

Fat–Bone Interactions in Adults With Spina Bifida

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Context: Spina bifida (SB) can lead to changes in body composition and bone mineral density (BMD) through diminished ambulation, renal impairment, and anticonvulsant medication. With increased life expectancy, diseases such as obesity and osteoporosis are emerging comorbidities in SB, with limited data to guide management.

Objective: To examine the relationship between cardiometabolic factors, body composition, BMD, and minimal trauma fractures (MTFs) in adults with SB.

Design: Retrospective cross-sectional study.

Setting and Participants: Forty-nine adults with SB (median age, 32.7 years; interquartile range, 22.6 to 39.0) who had undergone dual-energy x-ray absorptiometry imaging at a single tertiary hospital from 2004 to 2015.

Results: The mean body mass index was 31.7 ± 7.5 kg/m²; 26 (53.1%) were obese. Using age- and sex-matched fat percentiles from the National Health and Nutrition Examination Survey III, 62.5% had a total body percentage fat greater than the 95th percentile. Low bone mass (defined as a Z-score of ≤ -2.0) was present in 21.9% at the L1 vertebra and in 35.1% at the femoral neck. Ten (20.4%) had a history of MTFs. A BMD or Z-score at L1, femoral neck, or total body site did not correlate with the occurrence of MTF. Fat mass was significantly and positively associated with BMD after adjustment for age, sex, and height and accounted for 18.6% of the variance in BMD ($P = 0.005$). The prevalence of metabolic comorbidities, such as hypertension (20.4%) and obstructive sleep apnea (16.3%), was high.

Conclusions: Obesity and low BMD are common in young adults with SB. An increased fat mass correlated significantly with BMD. The prevalence of metabolic complications in patients with SB is increased and deserves further study.

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Freeform/Key Words: body composition, bone density, myelomeningocele, obesity, osteoporosis, spina bifida

Spina bifida (SB) occurs when the neural tube fails to fuse during embryonic development, leading to neurologic deficits below the level of the lesion. Motor, sensory, and autonomic function can be affected, leading to reduced mobility, the risk of pressure sores, orthopedic

Abbreviations: BMD, bone mineral density; BMI, bone mineral index; DXA, dual-energy x-ray absorptiometry; FM, fat mass; LTM, lean tissue mass; MTF, minimal trauma fracture; OSA, obstructive sleep apnea; SB, spina bifida.

deformities, and neurogenic bladder and bowel. With the increasing life expectancy [1, 2], chronic diseases of adulthood, including obesity, osteoporosis, cardiovascular disease, and renal failure, are also emerging as health problems in this vulnerable group. However, limited data for adults with SB are available to guide the clinical management of these conditions.

Dual-energy x-ray absorptiometry (DXA) imaging allows for the simultaneous assessment of body composition and bone density. Obesity has been reported as early as 6 years of age in children with SB [3, 4] and becomes an increasing health issue with adulthood [5]. The mortality and morbidity of SB have been shifting from renal failure to those associated with metabolic derangements, such as stroke and acute myocardial infarction [6]. The distribution of fat is pathologically altered in those with SB, with a greater distribution in the lower limbs [7] and predominantly within the muscle compartment [8].

Obesity, however, might have protective effects on bone, with previous studies documenting a positive association between body mass index (BMI) and bone mineral density (BMD) [9, 10]. The reduction in fracture risk with an increased BMI is likely site-specific, with reduced hip and pelvis fractures but increased extremity fractures [11, 12]. This can be explained, in part, by soft tissue padding at these proximal sites. Fat might also exert positive effects on bone through the secretion of hormones such as insulin, amylin, and leptin and through the increased aromatization of testosterone to estrogen [13, 14].

A low bone mass is common in children with SB, with a prevalence of $\leq 65\%$ [15] and has been strongly associated with nonambulatory status and higher spinal level deficits [16, 17]. Other factors likely to play a role include vitamin D deficiency, anticonvulsant use, renal dysfunction, and urological intervention with intestinal segments, leading to metabolic acidosis. Fractures are prevalent in $\leq 30\%$ of children [18, 19] and predominantly involve the lower limbs. Measurement of bone density in those with SB is hampered by positioning difficulties and artifacts from metal implants from orthopedic operations. Furthermore, the absence of posterior elements in the vertebrae of patients with SB can spuriously lower the BMD of the lumbar spine [20]. Accordingly, some groups have measured the bone density of the L1 vertebra only, because this will be affected in only a few patients with high-level lesions [21, 22].

To the best of our knowledge, only one study has reported on the bone density in adults with SB [21]. That study included 21 participants with a mean age of 30 years. They had recruited from a rehabilitation service [21]. Using a T-score of < -2.5 for the diagnosis of osteoporosis, 33% had osteoporosis in at least one of the sites measured (L1 vertebra, femoral neck, or trochanter), and 47% had osteoporosis at either the femoral neck or trochanter. Given the young age of subjects in these studies, these prevalence rates were significantly greater than those of the general population; however, the significance of this finding is unclear, given the fracture rates in young adults with SB appear to be lower than those of unaffected adults [23, 24].

The aim of the present study was to characterize the changes seen in body composition and bone density in a young adult population with SB using DXA and to examine how the changes in body composition might affect the BMD and metabolic risk factors in these individuals.

1. Methods

A. Patients

A retrospective cross-sectional cohort study of 49 adults with SB who had undergone DXA imaging at a single tertiary hospital from 2005 to 2015 was undertaken. The medical indications for DXA imaging included a suspected low BMD in the setting of immobility, fracture, renal disease, anticonvulsant use, and hypogonadism. The Monash Health human research ethics committee approved the present study.

B. Data Collection and Clinical Measures

Patients were excluded if they had SB occulta or isolated caudal regression syndrome. Demographic data, the prevalence of hydrocephalus, current or previous use of anticonvulsant

medication, and minimal trauma fracture (MTF) were obtained from the medical record. A MTF was defined as a self-reported or radiologically proven fracture occurring after a fall from a standing height or less or a minimal trauma incident other than a fall (e.g., turning over in bed). Fractures of the skull, hands, and toes were excluded.

Ambulatory status was defined using the Hoffer ambulation scale [25] as community, household, functional, or nonambulators. The patients were then further divided into predominantly ambulators (community/household) and nonambulators (functional and nonambulators). The spinal level was obtained from the medical record and divided into high (above L3), mid (L3-L4), and low (L5 and below).

Data regarding urological intervention, which involved intestinal segments (bladder augmentation with intestinal segments or ileal conduit), were collected. The presence of renal dysfunction and metabolic acidosis was assessed from the estimated glomerular filtration rate, serum creatinine, bicarbonate, and chloride levels obtained from the medical records. Hypogonadism was defined in males as the use of androgen replacement therapy or low testosterone levels (<8 nmol/L) documented on two separate occasions and in females as the use of hormone replacement therapy for induction of pubertal development, menopause at age ≤ 40 years, or low estradiol levels (<73 pmol/L) on two separate occasions. Other biochemical variables included serum 25(OH) vitamin D, alkaline phosphatase, and γ -glutamyl transpeptidase at BMD measurement.

The medical conditions associated with obesity were documented and included cardiovascular disease, deep venous thrombosis, pulmonary embolism, obstructive sleep apnea (OSA), cerebrovascular disease, type 2 diabetes mellitus, and hypertension. If patients died during the study period, the age at death and the cause of death were recorded.

B-1. BMD measurements

BMD was measured using DXA at the L1 vertebra, femoral neck, and total body for all participants, unless limited by previous scoliosis surgery, femoral surgery, or difficulty with positioning. The presence of vertebral arch deficits can falsely lower the L1–L4 lumbar spine BMD. The L1 vertebra was therefore chosen, because it is rarely involved in SB, in line with previous studies [21, 22]. Patients were excluded if the entire lumbar spine was involved. A low BMD was defined as a Z-score of ≤ -2.0 , in accordance with the International Society of Clinical Densitometry guidelines for adults aged <50 years [26]. The coefficient of variation for BMD of a Lunar anthropomorphic lumbar spine phantom measured daily from mid-2004 to the end of 2011 was 0.51%. The coefficient of variation for the percentage body fat of a total body phantom measured weekly was 3.11%.

The total lean tissue mass and fat mass were derived from the whole body scan. All measurements were obtained using a General Electric Lunar Prodigy, software version 12 (Madison, WI) at a single center. The anthropometric measures of age, weight, and height were documented. In cases in which the true height could not be obtained, the patient's length lying flat was used for the BMI calculations.

The adults were categorized into four BMI groups: underweight (BMI, <18.5 kg/m²), normal (BMI, 18.5 to 25 kg/m²), overweight (BMI, >25 kg/m²), or obese (BMI, >30 kg/m²). The percentiles for fat mass were calculated from the National Health and Nutrition Examination Survey 1999 to 2004 body composition data matched for age and sex [27]. The fat mass was further divided into the trunk, arms, and legs, and the percentage of fat per segment was calculated by dividing the segment fat mass by the total mass of the segment. An increased fat mass was defined as $>35\%$ for females and $>25\%$ for males in accordance with the American Association of Clinical Endocrinologists/American College of Endocrinology guidelines [28].

C. Statistical Analysis

The distribution of data was explored using the Shapiro-Wilk test. All normally distributed data are expressed as the mean \pm standard deviation and the nonparametric data as the

median and interquartile range (IQR). Differences between groups were determined using the independent *t* test for normally distributed variables, the Mann-Whitney *U* test for non-parametric variables, and the Fisher exact test for categorical variables. Univariate analysis was used to determine the predictors of fracture.

Univariate regression analysis was performed to examine the correlation between the fat mass (FM) and lean tissue mass (LTM) with BMD at L1, the femoral neck, and total body separately. This was followed by a multivariate analysis after adjusting for age, sex, and height in these models. Multicollinearity was determined for multivariate regression models by calculating the variance inflation factor. Any regression model with a variance inflation factor >10 was excluded.

A *P* value of <0.05 was considered statistically significant, and all tests were two-sided. The analyses were conducted using SPSS statistics for Windows, version 22 (IBM Corp., Armonk, NY).

2. Results

A. Baseline Characteristics

The clinical characteristics of the 49 patients are summarized in [Table 1](#). The median age of the patients was 32.7 years (IQR, 22.6 to 39.0), 40.8% were male, and more than one-half were nonambulatory. Spinal level involvement was above L3 in 9 patients (18.4%), at L3-L4 in 16 patients (32.7%), and at L5 and below in 24 patients (49%). Also, 27 patients (55.1%) had a history of urological intervention with either bladder augmentation or formation of the ileal conduit. The mean duration of urological intervention was 24.8 ± 12.5 years. Hypogonadism was found in 5 patients, all male, of varying etiology. All received testosterone replacement. For 40 patients, the total body BMD was obtained, enabling the total and regional LTM and FM to be derived.

B. Body Composition

The mean BMI was 31.7 ± 7.5 kg/m², in the obese range for adults. Only 10 patients (20.4%) had a normal weight or were underweight, with 13 patients (26.5%) overweight and 26 (53.1%) obese. Of the 8 patients with a normal BMI, 7 (87.5%) had an increased percentage of body fat on DXA (defining as body fat >25% in men or >35% in women), which would classify these patients as obese in accordance with the American Association of Clinical Endocrinologists/American College of Endocrinology guidelines [28]. The prevalence of obesity according to total body fat percentage was 87.5%. Using age- and sex-matched fat percentiles from National Health and Nutrition Examination Survey III [27], 25 of 40 (62.5%) had a DXA total body fat percentage greater than the 95th percentile. When the regional fat percentiles were calculated, the percentage of tissue mass that was fat by site was at the 50th percentile for arms (IQR, 12.5% to 75%), the 95th percentile for legs (IQR, 90% to 95%), and 95th percentile for trunk (IQR, 50% to 95%).

The BMI was not significantly different statistically between the ambulatory and non-ambulatory patients (30.3 ± 6.8 kg/m² vs 32.0 ± 8.7 kg/m², respectively; *P* = 0.58). The distribution of fat and lean tissue according to ambulatory status is shown in [Table 2](#). Before adjustment for age, sex, and height, those who were nonambulatory had a greater percentage of leg mass that was fat than did the ambulatory patients (56.8% vs 46.7%; *P* = 0.006). No relevant difference was found in the percentage of arm or trunk mass that was fat between the ambulatory and nonambulatory patients. Similar results were found after adjusting for covariates. A trend was found toward a greater percentage of trunk fat and total body fat in those who were nonambulatory, but this did not reach statistical significance. The relationship between LTM and ambulatory status showed substantially greater leg, trunk, and total lean tissue for the ambulatory patients; however, the difference failed to maintain statistical significance after adjustment for age, sex, and height.

Table 1. Baseline Characteristics (n = 49, Unless Specified Otherwise)

Variable	Patients
Demographic	
Age, y	
Median	32.7
IQR	22.6 to 39.0
Male sex, n (%)	20 (40.8)
Anthropometric	
Weight, kg	71.1 ± 19.6
Height, cm	151.2 ± 11.0
BMI, kg/m ²	31.0 ± 7.5
Clinical, n (%)	
Spinal level	
High	9 (18.4)
Mid	16 (32.7)
Low	24 (49.0)
Nonambulatory	27 (55.1)
Anticonvulsant use	10 (20.4)
Hydrocephalus	36 (73.5)
Fracture	10 (20.4)
Urological intervention	27 (55.1)
Upper tract calculi	2 (4.1)
Renal impairment (eGFR <90 mL/min/1.73 m ² ; n = 42)	9 (18.4)
Hypogonadism	5 (10.2)
DXA	
LTM, kg	36.9 ± 9.0
FM, kg	30.4 ± 13.8
Bone mineral content, kg	2.40 ± 0.51
L1 BMD (n = 32), g/cm ²	1.04 ± 0.15
L1 Z-score	
Median	-1.1
IQR	-1.9 to -0.1
FN BMD (n = 37), g/cm ²	0.81 ± 0.13
FN Z-score	
Median	-1.5
IQR	-2.4 to -0.7
TB BMD (n = 40), g/cm ²	1.14 ± 0.10
TB Z-score	
Median	-0.3
IQR	-1.1 to 0.5
Radius BMD (n = 27), g/cm ²	0.86 ± 0.11
Radius Z-score	
Median	-0.6
IQR	-1.3 to 0.4
Biochemical	
Vitamin D (n = 23), nmol/L	58.0 ± 25.3
Creatinine, mmol/L	
Median	51
IQR	38 to 84
Bicarbonate, mmol/L	24.4 ± 3.4
Chloride, mmol/L	105.5 ± 3.3
ALP (U/L)	
Median	92.5
IQR	75 to 112.5
GGT (U/L)	
Median	20.5
IQR	14.5 to 50

Abbreviations: ALP, alkaline phosphatase; eGFR, estimated glomerular filtration rate; FN, femoral neck; GGT, γ -glutamyl transpeptidase; TB, total body.

Table 2. DXA and Body Composition Parameters According to Ambulatory Status

Region	Nonambulatory	Ambulatory	P Value
Lumbar spine Z-score (n = 32)	-1.46 (0.3 to -3.2)	-0.72 (1 to -3.7)	0.096
Femoral neck Z-score (n = 37)	-1.97 (-0.2 to -3.9)	-1.36 (-0.1 to -3.5)	0.097
Lean tissue (n = 40), %			
Arm	0.08 ± 0.04	0.08 ± 0.02	0.82
Leg	0.11 ± 0.03	0.14 ± 0.02	0.001
Trunk	0.24 ± 0.09	0.29 ± 0.06	0.06
Total	0.48 ± 0.17	0.56 ± 0.11	0.08
Fat (n = 40), %			
Arm	33.93 ± 14.53	33.01 ± 13.20	0.84
Leg	56.82 ± 9.97	46.65 ± 11.66	0.003 ^a
Trunk	47.94 ± 13.21	41.99 ± 12.19	0.051
Total	46.83 ± 12.03	40.76 ± 11.65	0.095

^aStatistically significant after adjusting for sex, age, and height.

C. Morbidity and Mortality

The patients had a mean follow-up duration of 6.54 ± 3.54 years, with four patients (8.1%) dying during the study period. The cause of death in one patient each was intra-abdominal sepsis (age, 21 years), acute hydrocephalus (age, 24 years), aspiration pneumonia (age, 41 years), and metastatic transitional cell carcinoma of the bladder in an ileal conduit (age, 50 years). The prevalence of metabolic comorbidities was high, including hypertension in 20.4%, OSA in 16.3%, deep vein thrombosis or pulmonary embolism in 8.2%, and type 2 diabetes mellitus requiring medication in 6.1%. In terms of cardiovascular disease, only one patient had a documented cerebrovascular accident, and none had documented ischemic heart disease.

D. DXA Parameters

L1 BMD was obtained in 32 patients (65.3%) and femoral neck BMD in 37 patients (75.5%). The median lumbar spine Z-score was -1.05 (IQR, -1.85 to 0.1), and the median femoral neck Z-score was -1.50 (IQR, -2.4 to -0.7). A reduced BMD, as defined by a Z-score of ≤ -2.0 , was present in 7 of 32 patients (21.9%) at L1 and in 13 of 37 patients (35.1%) at the femoral neck.

E. DXA and Fracture

Of the 49 patients, 10 (20.4%) had a history of fragility fracture, with one patient experiencing four fractures. Six patients had fractures that had occurred during childhood and four had experienced fractures in adulthood. None of those with fractures in childhood experienced additional fractures in adulthood. All fractures involved the lower limb except for two fractures of the shoulder.

Fractures were not significantly associated with the clinical risk factors for osteoporosis or anthropometric or DXA parameters on univariate analysis. The BMD and Z-scores at the L1 vertebra, femoral neck, and total body were not associated with the occurrence of fracture. In addition, BMI, height, weight, and DXA parameters of FM and LTM were not associated with the occurrence of fracture. Clinical variables, including sex, hydrocephalus, ambulatory status, urological intervention, presence of renal calculi, renal impairment, hypogonadism, and anticonvulsant use, were not associated with either BMD (femoral neck or lumbar spine) or fracture status.

F. Relationship Between Body Composition and Bone Mass

The relationship between BMD and body composition parameters was examined using linear regression modeling. Univariate analysis showed that 18.6% of the variance in total body

BMD was accounted for by the FM and 14% by the LTM ($P < 0.05$; Table 3). After adjustment for age, sex, and height, only FM had a statistically significant positive association with total body BMD ($R^2 = 0.399$; $P = 0.019$, $\beta = 0.427$, $P = 0.014$). The BMI also correlated extensively with the total body BMD on both univariate and multivariate analysis after adjusting for age and sex.

3. Discussion

To the best of our knowledge, the present study included the largest cohort of young adults with SB to date in whom bone density and body composition were examined. More than 50% of our cohort was obese using BMI criteria, and 87.5% were obese according to the total percentage of body fat, as calculated using DXA. This percentage is greater than the previously reported prevalence of 35% to 37% in young adults with SB [5, 29]. In a similar study of 18 adults with SB, Liu *et al.* [30] also found that the BMI calculated using either the length or arm span grossly underestimated the presence of obesity compared with that determined by trunk fat recorded using DXA. Coupled with the high prevalence of obesity-related complications found in this cohort, optimizing the cardiometabolic health of young adults with SB is important.

Obesity is a well-recognized problem for patients with SB and is thought to result primarily from reduced energy expenditure [3]. Other causes of weight gain can include hydrocephalus and cerebral abnormalities, leading to neurohormonal and appetite disturbances, and epigenetic factors, with maternal obesity and diabetes both risk factors for neural tube defects [4, 5, 31, 32]. In our study, leg fat was substantially increased, as expected; however, of greater concern was that the percentage of trunk fat was also at the 95th percentile. This has implications for cardiovascular health in this young population [33]. The lack of ambulation was associated with increased fat in the legs; however, only a trend was found toward reduced total body fat and trunk fat. This might have been because of our small sample size and the lack of functional measures; however, 50% of the ambulatory patients had a BMI within the obese range. The changes in body composition seen in our adult cohort could not be satisfactorily explained by ambulatory status. Other small studies of adults with SB have similarly shown no correlation between obesity and ambulatory status or physical activity levels [5, 30]. It could be that interventions to increase physical activity should be implemented earlier, considering that young children with SB (mean age, 9 years) were found to have no differences in trunk fat compared with their peers [7].

In a previous study of 225 subjects with SB, 43% had hypertension, 7.5% had diabetes, and 6.6% had renal impairment [34]. In a 40-year longitudinal study, the mortality rate per decade was 9% (7 of 77) for those aged 5 to 14 years, 13% (9 of 70) for those aged 15 to 24 years, and 15% (9 of 61) for those aged 25 to 34. The cause of death was mainly unexpected and included epilepsy, pulmonary embolus, acute hydrocephalus, and acute renal sepsis [35]. We found high rates of obesity-related diseases in our population, including hypertension (20.4%), OSA requiring continuous positive airway pressure (16.3%), and deep vein thrombosis

Table 3. Relationship Between Body Composition and Total Body BMD (n = 40)

Variable	R^2	P Value	β	P Value
Fat	0.186	0.005 ^a	0.432	0.005 ^a
Muscle	0.140	0.016 ^a	0.374	0.016 ^a
Model ^b	0.310	0.019 ^a		
Fat	NA	NA	0.427	0.014 ^a
Muscle	NA	NA	0.258	0.291

Abbreviation: NA, not applicable.

^aStatistically significant.

^bLinear regression model.

or pulmonary embolism (8.2%). From these data, deep vein thrombosis prophylaxis should be considered for nonambulatory young adults with SB who require surgery. Medications that can increase the risk of thrombosis such as oral contraceptive medication should also be used with caution in this population.

Obesity, however, can have positive effects on bone through fat–bone interactions. A greater BMD in those with a higher BMI could result from increased LTM or FM. In previous studies of body composition, LTM accounted for a larger variance in BMD than did FM in young females and males, although in postmenopausal women, the FM was relatively more important [39–41]. In our cohort, we demonstrated a relevant and positive correlation between FM and total body BMD, even after adjustment for age, sex, and height. The unusual loss of the muscle–bone relationship in young adults might result from the alterations in body composition seen in adults with SB. Whether this can explain why adults with SB appear to have a lower than expected risk of osteoporosis and fracture requires validation in other cohorts.

We were unable to demonstrate a relationship between BMD and the occurrence of fracture, in line with previous studies [15, 16, 42]. This was likely because of the small sample sizes in these studies and the inherent limitations of using DXA in this population. Overall, a low bone mass was seen in 21.9% of our patients at the L1 vertebra and in 35.1% of patients at the femoral neck. This was lower than the only other study of BMD in adult patients with SB, in which 47% of patients demonstrated low BMD at the femoral site [21]. Compared with the study by Valtonen *et al.* [21], our participants were of similar age and had an equivalent prevalence of obesity, ambulation, renal impairment, and anticonvulsant use. Also, more fractures were documented in their group than in ours (38.1% vs 20.4%); however, it is unclear whether the fractures identified by Valtonen *et al.* [21] were MTFs. The lower than expected rates of fracture described previously in young adults with SB suggests that treatment based on a low BMD alone should be approached with caution. Other risk factors such as a history of a fragility fracture or rapid bone loss might signal the need for intervention. Longitudinal studies of BMD are required, especially in older adults with SB, who will experience further bone loss with age.

Adults with SB have a high burden of chronic medical conditions and are at risk of bone disease. Despite the young age of our cohort (median age, 32.7 years), more than one-half of all patients were nonambulatory, one in five patients required anticonvulsant medications, and one in five patients had renal impairment. In addition, 1 in 4 men (5 of 20 men) had hypogonadism. We recommend DXA screening for adults with SB who have sustained a fragility fracture, have developed a chronic disease associated with bone loss, or require medications associated with bone loss [26]. This includes patients treated with antiepileptic medication and patients with hypogonadism and renal disease. In the absence of evidence-based guidelines to guide clinical practice, the treatment of young adults with low BMD needs to be individualized. Ensuring vitamin D levels are replete, treating hypogonadism in those without contraindications, encouraging weightbearing exercise (where possible), and optimizing general health are all appropriate. The use of specific osteoporosis medications should be assessed on a case-by-case basis by the treating physician but should be limited to individuals with fracture and/or a severe reduction in BMD, after discussion of the risks and benefits with the patient.

From the perspective of body composition, it is not surprising that obesity in adults with SB is common, given the high prevalence of childhood obesity described with this condition [3, 4]. In adults with SB and obesity, we recommend screening for hypertension, dyslipidemia, impaired glucose tolerance, and diabetes mellitus and for the associated comorbidities, including OSA [43]. Strategies are urgently needed to reduce the incidence and severity of childhood obesity and to effectively treat adult obesity, especially in this vulnerable group.

To the best of our knowledge, ours is the first study to assess both BMD and body composition, using DXA in adults with SB. Our findings highlight the importance of screening for obesity and its related complications. DXA allows for performance of detailed body composition analysis, overcoming the limitations with traditional anthropometric measurements

such as BMI. Patients with SB can experience growth restriction and scoliosis or joint contractures, which can further skew measurements of height. It also allows for the assessment of fat distribution, which is important when considering cardiovascular risk. However, the present study had a number of limitations, in particular, the small sample size, especially when reporting medical comorbidities in this group. We were unable to fully adjudicate or confirm death of our subjects who were lost to follow-up. In addition, adult patients with SB who continue to attend outpatient clinics at a tertiary referral center are more likely to have more severe neurologic disease and medical comorbidities than those in the general community. These medical comorbidities include renal disease, hypogonadism, and anticonvulsant use, all of which can contribute to bone disease and increase fracture risk. Furthermore, insufficient data were available regarding vitamin D status. Therefore, we were unable to examine any effect of vitamin D levels on BMD. As described previously, BMD measurements can be difficult to interpret in patients with SB and, furthermore, will be affected by body size. We have described an association between FM and BMD. However, owing to the cross-sectional nature of our study, this should be explored further.

4. Conclusions

Adults with SB have a high risk of both low BMD and obesity. Obesity could play a role in the skeletal phenotype of this population. The clinical assessment of bone health (in particular, in those with risk factors for bone disease) and screening for obesity with its related metabolic complications is recommended for adults with SB. The high prevalence of metabolic risk factors is of particular concern and requires further study.

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