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Adolescent paraplegia, morbid obesity, and pickwickian syndrome: outcome of gastric bypass surgery

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Abstract Loss of mobility, such as what occurs as a consequence of spinal cord injury or malformation, is a risk factor for excess weight gain and can confound weight management efforts. Despite well-documented outcomes of bariatric surgery in ambulatory patients, little information is available regarding weight loss surgery in adult or adolescent paraplegic patients. A 15-year-old adolescent boy with a body mass index of 60 kg/m^2 and complete paraplegia caused by spina bifida developed metabolic dysfunction, severe obstructive sleep apnea, and hypoxemia syndrome. In an effort to avoid a tracheostomy for worsening pickwickian syndrome, he was referred for weight loss surgery. Laparoscopic Roux-en-Y gastric bypass surgery was safely performed and resulted in loss of 55% of body weight (83.8% excess weight loss) for 2 years. Risk factors for cardiovascular disease markedly improved, and polysomnography demonstrated complete reversal of sleep apnea with substantial subjective improvement in daytime breathlessness and quality of life. Body composition analysis demonstrated preferential reduction in body fat mass compared with lean mass, without detrimental effect on bone mineral density. This case illustrates that paraplegia does not necessarily impair either weight loss efficacy or comorbidity resolution after Roux-en-Y gastric bypass surgery.

In the United States, pediatric obesity has reached epidemic proportions. Prevalence of obesity has tripled for the last 2 decades and now affects nearly 20% of children [1-3]. Of major concern is the fact that, on average, 4% of US children (more than 2 million) are extremely obese (99th percentile of body mass index [BMI] for age) [4]. There is currently no strong evidence that behavioral modification or pharmacotherapy results in significant and sustained weight loss for obese children [5]. This is especially true for those with extreme obesity. Bariatric surgery results in significant and sustained weight loss, as well as striking remission of many obesity-related comorbidities [6,7].

Spina bifida is one of the most common serious birth defects, occurring in 3 to 8 per 10,000 live births [8,9]. The spinal cord and vertebrae are malformed, usually leaving the lower limbs partially or completely paralyzed. Approximately 30% to 50% of those affected with spina bifida are functional ambulators, and 30% to 40% rely mainly or entirely on a wheelchair for mobility [10,11]. Lack of mobility and physical inactivity contribute to development of obesity.

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When bariatric surgical consultation was requested for an extremely obese adolescent with spina bifida who had developed severe comorbidities of obesity, there was no published information available on which to base medical decision making and family counseling. Indeed, based on the medical and surgical history (paraplegia, multiple prior operations), in this case, there was a good reason to hypothesize that the operation might not be as successful compared with outcomes that are seen in ambulatory patients. This case, therefore, highlights important aspects of the rationale, intervention, and outcome of laparoscopic Roux-en-Y gastric bypass (RYGB) in the management of an adolescent with complete paraplegia.

1. Case report

The subject of this report is a 15-year-old white adolescent boy born with spina bifida and paraplegia. His physical activity was limited because of complete paraplegia and dependence upon a wheelchair for mobility. He had progressively gained weight since 9 years old and had experienced multiple failed attempts at weight loss, including physician-directed weight management initiatives, as well as parent- and self-directed efforts since 11 years old. He lived with his mother, father, and sister, all of whom were overweight. Although he had participated in physical therapy once per week and used handheld weights 2 to 3 times per week, he was otherwise unable to do any substantial aerobic activity. His quality of life was increasingly eroded by his worsening obesity. For instance, his neurogenic bladder required intermittent catheterization, but as a result of his centrally obese habitus, he was unable to self-catheterize and often resorted to wearing adult diapers.

After repeated unsuccessful conventional weight loss attempts, he was referred in November of 2004 for consideration of weight loss surgery. His height was 139.7 cm and his weight was 117.9 kg, with a BMI of 60.4 kg/m^2 (Table 1). He presented with elevated fasting glucose, hyperinsulinemia, and elevated low-density lipoprotein cholesterol. He also had developed pulmonary insufficiency, and by formal polysomnography, he was diagnosed with severe obstructive sleep apnea syndrome (OSAS). His apnea-hypopnea index was 15.6 events per hour, more than 3× the upper limit of normal, and his minimum arterial oxygen saturation during sleep was 70% (Table 1). His sleep hygiene was impaired by frequent arousals (arousal index was 13.4 events per hour), and sleep efficiency (percentage of time spent in sleep after first onset of sleep) was 78%. Bilevel positive airway pressure (BiPAP) support was the initial prescribed treatment, but residual severe hypoxemia during rapid eye movement sleep was noted on titration polysomnogram. Indeed, the inadequate correction of his OSAS with BiPAP and

Table 1 Pre- and postoperative (1 and 2 year) characteristics

BMI change	50th percentile *	Preoperative	Postoperative	
			1 y	2 y
Weight (kg)	56.4	117.9	77	52.4
Height (cm)	170.1	139.7	139.7	139.7
BMI (kg/m^2)	19.8	60.4	39.4	26.9
Sleep study	**Reference range**			
Apnea hypopnea index	<5 events/h	15.6		1.2
Arousal index	≤10/h	13.4		3.9
Respiratory disturbance index	<5 events/h	30		1.8
Sleep efficiency	≥88%	78%		87%
Minimum O_2 desaturation	≥90%	70%		94%
Laboratory data	**Reference range**			
Triglycerides (mg/dL)	32-134	95	82	59
High-density lipoprotein cholesterol (mg/dL)	28-72	47	46	46
Low-density lipoprotein cholesterol (mg/dL)	55-120	126	128	88
Total cholesterol (mg/dL)	105-218	192	189	146
Insulin (μIU/mL)	5.0-26.0	70.4	11.1	6.8
Glucose (mg/dL)	70-105	109	82	81
Albumin (g/dL)	3.5-5.0	3.7	4.6	3.6
Vitamin B1 (μg/dL)	1.6-4.0	2.2	5.1	4.3
Iron (μg/dL)	75-150	59	73	98
Hemoglobin (g/dL)	13.0-16.0	14.9	17.1	15.8

* Fiftieth percentile for a 15-year-old adolescent boy.

concerns about daytime hypoxemia had prompted referral to an otolaryngologist in his local community for tracheostomy. However, the procedure was deemed too risky given his cervical adiposity, and the procedure was not performed.

Given the severity of his obesity and related comorbidities, coupled with his maturity, satisfactory compliance with prior medical regimens, supportive family environment, and realistic expectations, bariatric surgery was recommended. During the next 5 months after initial referral, a detailed educational program and informed consent process related to RYGB were completed by the patient and primary caregivers, and the patient underwent surgery for weight loss.

Under general endotracheal anesthesia, he was placed in supine position safely despite his kyphosis. For laparoscopic RYGB, a direct-viewing trocar entry into the peritoneal cavity was chosen, and pneumoperitoneum was safely established. Numerous intra-abdominal adhesions were encountered because of his prior surgical history, and the abdominal domain was limited because of his habitus (kyphosis and central obesity). Despite these challenges, laparoscopic RYGB was performed without complications. After the creation of a 30-mL gastric pouch, the jejunum was transected at 30 cm distal to the ligament of Treitz for creation of the 100-cm Roux limb. A stapled jejunojejunostomy was performed, the Roux limb was tunneled in a retrocolic fashion, and a 2-layered hand-sewn gastrojejunostomy was performed [12].

His postoperative course was uneventful, and he was discharged on postoperative day 5. His prescribed medications included vitamin B_{12}, 2 pediatric multivitamins, and calcium + vitamin D supplements. Because of his physical handicaps, our exercise physiologists carefully reviewed with him and his caregivers important concepts of energy balance and designed upper body aerobic workout routines that he could regularly use. Two years after RYGB, his weight had decreased from 117.9 to 52.4 kg, corresponding to a BMI of 26.9 kg/m^2. Despite his physical limitations, he was able to lose weight at a rate of 4.8% per month during the initial 6 months postoperatively.

He also demonstrated dramatic improvement in insulin resistance and dyslipidemia (Table 1) and symptomatic improvement in gastroesophageal reflux. Systolic and diastolic blood pressures fell by 16 and 10 mm Hg, respectively, for the first year. Medications for dyslipidemia and hypertension were discontinued, and elevated fasting glucose normalized. Remarkably, 2 years after RYGB, his polysomnogram indicated complete resolution of OSAS (Table 1) and dramatic improvement in sleep hygiene. He did not require BiPAP postoperatively. He also reported great improvements of his quality of life, including higher levels of energy, and he was better able to independently conduct activities of daily living.

2. Discussion

Spina bifida is a complex deformity that occurs as a result of the neural tube failing to close normally in the embryological period; bladder dysfunction, hydrocephalus, and lower extremity paralysis are 3 major associated impairments. Neurologic paralysis results in motor disabilities, particularly in ambulation. Reduced physical activity translates into a 10% to 20% reduction in energy expenditure [13]. In our current "obesogenic" environment, if energy intake exceeds expenditure, the biologic result is increased adipose tissue mass. Indeed, obesity is seen in approximately 50% of patients with spina bifida [14], and adiposity correlates with age and neurologic level [15]. Hence, this patient's disability likely promoted persistent energy imbalance.

Since the date of operation for the adolescent described in this case report, another case of a paraplegic adult who underwent RYGB was reported. In the adult case report, a 31% reduction in BMI was documented 1 year after RYGB (BMI reduced from 48 to 33 kg/m^2), with improvement in medical conditions associated with obesity [16], similar to the outcomes we describe.

However, our report extends the findings from the adult case report, particularly related to the analysis of body composition and bone mineralization. There is a positive relationship between BMI and age-adjusted bone mineral content [17], and there may be a trend to normative reduction in total bone mineral content with significant fall in BMI, due simply to the reduced mechanical load on the skeleton. In fact, in our cohort of ambulatory boys, we see that pronounced weight loss is accompanied by significant reduction of BMC as well as of bone mineral density (BMD), but these values remain in the reference range over time. On the contrary, paraplegia alone results in compromised BMD compared with normative data from ambulatory populations. Indeed, Quan et al [18] reported that BMD in the patients with spina bifida was 1 to 2 standard deviations below the mean of an ambulatory population, very similar to the BMD z score of −2.8 at 6 months and −2.5 at 2 years that we measured in our patient. Similarly, low BMD results have been seen in other nonambulatory full-time wheelchair users [18], and this discrepancy likely reflects the effect of varying degrees of physical inactivity on bone metabolism. Importantly, in our case, the total BMD showed no decrease between the first measurement at 6 months postoperatively and the 2-year measurement, despite significant weight loss, suggesting adequacy of calcium and vitamin D supplementation. Patients with spina bifida are expected to experience peak bone mineralization around the age of 19 years [18]. Thus, longer-term DEXA follow-up measurements in this patient are needed, given the fact that RYGB is an independent risk factor for hypovitaminosis D and impaired calcium absorption.

The survival rate of patients with spina bifida has improved over the years, and it is estimated that from 50% to 70% of children born with spina bifida today will survive into adulthood [19]. Unfortunately, development of extreme obesity coupled with severe comorbidities such as OSAS and risk factors for cardiovascular disease threaten to reverse this survival advantage. This case study illustrates that application of a standard surgical weight loss procedure by a multidisciplinary clinical management team can result in safe reversal of cardiovascular risk factors, metabolic disorders, and disordered breathing during sleep associated with severe obesity in a paraplegic adolescent. Furthermore, the improvement in quality of life and potential for self-care and independent living during transition to adulthood support the appropriateness of surgical treatment of severe obesity in adolescents with paraplegia.

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