineral density may help to identify those patients at greatest risk of suffering of multiple fractures. Walk ability and sport activity, associated with the development of muscle mass, are important factors in promoting bone and body growth, to reduce the risk of obesity and of pathological fractures.

Key Words:

Myelomeningocele, Body composition, Bone mineral density, Osteoporosis.

Introduction

Myelomeningocele (MMC) is a congenital malformation of the neural tube, and accounts for as many as 4.7 in 10,000 live births[1]. Before 1960, as few as 10% of these infants survived, most dying from infection and/or hydrocephalus[2]. Today, after shunt improvements and more aggressive treatments, at least 60%-80% of patients born with MMC will survive into adulthood. The main clinical problems are difficulty or inability to stand, walk, voluntarily control of bladder and bowel functions. Often they have lower extremity sensory and motor deficits that impair a normal ambulation and lead to variable degrees of physical inactivity, osteoporosis, and development of pathologic fractures[3,4]. Despite this latter risk, little is known about the body composition, bone mineral density (BMD) and its relationship with deambulation and fractures. Neurological level of spinal cord involvement and ambulatory status, may affect bone mineral density of both upper and lower extremities as measured by ^{125}I photon absorptiometry of the distal radius, tibia, and first metatarsal, respectively[5]. In MMC patients, the body composition is different in the upper and lower body. The portion of the body above the neurological lesion is normal in strength, dexterity and skin sensitivity. With a proper training, the upper body may compensate the paralysis of the lower extremities. Body composition, therefore, plays an important role in the functional ability of these patients. It has been reported that the amount of muscle mass in a child with MMC is lower than normal, due to the complete or partial enervation of mus-

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cles below the neurological lesion. Such enervation results in a decreased physical activity and a subsequent weight gain[6]. After the age of four, body cell mass and lean mass tissue become increasingly replaced by adipose tissue. Moreover, the percentage of body fat is significantly greater in those with high and mid-lesions, and in non-ambulatory patients. Obesity can cause additional problems, including physical and psychological morbidity, ambulatory deterioration, transference difficulties, higher incidence of pressure sores, difficulty with hygiene and stomal care, and more complications after surgery[6-7]. Dietary habits and caloric intakes should be evaluated in children with MMC whose weight exceeds the 25th percentile for age[8]. Positive effects of physical activity on fat mass (FM) and bone mineral density (BMD) in normal subjects and in those with disability, are reported in many studies[9-11]. Sport activity improves lean body mass and reduces fat mass.

The aim of this study was to examined and compared body composition and bone mineral density measurements between different subgroups of patients with MMC.

Materials and Methods

Patients

Because known bone mineral density standards in children are available for the age group 6 to 19 yrs, of approximately 350 patients referred to the Spina Bifida Center of the Catholic University of Rome, we enrolled 64 patients with MMC aged between 5 and 14 yrs. Four patients were excluded because their parents didn't give permission. Therefore, we enrolled 60 consecutive patients (mean age 10.4 ranging form 5 to 14 yrs, 34 males and 26 females). Informed consent was obtained from all parents or guardians. Before examination, we acquired personal data by asking each patient or parent, if the patient was younger than 10 years, to fill in a case form. Questions specifically concerned the following issues: educational level (e.g., school frequency, presence of assistant teacher for handicapped), urological aspects (incontinence, urethral catheterization, self-made catheterization, urine loss between the catheterizations, pharmacological therapy, infections) and physical activity (if they performed physiotherapy and/or sport activity, and how many hours during the week). Medullar and brain magnetic resonance imaging (MRI) were acquired (e.g., site and type of lesion). Ventriculo-peritoneal shunt implantation was performed on all children. Detailed clinical history, careful clinical examination (from the neurological and physical rehabilitation point of view) were always performed (e.g., tendon reflexes of the four limbs, cutaneous sensitivity, muscle strength assessed according to British Medical Research Council scale, joint function, trophysm, dysmetria). The level of spinal cord lesion was: 10 thoracolumbar, 35 lumbosacral and 15 sacral. The ambulatory status of individuals within this group varied greatly and was categorized into two groups, defined as follows: (1) non-ambulator: full-time wheelchair; (2) full-time ambulator: individual who walks with or without tutorial aid. Of the studied patients, 38 were in the first group and 22 in the second one. Regarding sport activity we observed: 28 subjects practised sport activity. Anthropometry was performed using standard techniques (measurement of skinfolds by a calliper, of waist, hip, upper and middle thigh). Body fat as percentage of body weight was calculated from a 4-skinfold thickness using Brook's method.

Bone Densitometry Measurements

The Bone Mineral Content (BMC) and BMD at the lumbar spine and femoral neck levels were performed in all patients by DEXA using a Hologic QDR 2000 analyser (Hologic, Waltham, MA, USA). Data were expressed as deviation or Z-score to allow comparison with age and gender. For reference values of BMD we used data from Zanchetta et al[12].

Statistics

Z-scores were calculated for bone mineral density measurements via the following formula: Z-score = (patient's measurement - population mean)/standard deviation (SD). For the statistical analysis was used the software STAT-OF (OK-USA). Mann-Whitney *U* tests were used to compare two independent samples with a skewed distribution. Spearman's correlation coefficient was calculated to test the association between variables with a non-normal distribution. A p value <0.05 was deemed to be significant.

Results

Characteristics of sample are resumed in Table I.

Table I. Characteristics of sample (SD: Standard Deviation; L; Lumbar; LS: Lumbosacral; S: Sacral).

Patients	Number
Gender (M/F)	34/26
Age (mean ± SD)	10.4 ± 4.1
Site of lesion (L/LS/S)	10/35/15
Physical ability (walking/non-walking)	22/38
Sport activity (Practised/non practised)	38/22
Weight percentile (mean)	42
Height percentile (mean)	17
Lumbar Z-score (mean ± SD)	-0.8 ± 0.3
Neck Z-score (mean ± SD)	-1.4 ± 0.4

Weigth, Height and Body Fat

A scatter g of weight and height showed that these subjects were underweight (mean 42th percentile) and underheight (mean 17th percentile) compared with reference data (Table I). The reduction of weight and height was significantly (p<0.01) evident in the non-walking group (38th and 10th percentile respectively). Body fat as percentage of body weight was greater than expected, ranging from 13 to 42%, with no differences between the ambulatory group (26%) and non-ambulatory one (25%) (Table II).

Bone Mineral Density (BMD)

Mean Z-score of bone mineral density for patients is shown in Table I. Lumbar Z-score of patients was lower than that in normal sample (-0.8 ± 0.3; p<0.08). Similarly neck Z-score was significantly lower in MMC patients (-1.4 ± 0.4; p<0.01).

BMD and Walking Ability

BMD both in normal children and in patients increases with age. We examined the relationship of BMD respect age in boys and girls with MMC separately, but not differences were observed. Regarding walking ability we observed that lumbar and neck Z- scores were significantly lower in non-walking group (mean lumbar Z-score: -1.02 ± 0.89 vs -0.36 ± 0.75; p<0.01; mean neck Z-score: -1.71 ± 0.66 vs -0.69 ± 0.55; p<0.001) (Figure 1).

BMD and Sport Activity

BMD at lumbar and neck sites was better in patients who practised sport activity in comparison with who did not. In fact, in walking and sport group we observed a better BMD respect only walking group (mean lumbar Z-score: -0.30 ± 0.18 vs -0.66 ± 0.25); similarly results were found at neck site (-0.61 ± 0.36 vs -0.88 ± 0.32 respectively). Also in non-walking group we observed better BMD at neck and lumbar sites in patients practised sport activity (mean lumbar z-score: -1.02 ± 0.52 vs -1.36 ± 0.66; mean neck z-score: -1.55 ± 0.42 vs -1.89 ± 0.61) (Figure 2).

Body Fat and Sport Activity

Correlation between body fat (as percentage of body weight) and sport activity, showed better results in patients practised sport activity respect other patients with the same disability (21% in patients walking and practising sport respect 27% in only walking patients; 22% in non walking and practising sport respect 26% in the sample of only non-walking) (Figure 3).

Discussion

In this study we assessed BMD and body composition in children with MMC and correlations with walking ability and sport activity. Literature evidenced that patients with MMC had higher risk of osteoporosis and pathologic bone fractures[13-16]. James[17] reported 44 lower extremity fractures occurring in 22 of 122 patients, and Drennan and Freehafer[18] reported 58 fractures occurring in 25 of 84 children[8]. All Authors felt that disuse of the limbs and overall physical inactivity contributed greatly to the increased risk

Table II. Characteristics of walking and nonwalking groups (°p<0.01; *p< 0.01; **p<0.001).

	Weight percentile°	Height percentile°	% Body fat	Lumbar Z-score*	Neck Z-score**
Walking	55th	28th	26th	-0.36 ± 0.75	-0.69 ± 0.55
Non walking	39th	10th	25th	-1.02 ± 0.89	-1.71 ± 0.66

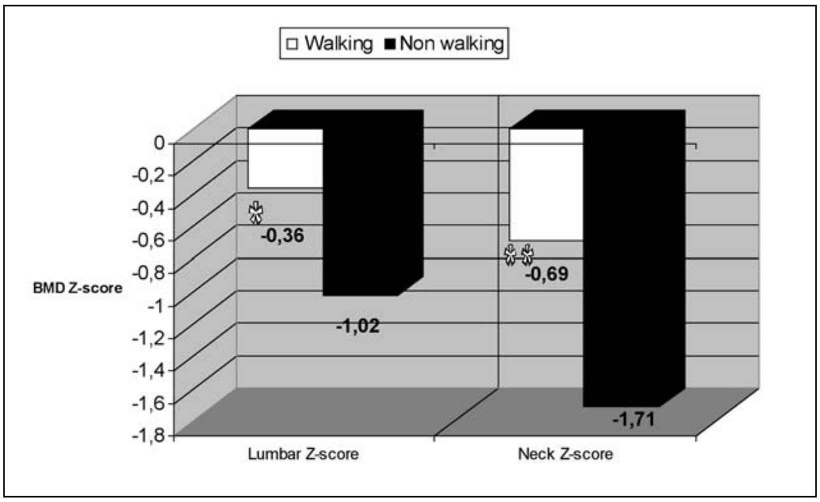

Figure 1. Correlation between BMD at lumbar and femoral sites and walking ability (*p<0.01; **p<0.001).

of bone fractures. In our children population with MMC, we did not have any lower extremity fractures but we evidenced a significant bone mineral density reduction and consequently an higher risk of osteoporosis and pathologic fractures. Many Authors reported that BMD has been correlated with bone strength and incidence of fractures[12-19]. Our data agree with these studies. In fact, they showed that BMD reduction is more evident at neck site where higher is the risk of pathologic fractures. Level lesion, because is the main cause of walking ability, is a very important cofactor for bone density and for risk of fractures. We have observed different BMD in our subgroups: the non-ambulator group (full-time wheelchair) had worse BMD respect the full-time ambulator (individual who walked autonomously or with tutorial aid). Due to loss of motor function, adolescents with MMC were restricted in their everyday activities, which led to an increasingly sedentary life style. This, in turn, leds to a decrease in physical fitness and an increase in body fat. Conversely, MMC children who practised sport activity had a higher BMD and a better Fat Mass respect patients with the same degree of disability. Deficits in lean muscle tissue in children with motor impairments were associated with a high prevalence of obesity and suboptimal physical fitness. Obesity was 2.5 times more evident among individuals with lower extremity disabilities than among those without any disability[20,21]. Obesity exacerbates problems associated with disability, such as limited ambulation, difficulty in transferring, risk of pressure sores, psychological morbidity, and possible

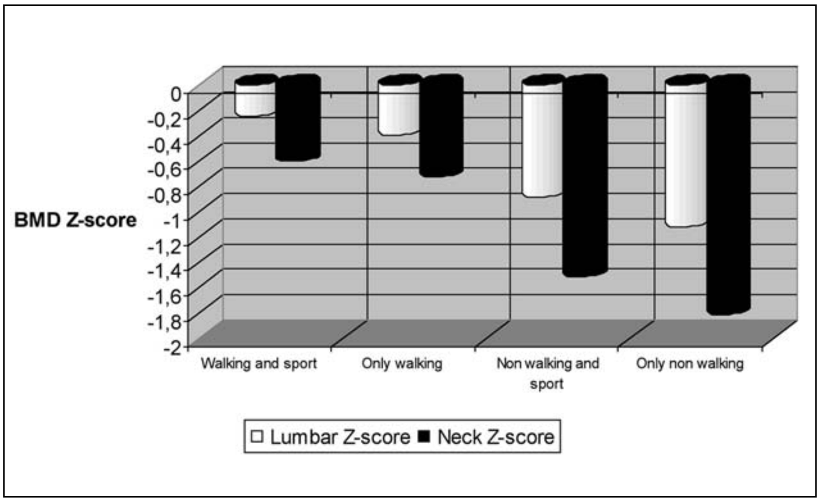

Figure 2. Correlation between BMD at lumbar and femoral sites, walking ability and sport activity.

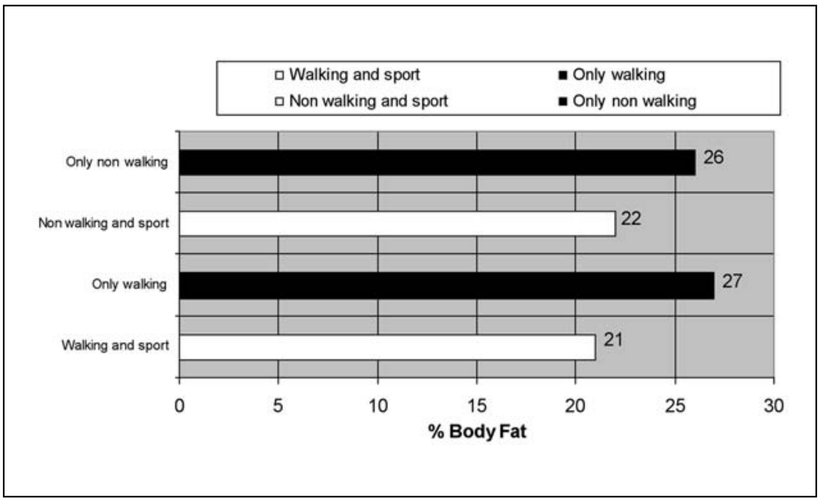

Figure 3. Correlation between % body fat (as percentage of body weight), walking ability and sport activity.

complications during surgery. Several studies have found that risk of morbidity from coronary disease and atherosclerosis was increased among men and women who had been overweight adolescents[7,22-24]. Recent scientific information suggests that the most effective interventions to address excess weight and obesity in children should include behavioural modification and education to improve eating habits and increase energy expenditure[25-30]. Roberts et al.[6] reported that in MMC after the age of four, body cell mass and lean mass tissue become increasingly replaced by adipose tissue. The percentage of body fat was significantly greater in those with high-lesions and mid-lesions, and in those in the non-ambulatory group. Shurtleff et al.[29] concluded that body fat composes a greater proportion of body mass in subjects with MMC in comparison with controls and that excess adipose tissue bears a statistical relationship to the degree of mobility and the lesion level. Dietary habits and caloric intakes should be evaluated in patients whose weight exceeds the 25th percentile for age. The effects of sedentary lifestyle and physical inactivity on bone fractures and BMD have been examined in several studies. James[17] found that in the asymmetrically affected patients, the nonactive or paralytic limb was more likely at risk for fracture compared with the active limb. Lin et al.[14] examined hemiplegic patients with cerebral palsy and found that BMC and BMD of the nonactive limb were 5.6% and 21% lower than measures for the active limb[15]. The present study confirms that MMC patients has an higher risk for obesity and osteoporosis but shows that is possible during childhood, to improve quality of life reducing risk of coronary disease, atherosclerosis and pathologic fractures, with active lifestyle and particularly practising sport. Sport activity improves lean mass, reduces fat mass and causes behavioural modification and education of eating habits.

In conclusion, patients with MMC have an higher risk of pathologic fractures due to osteoporosis. We suggest to perform BMD measurement in each child with MMC particularly in those with higher level of lesion and non-walking, particularly at high risk for pathologic bone fractures. To reduce the risk of obesity and pathologic fractures, the patients may practise sport activity which reducing fat mass and improving bone density, self esteem and quality of life.

References

1) Botto L, Moore CA, Khoury MJ, Erickson JD. Neural tube defects. New Engl J Med 1999; 341: 1509-1519.

2) Shurtleff D. Myelodysplasia: problems of long-term survival and social function. Western J Med 1975; 122: 199-205.

3) Anshuetz RH, Freehafer AA, Shaffer JW, Dixon MS Jr. Severe fracture complications in myelodysplasia. J Pediatr Orthop 1984; 4: 22-24.

4) Valtonen KM, Goksör LA, Jonsson O, Mellström D, Alaranta HT, Viikari-Juntura ER. Osteoporosis in adults with meningomyelocele: an unrecognized problem at rehabilitation clinics. Arch Phys Med Rehabil 2006; 87: 376-382.

5) Rosenstein BD, Greene WB, Herrington RT, Blum AS. Bone density in myelomeningocele: the effects of ambulatory status and other factors. Dev Med Child Neurol 1987; 29: 486-494.

6) Roberts D, Shephard RW, Shephard K. Anthropometry and obesity in myelomeningocele. J Paediatr Child Health 1998; 27: 83-90.

7) Yekutiel M, Brooks ME, Ohry A, Yarom J, Carel R. The prevalence of hypertension, ischaemic heart diseases in spinal cord injured patients and amputees. Paraplegia 1989; 27: 58-62.

8) Bandini LG, Schoeller DA, Fukagawa NK, Wykes LF, Dietz WH. Body composition and energy expenditure in adolescents with cerebral palsy or myelodysplasia. Pediatric Res 1991; 29: 70-77.

9) Ekvall SW. Myelomeningocele. In: Ekvall S, (ed): "Pediatric Nutrition in Chronic Diseases and Developmental Disorders". New York: Oxford University Press, pp 107-113, 1993.

10) Linden C, Alwis G, Ahlborg H, Gardsell P, Valdimarsson O, Stenevi S, Besjakov J, Karisson MK. Exercise, bone mass and bone size in prebubertal boys: one-year data from the pediatric osteoporosis prevention study. Scand J Med Sci Sports 2007; 17: 340-347.

11) Chad KE, McKay HA, Zello GA, Bailey DA, Faulkner RA, Snyder RE. Body composition in nutritionally ambulatory and non-ambulatory children with cerebral palsy and healthy reference group. Dev Med Child Neurol 2000; 42: 334-339.

12) Zanchetta JR, Plotkin H, Alvarez FML. Bone mass in children: normative values for 2-20 year-old population. Bone 1995; 16(suppl): 393S-399S.

13) Chan GM, Hess M, Hollis J, Book LS. Bone mineral status in childhood accidental fractures. Am J Disease Child 1984; 138: 569-570.

14) Lin PP, Henderson RC. Bone mineralization in the affected extremities of children with spastic hemiplegia. Dev Med Child Neurol 1996; 38: 782-786.

15) Henderson RC, Lin PP, Greene WB. Bone mineral density in children and adolescents who have spastic cerebral palsy. J Bone Joint Surg 1995; 77: 1671-1681.

16) Quan A, Adams R, Ekmark E, Baum M. Bone mineral density in children with myelomeningocele. Pediatrics 1998; 102: 232-236.

17) James CC. Fractures of the lower limbs in spina bifida cystica: a survey of 44 fractures in 122 children. Dev Med Child Neurol Suppl 1970; 22(Suppl 22): 88.

18) Drennan JC, Freehafer AA. Fractures of the lower extremities in paraplegic children. Clin Orthop Relat Res 1971; 77: 211-217.

19) Dosa NP, Eckrich M, Katz DA, Turk M, Liptak GS. Incidence, prevalence, and characteristics of fractures in children, adolescents, and adults with spina bifida. J Spinal Cord Med 2007; 30(Suppl 1): S5-S9.

20) Troiano RP, Flegal KM, Kuczmarski RJ, Campbell SM, Johnson CL. Overweight prevalence and trends for children and adolescents: the National Health and Nutrition Examination Surveys, 1963 to 1991. Arch PediatrAdol Med 1995; 149: 1085-1091.

21) Weil E, Wachterman M, McCarthy EP, Davis RB, O'Day B, Iezzoni LI, Wee CC. Obesity among adults with disabling conditions. JAMA 2002; 288: 1265-1268.

22) Bauman WA, Spungen AM. Carbohydrate and lipid metabolism in chronic spinal cord injury. J Spinal Cord Med 2001; 24: 266-277.

23) Imai K, Kadowaki T, Aizawa Y, Fukutomi K. Morbidity rates of complications in persons with spinal cord injury according to the site of injury and with special reference to hypertension. Paraplegia 1994; 32: 246-252.

24) Rendeli C, Ausili E, Castorina M, Antuzzi D, Tabacco F, Caldarelli M. Homocysteine, folate, lipid profile and MTHFR genotype and disability in children with myelomeningocele. Child Nerv Syst 2006; 22: 1316-1321.

25) Block P, Skeels SE, Keys CB, Rimmer JH. Shake-It-Up: health promotion and capacity building for people with spinal cord injuries and related neurological disabilities. Disabil Rehabil 2005; 27: 185-190.

26) Bar-Or O. Obesity: the optimal exercise program. In: Godberg E, Ed. Sports and Exercise for Children With Chronic Health Conditions. Champaign, IL: Human Kinetics; 1995.

27) Bar-Or O, Foreyt J, Bouchard C, Brownell KD, Dietz WH, Ravussin E, Salbe AD, Schwenger S, St Jeor S, Torun B. Physical activity, genetic, and nutritional considerations in childhood weight management. Med Sci Sport Exerc 1998; 30: 2-10.

28) Sothern MS, Schumacher H, von Almen TK, Carlisle LK, Udall JN. Committed to kids: an integrated, four-level approach to weight management in adolescents. J Am Diet Assoc 2002; 102: S81-S85.

29) Shurtleff DB, Lamers J, Goiney T, Gordon L. Are myelodysplastic children fat? Anthropometric measures: a preliminary report. Spina Bifida International 1982; 4: 1-21.

30) Catherine B, Grogan M, Shirley M, Ekvall P. Body Composition of children with myelomeningocele, Determined by 40K, urinary creatinine and anthropometric measures. J Am Coll Nutr 1999; 18: 316-323.