Metabolic Syndrome in Children With Myelomeningocele and the Role of Physical Activity: A Narrative Review of the Literature

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Objectives: The purpose of this review is to describe the current scientific literature on the prevalence of metabolic syndrome in children with myelomeningocele and to gain insight into the baseline levels of aerobic fitness, endurance, and strength in this population in order to identify gaps in knowledge, suggest potential primary prevention strategies, and provide recommendations for future studies. **Methods:** A literature review of articles published in English and French between 1990 and April 2020 was conducted. **Results:** Obese adolescents with myelomeningocele have an increased prevalence of components of the metabolic syndrome. Children and adolescents with myelomeningocele have decreased aerobic fitness and muscular strength, decreased lean mass, and increased fat mass, all of which, when combined with higher levels of physical inactivity, put them at higher risk of developing metabolic syndrome and cardiovascular diseases. **Conclusion:** Until more research is conducted, addressing weight-related challenges and promoting healthy habits (such as optimal activity levels) could be easily integrated into yearly myelomeningocele clinics. An actionable suggestion might be to systematically weigh and measure children in these clinics and utilize the results and trends as a talking point with the parents and children. The follow-up appointments could also be used to develop physical activity goals and monitor progress. We recommend that the health care practitioner tasked with this intervention (physician, nurse, etc.) should be aware of locally available accessible sports platforms and have knowledge of motivational interviewing to facilitate removal of perceived barriers to physical activity. **Key words:** *metabolic syndrome, myelomeningocele, pediatric, physical activity, spina bifida, youth*

Introduction

Spina bifida (SB) is a congenital disorder resulting from the defective closure of the neural tube during the fourth week of gestation.[1] It refers to a group of developmental defects of the spinal column ranging from spina bifida occulta, a very common condition without any impact on function, to myelomeningocele (MMC), the most severe form of SB.[2] In MMC, the spinal cord and meninges protrude posteriorly through incompletely closed vertebrae and form a sac, which may or may not be covered by skin, on the infant's back.[3] Its incidence is approximately 0.20 to 0.40 per 1000 births per year in Canada and the United States.[4]

MMC results in varying degrees of impairments depending on the level and severity of the lesion. Patients may exhibit sensory and motor deficits below the level of the lesion ranging from weakness to paralysis, spasticity or hypotonia, bowel and bladder dysfunction, Chiari II malformation with associated hydrocephalus, neuroendocrine disorders, and cognitive impairments as well as orthopedic malformations such as neurogenic club foot, scoliosis, kyphosis, contractures, and hip dislocations, thus requiring extensive multidisciplinary medical care.[1] With current medical progress, the life expectancy of people with MMC has increased.[5] A new approach in management of these patients is therefore required, not only focusing on the disorder itself but also on health promotion and primary prevention to ensure optimal health and well-being. Indeed, due to risk factors inherent to their condition, the prevalence of obesity in children and adolescents with MMC has been estimated to be up to 64%, approximately twice that of their neurotypical

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peers.[6-8] Furthermore, whereas childhood obesity is a worldwide issue, children with physical disabilities have been excluded from research focusing on health promotion challenges and exercise recommendations.[9] This paucity of research may explain why although dietary and physical activity interventions are recommended by health care providers, assistance with implementation for children and adolescents with MMC is provided inconsistently.[10-12] Because there is strong evidence that childhood lifestyle habits often continue through adulthood, it is crucial to identify and address these habits early.[13]

Accordingly, the aim of this narrative review is to describe the current scientific literature on the prevalence of metabolic syndrome in children with MMC and to gain insight into the baseline levels of aerobic fitness, endurance, and strength in this population in order to identify gaps in knowledge, suggest potential primary prevention strategies, such as physical activity, and provide recommendations for future studies.

Methods

A literature review of articles published in English and French was conducted. The strategy used for this review included searching PubMed and Google Scholar as well as cross-referencing citations of published papers. Search terms included metabolic syndrome, metabolic syndrome disease, metabolic syndrome disorder, obesity, aerobic fitness, endurance, strength, and physical activity, where each term was combined with spina bifida and myelomeningocele. Terms were searched as key words and, when available, as MeSH terms. The search was limited to literature published between 1990 and April 2020. From the initial 301 records identified and screened, a total of 36 studies were included in this review. Studies were included if they included children or adolescents with myelomeningocele and assessed metabolic syndrome components (obesity, dyslipidemia, blood pressure, hyperglycemia, and insulin resistance) and/or physical fitness (including at least one of the following parameters: aerobic fitness, cardiorespiratory endurance, muscle strength, or body composition). Due to the scarcity of literature specific to pediatric MMC, some literature on children with the broader term SB, adults with MMC, pediatric spinal cord injury (SCI), and pediatrics in general was included to provide further insight into the possible effect of metabolic syndrome and the role of exercise. Although we sought to gain insight on MMC, occasionally studies mentioned only the broader undifferentiated term SB; we chose to include these studies when it was clear that the patients had clinical presentations more consistent with MMC than SB occulta.

Results

All studies identified, their methods, protocols used to evaluate outcome measures, and their results are presented in **Tables 1** and **2**.

Metabolic syndrome

Metabolic syndrome is defined as having three or more risk factors amongst obesity, elevated blood pressure, elevated triglycerides, decreased high-density lipoprotein cholesterol, and hyperglycemia.[14] Adults with three or more criteria of metabolic syndrome are three times more at risk of developing cardiovascular disease and five times more at risk of developing diabetes and other comorbidities.[15] Furthermore, each additional risk factor increases the risk for cardiovascular disease mortality.[16-18] A study done by Nelson et al.[15] concluded that, similar to adults with SCI, obese adolescents with spinal cord dysfunction have an increased prevalence of metabolic syndrome components. Thus, implementing positive health behaviors early in life is crucial as obese children have a 2.9 times higher risk of developing metabolic syndrome during adulthood,[18,19] further increasing their risk of having a cardiovascular disease.[16-18]

For the metabolic syndrome components, **Table 1** provides a breakdown of each study's design, participant description, assessment tools and protocols, outcome measures, and results.

Obesity

Obesity is the result of chronic positive energy balance, where total energy intakes exceeds total energy expenditure.[15,20] In children with MMC, obesity ranges from 28% to 50% and from 34% to 82.4% among adolescents and adults, as assessed by

Table 1. Metabolic syndrome components in patients with myelomeningocele

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Roberts et al. (1991)[33]	Cross-sectional, outpatient (Australia) 110 subjects with MMC 0.3-27 yo; 45% male	Thickness of 4 skinfolds (biceps, triceps, subscapular, suprailiac)	Body fat	All neural groups had %BF greater than expected ($.001<p<.05$); %BF ranging from 24.4% to 25.8% using skinfold measurements
Mita et al. (1993)[22]	Cross-sectional 35 subjects with SB 2.9-16.7 yo; 49% male 129 control subjects matched for age 4.0-15.11 yo; 50% male	Thickness of 2 skinfolds (right mid-triceps and subscapular)	Body fat	%body fat > when higher level neurological involvement: high, 26.1 ± 5.0%; mid, 23.0 ± 6.3%; low, 20.4 ± 8.6% (difference nonsignificant) **Community ambulators**: 37.6 ± 8.4% **Noncommunity ambulators**: 23.1 ± 6.2%; $p < .1$
Fiore et al. (1998)[29]	Cross-sectional 100 subjects with MMC 0.5-19 yo (median 7.6 yo); 54% male	BMI	Body fat	20% > 75th percentile 40% frank overweight
Grogan & Ekvall (1999)[30]	Cross-sectional, outpatient (USA) 14 subjects with MMC 6-16 yo; 29% male	Thickness of 3 skinfolds (triceps, scapula, and thorax)	Body fat	Sum of skinfolds: 64 + 30.4 mm
van den Berg-Emons et al. (2003)[21]	Cross-sectional (Netherlands) 14 subjects with MMC 14-26 yo; 57% male	Thickness of 4 skinfolds (biceps, triceps, subscapular, suprailiac)	Body fat	All participants: 23.1 ± 7.1% **Ambulatory** 22.3 ± 8.1% **Nonambulatory** 25.0 ± 4.4%; $p = .60$
Rendeli et al. (2004)[59]	Cross-sectional (Italy) 81 subjects with SB 1-16 yo; 56% male 150 controls 1-16 yo; 60% male	BMI HDL, TG, LDL VLDL, TC	Body fat Dyslipidemia	**SB group**: 18.9 ± 2.2 kg/m^2 **Control group**: 20.9 ± 1.2 kg/m^2 No sig. difference for TC, HDL, LDL, VLDL, or TG serum levels In MMC nonwalking girls, higher levels of VLDL ($p = .04$) TC serum levels were sig. higher ($p = .01$) in MMC nonwalking girls

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Table 1. Metabolic syndrome components in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Nelson et al. (2007)[15]	Cross-sectional, outpatient (USA) 34 subjects with SB 16.3 ± 2.5 yo 52.9% male 20 subjects with SCI 16.9 ± 3.0 55% male 60 controls 16.2 ± 2.5 yo 45% male	DEXA BMI	Body fat	% Trunk fat **SB group:** 31.2 ± 9.6, ($p \leq$ .05 with control and SCI groups) **SCI group:** 30.9 ± 13.4 ($p \leq$ .05 with control group) **Control group:** 25.7 ± 12.8 % Body fat **SB group:** 40.0 ± 8.5 ($p \leq$ .05 with control and SCI groups) **SCI group:** 33.9 ± 12.5 ($p \leq$ .05 with control group) **Control group:** 28.0 ± 11.2 BMI z score **SB group:** 1.43±1.07 ($p \leq$ .05 with control and SCI groups) **SCI group:** 0.07 ± 2.29 ($p \leq$ .05 with control group) **Control group:** 0.73 ± 1.11
		Systolic blood pressure	Blood pressure	High blood pressure **Nonobese SB group:** 10%; **Obese SB group:** 29.2% **Nonobese SCI group:** 20%**; Obese SCI group:** 30% **Nonobese control group:** 12.2 %**; Obese control group:** 26.3 %
		Fasting blood glucose Fasting insulin HOMA-IR	Insulin-resistance	Impaired fasting blood glucose / elevated HOMA-IR **Nonobese SB group:** 0% / 0%**; Obese SB group:** 0% / 12.5% **Nonobese SCI group:** 0% / 0%**; Obese SCI group:** 0% / 60% **Nonobese control group:** 2.5% / 4.9%**; Obese control group:** 0% / 42.1%
		HDL, TG, LDL, TC	Dyslipidemia	Low HDL / High TG **Nonobese SB group:** 70% / 10%**; Obese SB group:** 75% / 33.3% **Nonobese SCI group:** 80% / 20%**; Obese SCI group:** 100% / 70% **Nonobese control group:** 56.1% / 24.4%**; Obese control group:** 84.2% / 36.8%

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Table 1. Metabolic syndrome components in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
			Metabolic syndrome	Has metabolic syndrome **Nonobese SB group:** 0%**; Obese SB group:** 45.8% **Nonobese SCI group:** 10%**; Obese SCI group:** 100% **Nonobese control group:** 2.4 %**; Obese control group:** 63.2%
Liusuwan et al. (2007)[31]	Cross-sectional, outpatient (USA) 66 subjects with SB 15.8 ± 2.6 yo 33 subjects with SCI 17.5 ± 2.2 yo 85 controls 15.9 ± 2.4 yo 31 overweight controls 15.6 ± 2.6 yo	DEXA BMI	Body fat	BMI **SB group**: 28.7 ± 6.2 kg/m^2 ($p < .05$ with control groups) **SCI group**: 19.5 ± 3.7 kg/m^2 ($p < 0.05$ with control groups) **Control group:** 20.6 ± 2.3 kg/m^2 **Overweight control group**: 33.1 ± 5.2 kg/m^2 ($p < .05$ with control group) % Body fat **SB group**: 38.9 ± 9.4% ($p < .05$ with control groups) **SCI group**: 26.7 ± 9.1% ($p < .05$ with control groups and SB group) **Control group**: 21.2 ± 7.1% **Overweight control group**: 39.0 ± 8.1% ($p < .05$ with control group)
Widman et al. (2007)[13]	Cross-sectional, outpatient (USA) 37 subjects with SB 11-21 yo; 49% male 34 healthy subjects, matched by age	DEXA	Body fat	**SB group:** Male: 36.3 ± 9.3%; Female: 46.2 ± 5.0% (SB vs. control, $p < .05$) **Control group:** Male: 16.1 ± 5.2%; Female: 25.7 ± 4.1%
Ausili et al. (2008)[23]	Cross-sectional, outpatient (Italy) 60 subjects with MMC 5-14 yo; 57% male	Thickness of 4 skinfolds (waist, hip, and upper and middle thigh)	Body fat	**Ambulatory**: 26%**; Ambulatory + sports**: 21%**;** **Ambulatory no sports**: 27% **Nonambulatory**: 25%**; Nonambulatory + sports**: 22%**;** **Nonambulatory no sports**: 26%

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Table 1. Metabolic syndrome components in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Buffart et al. (2008)[25]	Cross-sectional, outpatient (Netherlands) 31 subjects with MMC 16-30 yo; 58% male	Thickness of 4 skinfolds (biceps, triceps, subscapular, suprailiac)	Body fat	All participants: 74.4 ± 40.6 mm **Ambulatory:** 59.1 ± 33.4 mm**; Nonambulatory:** 88.6 ± 42 mm; $p = .05$ **Male:** 50.7 ± 24.2 mm; **Female:** 110.4 vs. 32.6 mm; $p < .001$
		HDL, TG, LDL, TC	Dyslipidemia	High total cholesterol / high LDL / very high LDL / low HDL / high TG **Total:** 3 / 6 / 3 / 19 / 0% **Ambulatory**: 0 / 0 / 0 / 15 / 0% **Nonambulatory**: 6/ 11 / 5/ 22 / 0% **Male**: 6 / 11 / 6 / 11 / 0% **Female**: 0 / 0 / 0 / 31 / 0%
		Blood pressure	Blood pressure	Prehypertension / stage I hypertension / stage II hypertension **Total:** 63 / 17 / 3% **Ambulatory:** 42 / 25 / 0% **Nonambulatory:** 78 / 11 / 6% **Male:** 64 / 24 / 6%; **Female:** 61 / 8 / 0%
Buffart et al. (2008)[8]	Cross-sectional, outpatient (Netherlands) 50 subjects with MMC 16-30 yo; 50% male	Thickness of 4 skinfolds (biceps, triceps, subscapular, suprailiac)	Body fat	All participants: 74.8 ± 38.8 mm, 159 ± 77% of reference values **Male:** 51.2 ± 24.6 mm, 146 ± 79% of reference values **Female:** 100.4 ± 35.1 mm, 173 ± 73% of reference values
Buffart et al. (2008)[27]	Cross-sectional, Outpatient (Netherlands) 51 subjects with MMC 16-30 yo; 51% male	Thickness of 4 skinfolds (biceps, triceps, subscapular, suprailiac)	Body fat	**Sports:** 74 ± 36.8 mm **No sports:** 75.5 ± 39.4 mm; $p = .4$
Buffart et al. (2008)[24]	Cross-sectional, outpatient (Netherlands) 51 subjects with MMC 16-30 yo; 51% male	Thickness of 4 skinfolds (biceps, triceps, subscapular, suprailiac)	Body fat	All participants: 74.4 ± 38.5 mm Increased ambulatory status had less body fat ($p = .03$) vs. less ambulatory status **Community:** 59.1 ± 29.2 mm**; Household:** 65.5 ± 32.3 mm; **No:** 86.0 ± 42.0 mm

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Table 1. Metabolic syndrome components in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Buffart et al. (2009)[26]	Cross-sectional, outpatient (Netherlands) 51 subjects with MMC 16-30 yo; 51% male	Thickness of 4 skinfolds (biceps, triceps, subscapular, suprailiac)	Body fat	All participants: 74.4 ± 38.5 mm **Community:** 59.1 ± 29.2 mm**; Household:** 65.5 ± 32.3 mm; **No:** 86.0 ± 42.0 mm
Dosa et al. (2009)[7]	Cross-sectional, outpatient (USA) 203 subjects with SB 6-58 yo (median 19 yo); 45% male	BMI	Body fat	Are obese **All**: 23%; **Male**: 18%; **Female**: 28% **6-11 yo**: 18%; **Male**: 9%; **Female**: 22% **12-19 yo**: 8%; **Male**: 6%; **Female**: 9% **20-58 yo**: 37%; **Male**: 27%; **Female**: 48% Are overweight/obese **6-11 yo: Ambulatory**: 12.5%/ 20.8%; **Nonambulatory**: 20% / 10% **12-19 yo: Ambulatory**: 22.4%/ 8.2%; **Nonambulatory**: 7.7% / 7.7% **20-58 yo: Ambulatory**: 26.7% / 31.1%; **Nonambulatory**: 20.4% / 42.9%
Mueske et al. (2015)[32]	Cross-sectional, outpatient (USA) 82 subjects with MMC 9.8 ± 2.7 yo; 55% male 119 controls 10.4 ± 2.4 yo; 55% male	DEXA BMI	Body fat	% Trunk fat **MMC group**: 29.2 ± 9.5 (p = .007)**; Male**: 27.4 ± 9.7; **Female**: 31.3 ± 9.0 Sacral 25.8 ± 9.5; Low lumbar 26.9 ± 9.1; Mid lumbar + 31.4 ± 9.2 **Control group**: 25.6 ± 9.0; **Male**: 24.7 ± 9.7; **Female**: 26.6 ± 8.1 % Total body fat **MMC group**: 35.2 ± 9.0 (p < .001)**; Male**: 33.5 ± 9.5; **Female**: 37.3 ± 8.0 Sacral: 31.5 ± 8.5; Low lumbar: 32.5 ± 8.0; Mid lumbar+**:** 37.7 ± 8.7 **Control group**: 29.8 ± 8.4**; Male**: 28.7 ± 9.5; **Female**: 31.1 ± 6.9 BMI (kg/m^2) **MMC group**: 21.0 ± 6.1 Sacral: 19.4 ± 5.2; Low lumbar: 18.4 ± 3.1; Mid lumbar+: 22.5 ± 6.7 **Control group**: 20.0 ± 4.7

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Table 1. Metabolic syndrome components in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Van Speybroeck et al. (2017)[37]	Cross-sectional, outpatient 28 subjects with MMC 10.0 ± 2.1; 61% male 58 controls 10.4 ± 2.4 yo; 52% male	DEXA BMI	Body fat	BMI (kg/m^2) **MMC group**: 20.9 ± 8.0 (p = .73) Sacral: 16.4 ± 5.8; Low lumbar: 19.6 ± 7.9; Mid lumbar: 21.3 ± 6.3 **Control**: 18.3 ± 7.3 % Trunk fat **MMC group:** 29.3 ± 10.3% (p = .12) Sacral: 23.6 ± 10.0%; Low lumbar: 27.4 ± 12.6%; Mid lumbar**:** 33.0 ± 8.7% **Control:** 25.8 ± 9.5% % Total body fat **MMC group:** 35.2 ± 9.4% (p = .01) Sacral: 29.1 ± 9.1%; Low lumbar: 33.0 ± 11.6%; Mid lumbar: 39.2 ± 7.2% **Control**: 29.9 ± 8.8%
		HDL, TG, LDL, TC	Dyslipidemia	MMC had lower levels of HDL (p = .03); did not differ in TC, TG, or LDL ($p \geq .26$). Levels of HDL tended to decrease with increasing neurosegmental level while levels of TG tended to increase.
		Fasting blood glucose Fasting insulin HOMA-IR	Insulin-resistance	MMC tended to higher levels of insulin (p = .10) and HOMA-IR (p = .12); not all differences reached statistical significance. Glucose levels did not differ (p = .68).

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Table 1. Metabolic syndrome components in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Crytzer et al. (2018)[28]	Cross-sectional, outpatient (USA) 29 subjects with SB Group who reached ventilatory breakpoint: 28.53 ± 13.08 yo Group who did not reach ventilatory breakpoint: 34.20 ± 11.00 yo	BMI, using arm span	Body fat	Group who reached ventilatory breakpoint: 23.34 ± 7.16 kg/m^2 Group who did not reach ventilatory breakpoint: 26.17 ± 8.34 kg/m^2
Stiles-Shields & Holmbeck (2019)[10]	Cross-sectional, outpatient (USA) 167 subjects with SB (56.9% MMC) 15-24 yo; 60.5% male	BMI	Body fat	24.87 ± 6.15 kg/m^2 MMC had higher BMIs than those with other types of SB (U = 2629, p = .02).

Note: Data are expressed in terms of mean ± *SD*. %BF = percent body fat; BMI = body mass index; DEXA = dual energy x-ray absorptiometry; HDL = high-density lipoprotein; HOMA-IR = homeostasis model assessment of insulin resistance; LDL = low-density lipoprotein; MMC = myelomeningocele; SB = spina bifida; SCI = spinal cord injury; sig. = significant; TC = total cholesterol; TG = triglycerides; VLDL = very low-density lipoprotein; yo = years old.

Table 2. Aerobic fitness, endurance, and strength in patients with myelomeningocele

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Coutts et al. (1993)[87]	Cross-sectional, outpatient 42 subjects with SB 7-18 yo; 45% male	Wingate 30 s anaerobic power test Progressive maximal oxygen uptake test on an electronically brake arm or cycle ergometer Handgrip strength	VO2 peak	7-12 yo: Male: 0.81 L/min; Female: 1.03 L/min 13-18 yo: Male: 1.47 L/min; Female: 1.05 L/min
			Maximum workload	7-12 yo: Male: 56 W; Female: 59 W 13-18 yo: Male 158 W; Female: 79 W
			HR	7-12 yo: Male: 178 bpm; Female: 174 bpm 13-18 yo: Male: 185 bpm; Female: 183 bpm
			Handgrip strength	7-12 yo: Male: 353 N; Female: 392 N 13-18 yo: Male: 686 N; Female: 471 N
Sherman et al. (1997)[66]	Cross-sectional 12 subjects with MMC 10-17 yo; 33% male 12 healthy subjects matched for age, gender, and arm span	Cardiopulmonary exercise testing	VO2 peak	**MMC group:** 13.8 ± 4.8 mL/kg/min **Control group:** 21.3 ± 7.5 mL/kg/min $p = .02$
			HR peak	**MMC group:** 153.5 ± 21.4 bpm **Control group:** 127.6 ± 22.1 bpm $p = .09$
Klimek-Piskorz & Piskorz (2002)[71]	Cross-sectional, outpatient 10 subjects with MMC 17.6 ± 0.6 yo; 100% male	Upper limb graded cycle ergometer test	VO2 peak	46.3 ± 2.2 mL/kg/min
			Maximum workload	157.5 ± 38 W
			HR peak	191 ± 6 bpm
van den Berg-Emons et al. (2003)[21]	Cross-sectional, outpatient (Netherlands) 14 subjects with MMC 14-26 yo; 57% male	Progressive maximal exercise test in an electronically braked arm or cycle ergometer	VO2 peak	All participants: 27.3 ± 7.4 mL/kg/min **Ambulatory:** 30.1 ±6.2 mL/kg/min **Nonambulatory:** 22.5 ± 7.5 mL/kg/min $p = .15$
			HR peak	All participants: 185 ± 18 bpm **Ambulatory:** 193 ±5 bpm **Nonambulatory:** 171 ± 25 bpm $p = .15$

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Table 2. Aerobic fitness, endurance, and strength in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Norrlin et al. (2003)[73]	Cross-sectional, outpatient (Sweden) 32 subjects with MMC 6-11 yo; 59% male	Isometric hand strength with hand-held dynamometry	Muscle strength	Hand grip strength **Community ambulators:** 74.5 ± 25.2 N **Household ambulators:** 62.5 ± 25.7 N **Nonfunctional ambulators:** 79.6 ± 25.2 N **Nonambulators:** 57.9 ± 13.6 N 71.03 ± 23.30% of reference values
Schoenmakers et al. (2004)[88]	Cross-sectional, outpatient (Netherlands) 30 subjects with MMC 1-17 yo; 47% male 14 subjects with LMMC 1-17 yo; 57% male	Manual muscle testing	Muscle strength	Hip flexor muscles: MMC 4.9 ± 0.5; LMMC 4.9 ± 0.3; p = 098 Hip abductor muscles: MMC 4.5 ± 0.9; LMMC 4.6 ± 0.7; p = .59 Hip extensor muscles: MMC 3.5 ± 1.5; LMMC 4.4 ± 1.1; p = .01 Knee extensor muscles: MMC 4.9 ± 0.5; LMMC 5.0 ± 0.0; p = .50 Ankle dorsal-flexor muscles: MMC 4.4 ± 1.2; LMMC 4.07 ± 1.6; p = .49 Calf muscles: MMC 2.9 ± 1.5; LMMC 4.0 ± 1.6; p = .01
Widman et al. (2007)[13]	Cross-sectional, outpatient (USA) 37 subjects with SB 11-21 yo; 49% male 34 healthy subjects, matched by age	Ramp protocol with a magnetically braked arm ergometry Peak dynamic muscle strength using LIDO dynamometer	VO2 peak	**SB group** Male: 20.6 ± 7.6 mL/kg/min; Female: 14.2 ± 4.2 mL/kg/min **Control group** Male: 30.8 ± 6.0 mL/kg/min; Female: 21.0 ± 4.8 mL/kg/min p < .05
			Maximum workload	**SB group** Male: 61.9 ± 17.9 W; Female: 48.9 ± 15.3 W **Control group** Male: 3.0 ± 21.0 W; Female: 56.4 ± 11.4 W SB male vs. control male; p < .05
			HR peak	**SB group** Male: 163.4 ± 18.7 bpm; Female: 167.3 ±19.3 bpm **Control group** Male: 169.7 ± 24.2 bpm; Female:159.2 ± 20.2 bpm

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Table 2. Aerobic fitness, endurance, and strength in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
			Muscle strength	Shoulder flexion strength **SB group** Male: 0.54 ± 0.14 N-m/kg; Female: 0.46 ± 0.14 N-m/kg **Control group** Male: 0.70 ± 0.14 N-m/kg; Female: 0.51 ± 0.07 N-m/kg SB male vs. control male; $p < .05$ Shoulder extension strength **SB group** Male: 0.62 ± 0.14 N-m/kg; Female: 0.52 ± 0.18 N-m/kg **Control group** Male: 0.94 ± 0.19 N-m/kg; Female: 0.67 ± 0.14 N-m/kg ($p < .05$) Elbow flexion strength **SB group** Male: 0.58 ± 0.20 N-m/kg; Female: 0.38 ± 0.11 N-m/kg **Control group** Male: 0.61 ± 0.15 N-m/kg; Female: 0.37 ± 0.05 N-m/kg Elbow extension strength **SB group** Male: 0.56 ± 0.14 N-m/kg; Female: 0.43 ± 0.12 N-m/kg **Control group** Male: 0.57 ± 0.10 N-m/kg; Female: 0.45 ± 0.09 N-m/kg
Bruinings et al. (2007)[67]	Cross-sectional, outpatient (Netherlands) 18 subjects with MMC 16-30 yo; 2% male 18 healthy subjects matched for age and gender	Progressive maximal exercise test on an electronically braked arm or cycle ergometer	VO2 peak	**Ambulatory:** 31.4 ± 8.3 mL/kg/min, 34% lower than healthy subjects ($p = .02$) **Nonambulatory:** 23.5 ± 5.33 mL/kg/min, 54% lower than healthy subjects ($p < .001$)

(continues)

Table 2. Aerobic fitness, endurance, and strength in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Buffart et al (2008)[25]	Cross-sectional, outpatient (Netherlands) 31 subjects with MMC 16-30 yo; 58% male	Progressive maximal exercise test in an electronically braked arm or cycle ergometer	VO2 peak	All participants: 1.47 ± 0.51 L/min **Ambulatory:** 1.62 ± 0.59 L/min **Nonambulatory:** 1.36 ± 0.45 L/min $p = .18$ Male: 1.76 ± 0.47 L/min; Female: 1.07 ± 0.27 L/min $p = .001$
Buffart et al (2008)[8]	Cross-sectional, outpatient (Netherlands) 50 subjects with MMC 16-30 yo; 50% male	Progressive maximal exercise test on an electronically braked arm or cycle ergometer Strength of hip flexors, knee extensors, shoulder abductors, and elbow extensors through the "Break" Testing Method with hand-held dynamometry	VO2 peak	All participants: 22.6 ± 8.2 mL/kg/min; 67 ± 15% of reference values **Male:** 1.78 ± 0.51 L/min, 71 ± 13% of reference values **Female:** 1.18 ± 0.30 L/min, 61 ± 18% of reference values $p < .001$ **Community ambulators:** 29.0 ± 7.7 mL/kg/min **Household ambulators:** 22.3 ± 6.6 mL/kg/min **Nonambulators:** 19.2 ± 6.8 mL/kg/min $p < .001$
			Maximum workload	All participants: 91 ± 42 W **Male:** 113 ± 43W; **Female:** 69 ± 28 W **Community ambulators:** 123 ± 42 W **Household ambulators:** 97 ± 35 W **Nonambulators:** 73 ± 34 W
			HR peak	All participants: 174 ± 19 bpm, 90 ± 10% predicted maximum **Male:** 179 ± 16 bpm, 92 ± 8% of predicted maximum **Female:** 169 ± 20 bpm, 89 ± 10% of predicted maximum **Community ambulators:** 173 ± 21 bpm, 87 ± 10% of predicted maximum **Household ambulators:** 183 ± 14 bpm, 95 ± 8% of predicted maximum **Nonambulators** :172 ± 18 bpm, 91 ± 10% of predicted maximum

(continues)

Table 2. Aerobic fitness, endurance, and strength in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
			Muscle strength	Subnormal muscle strength 61% participants **Community ambulators**: 79% **Ambulators**: 57% **Nonambulators**: 54% **Male:** 58%; **Female:** 64%
Buffart et al (2008)[27]	Cross-sectional, outpatient (Netherlands) 51 subjects with MMC 16-30 yo; 51% male	Progressive maximal exercise test on an electronically braked arm or cycle ergometer	VO2 peak	**Participating in sports:** 1.58 ± 0.53 L/min **No participation in sports:** 1.27 ± 0.5 L/min $p = .13$
		"Break" Testing Method with hand-held dynamometry (hip flexors, knee extensors, shoulder abductors and elbow extensors)	Muscle strength	**Sports:** 52% with subnormal muscle strength **No sports** :81% with subnormal muscle strength $p = .08$
Buffart et al (2008)[24]	Cross-sectional, outpatient (Netherlands) 51 subjects with MMC 16-30 yo; 51% male	Progressive maximal exercise test on an electronically braked arm or cycle ergometer	VO2 peak	All participants :22.6 ± 8.2 mL/kg/min **Community ambulators:** 29 ± 7.7 mL/kg/min **Household ambulators:** 22.3 ± 6.6 mL/kg/min **Nonambulators:** 19.2 ± 6.8 mL/kg/min Average VO2 peak 42% lower than normative values (1.48 ± 0.522 mL/kg/min vs. 2.56 ± 0.412 mL/kg/min, respectively) Persons with a higher level of ambulatory status had higher VO2 peak (1.85 ± 0.572 mL/kg/min vs. 1.29 ± 0.402 mL/kg/min; $p < .001$)
Buffart et al (2009)[26]	Cross-sectional, Outpatient (Netherlands) 51 subjects with MMC 16-30 yo; 51% male	Progressive maximal exercise test on an electronically braked arm or cycle ergometer	VO2 peak	All participants: 1.48 ± 0.52 L/min **Community ambulators:** 1.85 ± 0.57 L/min **Household ambulators:** 1.44 ± 0.45 L/min **Nonambulators:** 1.29 ± 0.40 L/min
Danielsson et al (2008)[89]	Cross-sectional, outpatient (Sweden) 38 subjects with MMC 3.8-16.8 yo 53% male	Manual muscle testing	Muscle strength	Knee extensors muscle strength 36.8% graded 0-3; 63.3% graded 4-5 **Ambulators:** 100% graded 4-5 **Nonambulators:** 26.3% graded 4-5 $p < .0001$

(continues)

Table 2. Aerobic fitness, endurance, and strength in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
de Groot et al (2008)[70]	Cross-sectional, outpatient (Netherlands) 23 subjects with SB 6-17 yo; 57% males	Graded treadmill test 6MWT	VO2 peak	All participants: 33.14 mL/kg/min **Ambulators:** 34.77 mL/kg/min **Community ambulators:** 26.2 mL/kg/min; $p < .05$ 85% reached critical values
			HR peak	All participants: 172.2 ± 21.2 bpm **Ambulators:** 175.5 ± 20.8 bpm **Community ambulators:** 158.5 ± 19.1 bpm $p < .05$
			6MWD	All participants: 391.4 ± 61 m; 48.5 ± 8.3% of the predicted **Ambulators:** 408.5 ± 57.2 m; 50.2 ± 8.3% of the predicted **Community ambulators:** 333.4 ± 30.6 m; 41.1 ± 2.7% of the predicted; $p < .05$
de Groot et al (2009)[69]	Cross-sectional, outpatient (Netherlands) 20 subjects with SB 10.3 ± 4.9 yo; 45% male	Graded treadmill test 3-minute supramaximal test 6MWT	VO2 peak	All participants: 34.1 ± 8.3 mL/kg/min **Ambulators:** 39.4 ± 5.7 mL/kg/min **Community ambulators:** 28.7 ± 7 mL/kg/min $p > .05$
			VO2 supramaximal	All participants: 34.8 mL/kg/min; no significant differences with VO2 peak ($p = .274$)
			HR peak	All participants: 183.8 ± 19.9 bpm **Ambulators:** 184.7 ± 20.4 bpm **Community ambulators:** 182.3 ± 20.3 bpm $p > .05$
			6MWD	All participants: 418 ± 95 m **Ambulators:** 473 ± 45.5m **Community ambulators:** 357 ± 100 m $p < .05$

(continues)

Table 2. Aerobic fitness, endurance, and strength in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Schoenmakers et al. (2009)[68]	Cross-sectional, outpatient (Netherlands) 16 subjects with MMC 9.9 ± 3.2 yo 7 subjects with LMMC 11.6 ± 2.7 yo (total: 57% male)	Maximal exercise test in a treadmill 6MWT Manual muscle testing Isometric muscle strength	VO2 peak HR peak 6MWD Muscle strength	VO2 peak lower compared to reference values ($p < .05$) HRpeak lower compared to reference values ($p < .05$) **MMC:** 353 ± 108 m; **LMMC:** 424 ± 65 m; $p = .07$ Lower compared to reference values ($p = .03$) >25% hip abductors and the plantar flexors <3 grade <50% hip extensors = 5 grade Muscle strength of upper and lower extremity muscles lower compared to reference values ($p < .01$)
Hassan et al (2010)[72]	Cross-sectional, outpatient (Netherlands) 22 subjects with SB (15 with MMC; 7 with LMMC) 10.3 ± 3.1 yo; 59% male	6MWT	6MWD	391 ± 61 m 60.0± 9.4% of the predicted distances derived from Li et al.[90] ($p < .001$) 62.2 ± 9.4% of the predicted distances derived from Geiger et al.[91] ($p < .001$)
de Groot et al (2011)[63]	Cross-sectional, outpatient (Netherlands) 23 subjects with MMC 10.7 ± 3.5 yo; 48% male	Graded treadmill exercise test 6MWT	VO2 peak HR peak 6MWD	1.27 ± 0.6 L/min 185 ± 21.1 bpm 408 ± 94.7m
Crytzer et al (2018)[28]	Cross-sectional, outpatient (USA) 29 subjects with SB Group who reached ventilatory breakpoint: 28.53 ± 13.08 yo Group who did not reach ventilatory breakpoint: 34.20 ± 11.00 yo	Graded maximal exercise test on an electronically braked arm ergometer	Ventilatory breakpoint, %VO2 peak	Group who reached ventilatory breakpoint: 61.76 ± 16.26

(continues)

Table 2. Aerobic fitness, endurance, and strength in patients with myelomeningocele *(cont.)*

Authors	Design and setting/ Participants	Assessment tools and protocols	Outcome measure	Results
Nowak et al (2020)[92]	Cross-sectional, outpatient (Poland) 95 subjects with SB (all wheelchair users) 5-15 yo; 47% male	30-second Wingate Test on arm-crank ergometer (anaerobic) Handgrip strength	Mean power Peak power Relative mean power	- All values were increasing with age except for relative peak power where the mean score was lower in boys aged 12-13.9 yo when compared to 10-11.9 yo and handgrip strength where the mean score of girls aged 14-15.9 yo was lower than girls aged 12-13.9 yo. - Boys achieved higher scores than girls in all age groups. - When compared to the reference values for able-bodied children, the scores of children with SB were considered "very poor."

Note: Data are expressed in terms of mean ± *SD*. bpm = beats per minute; HR = heart rate; LMMC = lipomyelomeningocele; MMC = myelomeningocele; 6MWD = 6-minute walk distance; 6MWT = 6-minute walk test; SB = spina bifida; VO2 peak = peak oxygen consumption; yo = years old.

a variety of methods, most commonly with skinfold thickness measurements, approximately twice that of their typically developing peers[7,8,10-13,15,21-33] (**Table 1**).

Children with MMC and obesity are at risk of the same deleterious effects of obesity as typically developing children: high blood pressure, type II diabetes mellitus, atherosclerosis, non-alcoholic fatty liver disease, obstructive sleep apnea, and muscle and joint pain.[34-37] Due to their condition, they are also at risk of additional secondary complications such as pressure injuries, respiratory and gastrointestinal problems, depression, and mobility impairment.[6,38] Despite stable neurological functions, obesity when coupled with decreased strength can decrease walking ability in about one-third of community ambulators.[39] Obesity has been shown to affect functional independence in certain wheelchair users by increasing their difficulties with activities of daily living, such as bathing, dressing, locomotion, transfers, catheterization, and toileting.[35,40-42] For similar lesion levels, certain individuals with SCI and obesity underperform, due to their weight, in many activities of daily living.[40] Because of an absence of availability of bariatric ultralight weight wheelchairs, wheelchair users with obesity are also faced with more rolling resistance on flat and inclined surfaces, which increases the strength required to maneuver their wheelchair.[41] Thus obesity in patients with MMC should not be overlooked as for some it may compromise their quality of life by hindering independence, limiting participation in the community, limiting physical activity, and causing social isolation.[7,35,36,40,41,43]

A recent study conducted by McPherson et al.[6] showed that children acknowledge the importance of discussing weight-related issues and want to be involved. This further demonstrates the importance for health care providers to address these challenges and involve parents and children in appropriate interventions.

Causes of obesity. The causes of obesity in children with MMC are threefold. First, they often have more sedentary lifestyles due to a myriad of factors (mobility restrictions, cognitive impairments, incontinence, pressure injuries, and neuroendocrine disorders) that may hinder their ability to maintain a healthy weight by participating in physical activity.[6,7,21,35,44] Overall, energy expenditure in children and adolescents with MMC is significantly less than in their typically developing peers.[30,31,45-47]

Second, children and adolescents with MMC tend to have poorer diets.[6,10,48,49] For a subset of this population, particularly those with Chiari II malformations, this may be partly explained by dysphagia that can lead to a preference for certain textures, which unfortunately tend to be primarily energy-rich foods.[6,7]

Third, they have lower basal metabolic rates than their typically developing peers,[15,21,45,46] most probably due to differences in body composition, specifically a lesser total lean mass.[20,31,46,47] Indeed, children with MMC have higher percent body fat (21%-55%) than their typically developing peers.[30-32] As MMC predominantly affects the lower extremities, body fat is primarily distributed in the lower limbs as a result of muscle paresis, atrophy, and increased adipocytes in the atrophied tissue.[32] Although some studies have shown a positive relationship between percent body fat and the level of the lesion, with greater levels of body fat in higher levels,[22,32,50] others have shown no relationship.[23,33] Children and adolescents with MMC also have reduced lean body mass.[7,15,23,33,36,45] Interestingly, Shepherd et al.[50] stated that children with MMC younger than 4 years had body composition similar to their typically developing peers and that increased body fat was acquired with age and decrease in ambulatory activity. This fact reinforces the importance of early physical activity and nutritional interventions.

Appropriate assessment of obesity in children with MMC. The Canadian Pediatric Society recommends that all children over the age of 2 years have their growth monitored as it represents an ideal opportunity for health care providers to identify and address obesity early on.[51] The Center for Disease Control and Prevention (CDC) guidelines suggest using the body mass index (BMI), calculated as weight in kilograms divided by height in meters squared, as a representation of body fat; a child 2 to 18 years old is considered to be overweight when they are within the 85th to 95th percentile and obese

if within the >95th percentile.[52] Although BMI assessment is simple, cheap, and noninvasive, it can result in overestimation of BMI in nonobese MMC patients and underestimation in SCI patients.[15] Indeed, Liusuwan et al.[20] showed that the percent body fat in children with SCI was underestimated by approximately 10% per BMI value. Furthermore, accurate measurement of height is critical in calculating BMI, and it can be challenging to obtain in MMC patients with lower limbs contractures and scoliosis. When combined with lower limb muscle mass loss and hypoplasia due to paralysis, increased total body and trunk fat mass, decreased lean mass, and shorter stature, it skews BMI calculations, which further suggests that BMI may not be an appropriate screening tool to classify children with MMC as obese, overweight, or underweight.[15,47,50,53] There is currently no gold standard alternative for assessing and classifying the weight of children with MMC. However, it has been suggested that fat mass as measured by dual-energy X-ray absorptiometry (DEXA) correlated best with waist circumference and skinfold thickness measurements,[30,33,54] and thus the latest has been recommended for assessing body fat in children with MMC.[12,30]

Blood pressure

According to the National Health and Nutrition Examination Surveys, hypertension in the general pediatric population increased from 1988 to 2006.[55] MMC patients may be at higher risk of developing hypertension as they have a higher prevalence of obesity and renal and urinary tract dysfunction.[56] In recent studies, about 30% of obese adolescents with MMC were shown to have hypertension[15,25,37]; this percentage is much higher than what had been reported in typically developing children (2.6%-10.7%).[57] In addition, a large proportion of adolescents and young adults with MMC were found to be prehypertensive.[25,58]

Dyslipidemia

Van Speybroeck et al.[37] showed that children with MMC had lower serum high-density lipoprotein cholesterol (HDL) levels but did not differ from controls in levels of cholesterol, triglycerides, or low-density lipoprotein cholesterol (LDL). Abnormalities in lipid profile were associated with adiposity and level of lesion, whereby the profile was altered in patients with lumbar involvement but was normal in patients with sacral involvement. Similarly, Buffart et al.[25] reported that HDL level was reduced in 19% of the adolescents and young adults with MMC. In addition, 29% of the participants had elevated levels of total cholesterol, 38% had elevated levels of LDL, and 3% had elevated levels of triglycerides (TG), which may be explained by the fact that their population was much older than those in Van Speybroeck et al.[25] Nelson et al.[15] did not find any differences in lipid and lipoprotein profiles between children and adolescents with MMC and their typically developing peers of the same age who were obese. Comparably, Rendeli et al.[59] did not find any differences in lipid and lipoprotein profiles between a group of nonobese children and adolescents with MMC and their neurotypical nonobese peers. There were also some trends toward worsening dyslipidemia profiles in individuals who were nonambulatory versus ambulatory (see **Table 1** for details).[25,59] Although not completely consistent between studies, these findings suggest that obesity is the primary contributing factor to developing additional risk factors of metabolic syndrome such as dyslipidemia, and ambulation may be an important protective factor against dyslipidemia in individuals with MMC.

Insulin resistance

Insulin resistance is highly prevalent among children and adolescents with severe obesity, and thus patients with MMC may be at higher risk of developing insulin-resistance when compared to their typically developing peers.[60] We were able to find only two studies that looked at insulin resistance in this patient population. Nelson et al.[15] reported no differences between groups for fasting blood glucose but excluded participants with a prior known diagnosis of type 1 and type 2 diabetes. When comparing the children and adolescents with obesity, 30% with SCI had elevated fasting insulin compared with 10% of control participants and 6% of participants with SB, although these differences did not reach significance.[15] Van Speybroeck et al.[37] showed that children with MMC, more notably those with higher truncal fat, tended to have higher

levels of insulin and homeostasis model assessment of insulin resistance (HOMA-IR) when compared to their typically developing peers; however the differences did not reach significance, and fasting glucose levels did not differ between the two groups. In the limited research available on the subject, no significant differences are found in insulin resistance and glucose intolerance between adolescents and young adults with MMC, SCI, and controls.

Physical activity

In the general population, physical activity is known to decrease cardiovascular disease risk by increasing levels of HDL, decreasing blood pressure and insulin resistance, and improving aerobic capacity, muscle strength, and bone density.[61] Therefore logically, physical activity should be the foundation of healthy lifestyle recommendations in patients with MMC; however, very few studies have focused on the effects of exercise in this population.[62-64]

Aerobic fitness and endurance

Children with MMC have decreased aerobic fitness levels when compared to their typically developing peers (**Table 2**).[8,13,24,65-68] More specifically, their VO2 peak is 32% to 54% lower.[8,13,24,65-67] Because children with MMC are generally less active than their peers, it can lead to higher levels of deconditioning. As one might expect, patients with MMC with higher levels of everyday physical activity were found to be fitter than their peers who were less physically active.[21] Whether there is an association between VO2 peak levels and ambulatory status is debated in the literature.[8,21,24-26,67,69,70] It has been suggested that the difference seen in some studies may be explained by the fact that aerobic capacity is also influenced by the amount of active muscle mass, which may be diminished due to lower limb paresis in nonambulatory patients.[8] The maximum workload capacity in patients with SB has also been found to be 13% to 25% lower than in their neurotypical peers.[8,13,71] Finally, it has been reported that distance walked in the 6MWT (6 minute walk test) was 60% to 62% that of their typically developing peers. In addition, independent and unrestricted ambulators were shown to have higher 6MWT distance when compared to community ambulators, who are independent outdoor ambulators with or without use of braces and/or assisted devices but who use a wheelchair for longer distances. Also, patients without hydrocephalus and Chiari II malformation tended to have a higher 6MWT distance when compared to patients with hydrocephalus and Chiari II malformation.[63,68,69,72] Thus, higher levels of aerobic fitness and endurance appear to be associated with more daily physical activity, possibly ambulation independent of gait aids, and absence of history of hydrocephalus.

Muscle strength

Children and adolescents with MMC have less upper and lower body muscle strength than their typically developing peers, with poorer strength being associated with lower ambulatory status (**Table 2**).[8,13,27,65,68,73] However, Buffart et al.[8] concluded that 61% of their subjects had subnormal strength, with a greater percentage of ambulatory persons having lower muscle strength (79%) than nonambulatory individuals (54%). Such difference is likely explained by the fact that nonambulators tended to have higher muscle strength in the upper limbs, possibly due to daily wheelchair propulsion.[8] Thus, poorer strength may be also explained by muscles disuse and deconditioning in addition to the expected loss of muscle function below the level of the lesion. Nevertheless, Norrlin et al.[73] stated that lower levels of handgrip strength were found in children with MMC, independent of their ambulatory status. Poor hand strength was significantly correlated with the need for caregiver assistance, although most of these patients also had brainstem dysfunction. Therefore, it may be harder for patients with MMC to perform activities of daily living because of lower aerobic capacity and muscle strength. Finally, Buffart et al.[27] reported that 52% of adolescents and young adults with MMC who participated in sports had subnormal muscle strength compared to 81% of youth who do not participate in sports, thus highlighting the importance of physical activity.

Physical activity levels and impact of exercise on health in children with myelomeningocele

Children, adolescents, and young adults with MMC, specifically those who use a wheelchair[24,74]

or have a shunted hydrocephalus,[21,44] tend to be substantially less physically active in both type of activity and intensity levels when compared to patients without disabilities.[24,44,46,68,75,76] Indeed, a recent study concluded that children and adolescents with SB were 2.5 times less physically active than their typically developing peers.[74] Also, only 19% of the subjects met the physical activity intensity guidelines for children (>60 minutes of moderate to vigorous intensity of which 30 minutes should be of vigorous intensity) during school days and 8% during weekend days.[77] In addition, Buffart et al.[27] showed that adolescent and young adults with MMC had self-reported levels of physical activity considerably lower than other patients with a variety of physical disabilities, as measured with the Physical Activity Scale for Individuals with Physical Disabilities (PASIPD). Finally, Kelly et al.[78] suggested that physical activity levels in SB patients might be age-related, as youth ages 2 to 5 years participated more often in physical and skill-based activities than older youth.

Physical activity in children with MMC leads to positive changes in body composition such as higher lean mass, lower fat mass, and increased bone mineral density.[23,30] It can also result in higher levels of strength and physical fitness.[21,44,62] Furthermore, Roebroeck et al.[44] reported that ambulatory status was associated with everyday level of physical activity. Accordingly, in patients with MMC, physical activity leads to greater functional independence, higher quality of life, and self-worth.[25,27,79] It also increases participation, social integration, and life satisfaction and improves psychological well-being.[27,35]

What affects participation in physical activity in children with myelomeningocele?

Barriers

There are barriers to physical activity participation specific to children with MMC. Those include mobility impairments, actual and/or perceived medical contraindications to sports participation, incontinence, and pressure injuries.[6,7] Buffart et al.[27] showed that ambulatory status, the presence of shunted hydrocephalus, and functional independence were not related to sports participation. Contrarily, Kelly et al.[78] reported that youth having had hydrocephalus, those with major medical conditions, and children 6 to 12 years with bowel issues had lower levels of physical and social participation. Similarly, Bloemen et al.[80] concluded that medical problems, bowel and bladder care (especially when the child is incapable of self-catheterization), injuries, pain, and deformities are important physical activity barriers associated with SB. Being overweight or obese was also considered a barrier to exercise as it makes transfers more difficult.[80] Cognitive impairments, especially a lack of understanding of one's abilities and limitations in executive functioning, may also hinder physical activity.[35,44] However, in a study conducted by Buffart et al.,[27] sports participation was not associated with educational level or cognitive functioning.

Environmental factors such as a lack of accessible facilities, including not having access to a private, appropriately equipped public bathroom, lack of transport, lack of special equipment, and lack of trained staff have also been identified as common barriers to physical activity in children with SB.[35,80] Other barriers such as inadequate family resources, cost of adaptive equipment, and suboptimal adaptive physical education in schools may also play a role for some children.[7] Finally, in adolescents and young adults with MMC, Buffart et al.[27] showed that lack of time and lack of interest were the most frequently mentioned barriers for sports participation.

Facilitators

A study by Bloemen et al.[80] identified various facilitators to participation in physical activity in children with SB. Those included having access to appropriate assistive devices for optimal mobility and self-care and having access to trained fitness staff who are capable and willing to modify activities and who use an approach oriented toward solutions. Participation facilitators also included having the opportunity to participate in adapted sports with a suitable effort to rest ratio and where it is possible to meet new friends.

Perceived physical competence, physical appearance, and self-efficacy are also considered important facilitators of physical activity amongst children and adolescents with MMC.[27,81] Accordingly, activities that focus on achieving a

satisfactory level of fitness, wheelchair skills, and self-confidence should be encouraged. Buffart et al.[27] have shown that greater social support, particularly from the patient's family, is also important for adolescents and young adults with MMC to participate in sports. Finally, people who perceive higher enjoyment during sports tend to be more physically active, hence the importance of individually tailoring exercise programs improve compliance.[27]

Summary and Recommendations

The purpose of this narrative review was to describe the current scientific literature on the prevalence of metabolic syndrome in children with MMC and to gain insight into the baseline levels of aerobic fitness, endurance, and strength in this population in order to identify gaps in knowledge, suggest potential primary prevention strategies, such as physical activity, and provide recommendations for future studies.

In light of our review, here are some key findings and related recommendations for future studies. Children and adolescents with MMC tend to have decreased lean body mass and basal metabolic rate leading to overall decreased energy needs and decrease energy expenditure, as well as poorer diets, all of which put them at higher risk of obesity. They have also been shown to have comparatively decreased aerobic fitness and muscular strength and increased fat mass,[21,65,82] which, when combined with lower levels of physical activity, put them at higher risk of developing metabolic syndrome.[7,15,25,61,76] Indeed, Nelson et al.[15] showed that obese adolescents with spinal cord dysfunction have an increased prevalence of metabolic syndrome components. Although not completely consistent between studies, obesity seemed to be the primary contributing factor for developing additional risk factors of metabolic syndrome such as dyslipidemia. Ambulation seemed to be an important protective factor against some components of metabolic syndrome in individuals with MMC. Implementing positive health behaviors early in life is crucial as children with obesity have a 2.9 times higher risk of developing metabolic syndrome during adulthood,[18,19] which further increases their risk of having a cardiovascular disease.[16-18] However, considering the scarcity of the literature, more research is needed to determine the prevalence of components of the metabolic syndrome in children with myelomeningocele in particular and their impact on health. Also, future work should focus on the impact of various types of physical activity on metabolic syndrome risk factors specifically.

Physical activity in children with MMC leads to positive changes in body composition such as higher lean mass and lower fat mass.[23,30] It can also result in higher levels of strength and physical fitness[21,44,62] and leads to greater functional independence, higher quality of life, and greater self-worth.[25,27,79] It increases participation, social integration, and life satisfaction and improves psychological well-being.[27,35] Thus, logically, encouraging children and adolescents with MMC to engage in any kind of physical activity should be the foundation of healthy lifestyle recommendations and should be considered as a key metabolic syndrome primary prevention strategy.

For the health and well-being of children, the World Health Organization recommends that they engage in at least 60 minutes of moderate to vigorous intensity exercise daily.[83] However, there are currently no clear guidelines on exercise prescription in children with MMC. Recent exercise recommendations for cardiometabolic health benefits have been developed for adults with SCI consisting of 30 minutes of moderate to vigorous intensity aerobic exercise three times per week and strength training including all major muscle groups twice weekly.[84] In addition, Crytzer et al.[28] determined that initial exercise prescription in adolescent and adults with spina bifida can be set at around 61% of VO2 peak. Recent guidelines for physical activity in people who have SB were posted on the National Center on Health, Physical Activity and Disability (NCHPAD) website.[85] The guidelines appear to be inclusive of all ages. In the absence of specific data for people with SB, they recommend following the American College of Sports Medicine guidelines for physical activity, which suggest a minimum of 150 minutes of moderate intensity physical activity per week, divided into 30 to 60 minute blocks, undertaken 3 to 5 days per week,

incorporating flexibility, strengthening, and aerobic activity.[85] Finally, the recent guidelines on the care for people with SB recommend that unless a health care provider advises otherwise for medical reasons, children with SB 6 years or older should engage in 60 minutes or more of physical activity daily. Vigorous aerobic exercise should be done at least three times per week. Similarly, muscle strengthening exercises and bone strengthening activities should be performed at least three times per week as part of the 60 or more minutes.[86] Future studies should compare the cardiometabolic impact on patients with MMC of the various exercise guidelines already available in order to give more specific recommendations regarding type, duration, frequency, and intensity of exercise training programs for children with MMC.

Although there is a paucity of information regarding specific optimal exercise prescriptions for children with MMC, we suggest the following: a minimum frequency of 3 to 4 days per week, but optimally daily exercise of a moderate to vigorous intensity for 60 minutes total with at least 30 minutes of vigorous per day. Finally, the type of activity should be varied to prevent overuse or overtraining injuries, focusing on activities the child finds engaging and entertaining to foster long-term maintenance of physical activity, such as active play or games (encouraging inclusion in recess physical activities), dance, and/or sports (adapted to their abilities). The aforementioned activities have the advantage of combining strengthening and aerobic components while remaining fun and engaging to many children. For some children who are particularly deconditioned, incrementally increasing daily locomotion (ambulation or manual wheelchair propelling) may impact aerobic capacity accessibly. Stretching should be encouraged following the physical activity.

There are currently missed opportunities for health care providers to address weight-related challenges and involve parents and children in appropriate interventions. Until more research is conducted, addressing weight-related challenges and promoting healthy habits (such as optimal activity levels) could be easily integrated into yearly MMC clinics. An actionable suggestion might be to systematically weigh and measure children in these clinics and utilize the results and trends as a talking point with the parents and children. When feasible, waist circumference and skinfold thickness measurements could also be added to better assess body fat specifically in children with MMC. In addition, we recommend that health care practitioners tasked with this intervention (physician, nurse, etc.) should be aware of locally available accessible sports platforms and have knowledge of motivational interviewing to facilitate removal of perceived barriers to physical activity. The follow-up appointments could also be used to develop physical activity goals and monitor progress. Finally, health care providers should advocate for children with MMC, for example, by bringing key issues to the local community such as accessibility of sports facilities.

Conflicts of Interest

The authors declare no conflicts of interest.

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