Surgery for Obesity and Related Diseases ■ (2023) 1–9

SURGERY FOR OBESITY AND RELATED DISEASES

Review article

Metabolic and bariatric surgery for obesity in Prader Willi syndrome: systematic review and meta-analysis

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Received 3 April 2022; accepted 21 January 2023

Abstract Obesity is the leading cause of morbidity and mortality in patients with Prader-Willi Syndrome (PWS). Our objective was to compare changes in body mass index (BMI) after metabolic and bariatric surgery (MBS) for the treatment of obesity (BMI $\geq$35 kg/m^2) in PWS. A systematic review of MBS in PWS was performed using PubMed, Embase, and Cochrane Central, identifying 254 citations. Sixty-seven patients from 22 articles met criteria for inclusion in the meta-analysis. Patients were organized into 3 groups: laparoscopic sleeve gastrectomy (LSG), gastric bypass (GB), and biliopancreatic diversion (BPD). No mortality within 1 year was reported in any of the 3 groups after a primary MBS operation. All groups experienced a significant decrease in BMI at 1 year with a mean reduction in BMI of 14.7 kg/m^2 ($P < .001$). The LSG groups (n = 26) showed significant change from baseline in years 1, 2, and 3 (P value at year 3 = .002) but did not show significance in years 5, 7, and 10. The GB group (n = 10) showed a significant reduction in BMI of 12.1 kg/m^2 in the first 2 years (P = .001). The BPD group (n = 28) had a significant reduction in BMI through 7 years with an average reduction of 10.7 kg/m^2 (P = .02) at year 7. Individuals with PWS who underwent MBS had significant BMI reduction sustained in the LSG, GB, and BPD groups for 3, 2, and 7 years, respectively. No deaths within 1 year of these primary MBS operations were reported in this study or any other publication. (Surg Obes Relat Dis 2023;■:1–9.)

Keywords: Prader-Willi syndrome; Metabolic and bariatric surgery; Weight loss; Children; Adolescents

Prader-Willi syndrome (PWS) is a genetic disorder from a lack of expression of genes on the paternally inherited chromosome 15q11.2-q13 region resulting in an accumulation of numerous endocrine abnormalities. PWS is the most

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https://doi.org/10.1016/j.soard.2023.01.017
1550-7289/

common genetic cause of life-threatening obesity [1]. The exact mechanism driving obesity in PWS is unknown although numerous hormones are hypothesized to play a role, such as increased ghrelin and a deficiency in growth hormone (GH) [2]. Treatment with recombinant growth hormone (rhGH) in PWS has been shown to improve body composition [3]. Along with rhGH use, much of the current management of obesity in PWS involves dietary restriction and lifestyle management, including behaviorally focused and mental health interventions. There are also ongoing studies looking at pharmacologic interventions in PWS to manage obesity and hyperphagia [2,4–6].

Despite these treatment options severe obesity and its associated co-morbidities continue to plague many patients with PWS [1]. The use of metabolic and bariatric surgery (MBS) has been effective in providing significant weight loss and resolving co-morbidities in non-syndromic adults and adolescents, but MBS has been poorly studied in the PWS population [7]. We performed a systematic review of the literature on MBS use within the PWS population and conducted a meta-analysis to determine the change in body mass index (BMI) following the 3 most common MBS operations: laparoscopic sleeve gastrectomy (LSG), gastric bypass (GB), and biliopancreatic diversion (BPD).

Methods

Literature search

A librarian-mediated systematic review of the literature was completed in July 2022, which incorporated publications extending back to 1974. Publications on outcomes of MBS performed on patients with PWS were the focus of the systematic review using keywords described in the appendix. PubMed (including Medline Complete), Embase, and Cochrane Central databases were searched in compliance with PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) 2020 guidelines [8]. Two authors independently screened each record, reviewed each report, retrieved, and collected the data. Only English-written articles in peer reviewed journals were accepted, including single-case reports. Unpublished series and abstracts were not included.

Inclusion and exclusion criteria for the meta-analysis

To be included in the meta-analysis each patient had to have a diagnosis of PWS, baseline BMI of 35 kg/m^2 or greater with a minimum follow up of 12 months following an MBS operation. Publications were excluded from the meta-analysis if the weights were recorded without the accompanying height to enable the BMI calculations or if there were multiple different operations recorded but the BMI outcomes could not be associated with a particular operation thus making it impossible to place the patient into the appropriate operative group (Table 1) [10,11,13,14,17,36]. Meta-analysis of Observational Studies in Epidemiology (MOOSE) reporting guidelines for meta-analysis were followed (Fig. 1) [37].

Patients who met criteria for the meta-analysis were grouped into 4 groups based on procedure, which were (1) LSG, (2) GB including Roux-en-Y gastric bypass (RYGB) and one-anastomosis gastric bypass (OAGB), (3) BPD including BPD with duodenal switch (DS), and (4) "other". The 3 patients in the "other" group met criteria but were too few and had MBS operations that are no longer used, including jejunoileal bypass and vertical banded gastroplasty, so they were not included in the meta-analysis.

BMI change

Each article was reviewed and BMI data were collected as available at baseline and 1, 2, 3, 5, 7, and 10 years on patients that met criteria for the meta-analysis. Individual weight data was collected for each patient with PWS and these individual BMI data points at each follow-up year were used to calculate the respective BMI. This was also done for percent excess weight loss.

Although in the pediatric population the BMI% over the 95th percentile is more accurate than the raw BMI, this information was not available in any of the reports meeting criteria for the meta-analysis, and the raw BMI was used.

Co-morbid conditions

Co-morbidities were not reported or incomplete in 60% of the articles included in the meta-analysis. In addition, the methods for determining the diagnosis and remission of co-morbidities were inconsistent across numerous publications. Therefore, conclusions and statistical analysis on co-morbidity resolution following MBS is not possible.

Statistical analysis

Analysis of variance (ANOVA) single factor was used to determine a significant difference in baseline demographic characteristics between groups. ANOVA was also utilized to evaluate whether there were differences in BMI between groups (LSG versus GB versus BPD) at baseline and each follow-up year. A paired Student *t* test was used to compare BMI at each follow-up period to the baseline BMI for each patient of the respective group. Data analysis was limited by the decreasing number of patients with follow-up data from 5 years and onward. A probability value (*P* value) less than .05 was determined to be statistically significant.

Results

Systematic review

The librarian-mediated systematic review yielded 256 publications. Of these, 28 publications were appropriate for data extraction and 22 contained data that met criteria for the meta-analysis (Fig. 1). From these publications a

Table 1
Systematic review results including all publications reporting MBS operation on individuals with PWS

Study and publication year	Total patients with Prader-Willi syndrome, n	Patients included in meta-analysis, n	Primary procedure	Maximum follow-up	Mortality within 1 yr of operation, n	Risk of bias	
						Outcome 1	Outcome 2
Randolph et al. 1974 [9]	1	1	JIB	1 yr	0	Low	Critical
Soper et al. 1975 [10]	7	0	Gastric bypass, gastroplasty	N/A	0	Low	Critical
Anderson et al. 1980 [11]	11	0	10 gastric bypass, 1 gastroplasty	N/A	0	Low	Critical
Touquet et al. 1983 [12]	1	1	JIB	1 yr	0	Low	Critical
Sliber et al. 1986 [13]	3	1	JIB	N/A	2	Low	Critical
Brossy et al. 1989 [14]	1	0	BPD	N/A	0	Low	Critical
Miyata et al. 1990 [15]	1	0	VBG	3 yr	0	Low	Moderate
Laurent-Jaccard et al. 1991 [16]	3	1	BPD	4 yr	0	Low	Moderate
Dousei et al 1992 [17]	1	1	VBG	5 yr	0	Low	Critical
Chelala et al 1996 [18]	1	1	AGB	N/A	1	Low	Critical
Antal et al. 1996 [19]	2	0	BPD	2 yr	0	Low	Moderate
Grugni et al. 2000 [20]	1	1	BPD	3 yr	0	Low	Severe
Marinari et al. 2001 [21]	15	15	BPD	10 yr	0	Low	Moderate
Kobayashi et al. 2003 [22]	1	1	RYGB	1.5 yr	0	Low	Moderate
De Almeida et al. 2005 [23]	2	2	BPD	2 yr	0	Low	Moderate
Papavramidis et al. 2006 [24]	1	1	BPD/DS	1.5 yr	0	Low	Moderate
Till et al. 2008 [25]	1	1	LSG	1 yr	0	Low	Moderate
Marceau et al. 2010 [26]	3	3	DS	2 yr	1	Low	Moderate
Fong et al. 2012 [27]	3	3	LSG	2 yr	0	Low	Moderate
Yu et al. 2013 [28]	1	1	LSG	1 yr	0	Low	Moderate
Musella et al. 2014 [29]	3	3	OAGB	2 yr	0	Low	Moderate
Michalik et al. 2015 [30]	2	2	BPD	2 yr	0	Low	Severe
Alqahtani et al. 2016 [31]	24	19	LSG	5 yr	0	Low	Moderate
Cazzo et al. 2018 [32]	1	1	BPD	1 yr	0	Low	Moderate
Liu et al. 2019 [33]	5	5	2 LSG, 1 RYGB, 2 OAGB	10 yr	0	Low	Moderate
Martinelli et al. 2019 [34]	1	1	LSG	1 yr	0	Low	Severe
Tripodi et al. 2020 [35]	2	2	OAGB	2 yr	0	Low	Moderate
Hu et al. 2022 [36]	6	0	2 LSG, 3 RYGB, LSG-DJB	5 yr	0	Low	Critical

JIB = jejunoileal bypass; N/A = not applicable; BPD = biliopancreatic diversion; VBG = vertical banded gastroplasty; AGB = adjustable gastric banding; RYGB = Roux-en-Y gastric bypass; DS = duodenal switch; LSG = laparoscopic sleeve gastrectomy; LSG-DJB = laparoscopic gastric band-duodenal jejunal bypass; OAGB = one-anastomosis gastric bypass; MBS = metabolic and bariatric surgery; PWS = Prader-Willi Syndrome.

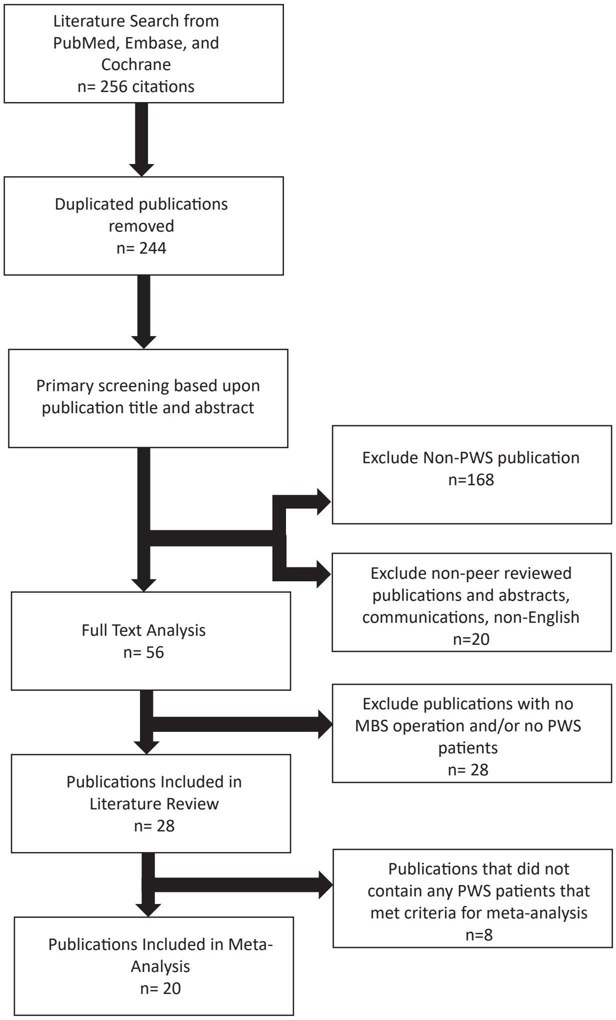

Fig. 1. Systematic review and meta-analysis: article selection.

total of 104 individuals with PWS that had MBS were discovered, of whom 67 (64%) met criteria for inclusion in our meta-analysis.

Meta-analysis: baseline characteristics

The age range of the patients with PWS included in the meta-analysis at the time of MBS was 5 to 40 years with 52% (33 patients) aged 17 years or younger. The 3 groups were statistically different in their baseline age. The average age of the LSG group was 12.8, the GB group was 18.4, and the BPD group was 21.4 years ($P < .01$). There was no significant difference in the baseline BMI (kg/m^2) between the 3 groups ($P = .12$). Our meta-analysis consisted of 56% male patients. Ethnicity and race of the patients with PWS were not recorded in most of the reviewed publications.

Meta-analysis: change in BMI

At the 1 year follow-up period individuals with PWS had a reduction of 14.9 kg/m^2, 11.4 kg/m^2, and 15.5 kg/m^2 for the LSG, GB, and BPD groups, respectively (Table 2).

The LSG group (n = 26) had a significant reduction in BMI through 3 years of follow-up with a reduction of 15.2 kg/m^2 from baseline at 3 years ($P = .002$). The gastric bypass group (n = 10) had the fewest number of patients with PWS and had significant reduction in BMI up to 2 years of follow-up with a reduction of 12.1 kg/m^2 ($P = .001$). The BPD-DS group (n = 28) had the most patients that followed up past 5 years and had significant reduction in BMI up until 7 years of follow-up with an average reduction of 10.7 kg/m^2($P = .02$) at year 7 (Table 2).

Overall, statistical analysis was limited by the loss of patients in the follow-up years 5, 7, and 10. However, the BPD-DS group seemed to confer the most weight loss in the long term with a reduction in BMI of 10.7 and 10.9 kg/m^2 at years 7 (n = 10) and 10 (n = 6), respectively. While the LSG group had weight gain with an increase in BMI of .3 and .8 kg/m^2 at years 7 (n = 2) and 10 (n = 2), respectively (Fig. 2).

Mortality and revisions

There was no surgical mortality or revisions in any of the groups in the meta-analysis within 1 year of the MBS operation (Table 1). In the BPD group, 3 of 28 had revisions between the 2- and 4-year follow-ups. One of these revisions occurred at 2 years for excessive weight loss and 2 for recurrence of severe obesity at 2 and 4 years. No other revisions were reported.

Surgical complications

There were no surgical complications reported in the LSG or GB groups. The BPD group (n = 28) had 3 cases of ventral hernias most likely related to open operations and 1 small bowel obstruction.

Nutritional complications

Nutritional complications were often not reported and, therefore, is unlikely to be an accurate reflection of nutritional deficiencies. However, there was 1 case of iron deficiency reported in the LSG group and 1 in the BPD group and 2 cases of osteoporosis reported in the BPD-DS group. There were no reported nutritional or surgical complications in the GB group.

Discussion

Postoperative mortality

Mortality from MBS is attributable to an operation if it occurs within the first year following the operation. In the recorded literature on individuals with PWS, there has not been a single death reported in the first year following a primary LSG, GB, or BPD, which encompasses publications included in our meta-analysis and those that were not included (Table 1).

Table 2
BMI and %EWL of individuals with PWS

Procedure	Total patients	Mean age	Baseline BMI	BMI 1 yr	2 yr	3 yr	5 yr	7 yr	10 yr	%EWL 1 yr	2 yr	3 yr	5 yr	7 yr	10 yr
LSG	26	12.8	51.0 ± 12	36.0 ± 9	33.7 ± 8	33.1 ± 9	41.7 ± 13	53.0 ± 6	53.6 ± 7	66.1	71.8	73.1	49.3	−1.3	−2.6
			P value	**<.001**	**<.001**	**.002**	.15	.9	.9						
			ΔBMI	-14.9	-16.3	-15.2	-11.2	0.3	0.8						
			n = 26	26	20	12	7	2	2	26	20	12	7	2	2
BPD/BPD-DS	28	21.4	55.2 ± 10	39.1 ± 8	39.4 ± 9	38.0 ± 8	40.2 ± 10	40.5 ± 8	38.8 ± 9	54.7	56.7	55.5	49.3	42.4	47.8
			P value	**<.001**	**<.001**	**<.001**	**.002**	**.02**	.13						
			ΔBMI	-15.5	-16.1	-15.0	-13.5	-10.7	-10.9						
			n = 28	22	21	16	14	10	6	22	21	26	14	10	6
OAGB/RYGB	10	18.4	47.7 ± 4	34.2 ± 5	35.5 ± 6	33.8 ± 3	36.7 ± 3	39.2	43.7	60.0	60.7	64.9	51.3	26.1	2.7
			P value	**.001**	**.001**	.06	.12	>.99	>.99						
			ΔBMI	-11.4	-12.1	-9.8	-6.9	-5.0	-0.5						
			n = 10	7	7	3	3	1	1	7	7	3	3	1	1
Total average	64	17.4	52.3 ± 10	37.0 ± 9	36.4 ± 9	35.7 ± 8	40.2 ± 10	42.3 ± 8	42.6 ± 10	60.1	61.7	61.1	46.4	31.0	26.4
			P value	**<.001**	**<.001**	**<.001**	**<.001**	**.02**	.16						
			ΔBMI	-14.7	-15.6	-14.6	-12.0	-8.6	-7.1						
			n = 64	55	48	31	24	13	9	55	48	31	24	13	9

BMI = body mass index; %EWL = percent excess weight loss; LSG = laparoscopic sleeve gastrectomy; BPD = biliopancreatic diversion; BPD-DS = biliopancreatic diversion with duodenal switch; OAGB = one-anastomosis gastric bypass; RYGB = Roux-en-Y gastric bypass; PWS = Prader-Willi Syndrome.
Bold *P*-values show significance (<.05).

However, there were 3 postoperative deaths that occurred within 1 year following MBS operations in outdated procedures that are no longer performed. Two deaths were reported in 1985 after jejunoileal bypass: 1 patient died of pulmonary embolism and the other patient died after perinephric infection [13]. In 1997, Chelala et al. reported the only adjustable gastric band placed for the treatment of obesity in an individual with PWS. Unfortunately, this resulted in a death from gastric bleeding 45 days after surgery [18].

Additionally, 4 other deaths have been reported in individuals with PWS following MBS, which occurred between 4 and 9 years after the surgery. These deaths should not be considered postoperative complications since they are related to progression of obesity characteristics in PWS. In 1980, Anderson et al. reported a death 4 years after a gastric bypass from congestive heart failure [11]. In 2001, Marinari et al. reported a death from respiratory failure 9 years after a BPD [21]. In 2010, Marceau et al. reported a mortality following a revision of a BPD/DS 4 years after the primary operation for recurrence of severe obesity, which resulted in death 53 days after the revisional operation from septicemia [26]. Hu et al. also reported a death 6 years after an MBS operation but it is unclear which operation was performed [36].

One death followed an endoscopically placed intragastric balloon [38]. This is not an MBS operation and is therefore not included in this review and meta-analysis.

Our review found no report of surgical mortality within the first year after a primary LSG, GB, or BPD in the PWS population. This is different from the publications of Gantz et al. [39] and Scheimann et al. [40], which reported high mortality rates after MBS but included outdated operations from decades ago and deaths that occurred many years after and unrelated to the operation itself.

Comparison of 1-year weight loss to patients without PWS

The 2020 Metabolic and Bariatric Surgery Accreditation and Quality Improvement Program (MBSAQIP) data showed an average 1-year BMI reduction of 11.3 kg/m^2, 14.1 kg/m^2, and 14.3 kg/m^2 for LSG, RYGB, and BPD, respectively. Compared with our meta-analysis, individuals with PWS had a reduction of 14.9 kg/m^2, 11.4 kg/m^2, and 15.5 kg/m^2 for LSG, GB (RYGB and OAGB), and BPD, respectively.

Safety

MBS has become much safer over the last 10 to 15 years in part because of the advent of minimally invasive surgery including laparoscopy and robotic surgery which decreases the risks of wound infection, small bowel obstruction, respiratory complications, and mortality [7]. Also, newer operations such as the LSG and modifications

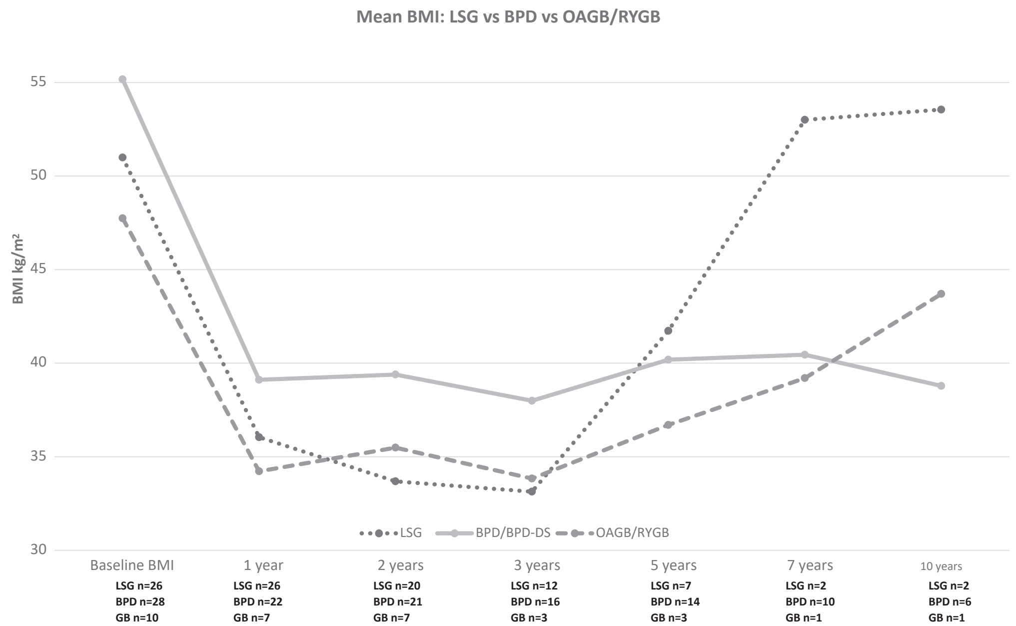

Fig. 2. Statistical analysis of the body mass index (BMI). There is no statistical significance between the baseline BMI or BMI at any follow-up period between any of the 3 groups.

of previous operations such as GB and BPD have contributed to the lower morbidity and mortality [7].

Of course, MBS operations are not only restrictive but are now known to result in changes in circulating gastrointestinal hormones affecting the hypothalamus and include decreasing ghrelin, increasing GLP-1 and PYY, which all contribute to the weight loss [27,31].

Cognitive impairment

Although some level of cognitive impairment is usually associated with PWS, recent guidelines no longer considered this a contraindication for MBS in children and adolescents suffering with severe obesity [41,42]. Nevertheless, careful, and thoughtful consideration must be given to the unique risks and possible benefits before deciding to proceed with MBS for an individual with PWS. An ethical framework for pediatric MBS evaluation has been developed by Moore et al. that can be helpful in guiding the family and surgical team in deciding whether MBS is appropriate for an individual adult or pediatric patient with PWS [43].

Change in BMI

In this meta-analysis, we found significant weight loss in the LSG, GB, and BPD groups until years 3, 2, and 7, respectively. The total average of all 64 patients with PWS that underwent MBS had significant BMI reduction through 7 years of follow-up data. At the 7-year follow-up there was a total of 14 patients with PWS and 12 (86%) of these patients had some degree of weight loss from their baseline weight. Nonetheless, understanding the long-term efficacy of these procedures is not possible since the data beyond 5 years are sparse.

Although we agree that the initial weight loss in the 5 years following a bariatric procedure in patients with PWS is not guaranteed to be sustained, unlike Scheimann et al., we do not see this as a justification for not offering MBS in this population [40]. The natural progression of PWS is a tendency toward weight gain as patients age [44]. Since obesity is the main driving force causing morbidity and premature mortality in patients with PWS, slowing down the magnitude of weight gain is critical. Patients with PWS that have uncontrolled obesity live, on average, 30 years less than those with proper weight control [1]. Thus, we believe a slowing of overall weight gain is likely to provide these patients with a valuable increase in life expectancy and quality of life. Remission or improvement of obesity-related co-morbidities, even if the weight loss is not maintained in the long term, may be of value. Despite some weight regain in the years following MBS both the patient and their family report an improved quality of life [21,23]. When analyzing the efficacy of MBS in patients with PWS, these patients should not be compared with patients unaffected by PWS since the mechanism of obesity and natural progression of weight regain are very different.

Hyperphagia

The hyperphagia experienced in PWS decreases the quality of life for the patient and their family. The level of hyperphagia can be systemically assessed by questionnaire [45].

High ghrelin levels are likely to be responsible for some of the observed hyperphagia seen in PWS. LSG has been shown to reduce hyperphagia in patients with PWS. Fong et al. followed the ghrelin levels after LSG and found a significant reduction, which was still present 3 years after surgery. Clinically, the patients with PWS in the Fong et al. publication also reported reduced appetite, which mirrored their reduction in ghrelin [27]. Since 70% of Ghrelin is produced by the stomach, the decrease in Ghrelin is likely related to the partial gastrectomy performed in the LSG which is also performed in the BPD-DS [46].

Selection of a weight loss procedure

It may be of value to speculate about the ideal procedure in patients with PWS. The operation would provide control of hyperphagia, significant short- and long-term weight loss, minimal surgical complication rates, and low but manageable nutritional risks. In our data collection, LSG appears to address several of these items, but a significant decrease in BMI may not be sustained past 3 years. Although our data are underpowered to statistically support any conclusions beyond 5 years, a BPD or BPD-DS may sustain a significant decrease in BMI out to 10 years, but requires greater compliance with nutritional supplementation. One strategy could be to perform the LSG for adolescents with severe obesity from PWS. Since the LSG is the first half of a BPD-DS the intestinal component could be added, if necessary, in adulthood converting the LSG to a BPD-DS if obesity recurs. The rational is by adding the intestinal component to the LSG, the BPD-DS provides additional hypo-absorption by decreasing the length of the small intestine that comes in contact with food and results in a mean threshold of absorption of 1600 calories per day regardless of the calories consumed, thus, decreasing the risk and/or severity of obesity recurrence [47].

As anti-obesity medications including GLP-1 receptor agonists continue to improve, there is likely to be also a role for combining surgery and pharmacotherapy to help some patients with PWS suffering with severe obesity to obtain their best result.

Limitations

There are many limitations to this study. The most glaring is the small number and the poor quality of the articles that report MBS performed on patients with PWS. Several articles could not be included in our meta-analysis because the specific operation could not be associated with weight data [10,11,13,14,17,36]. This meta-analysis is also limited by the small numbers of patients, poor quality and inconsistent long-term follow-up, and variability in the age between groups. In addition, 1 surgeon contributed to most of the LSG patients (73%), and they were primarily from a Middle Eastern background [31]. Therefore, the LSG group may not accurately represent the true diversity and age distribution of the PWS population. Nonetheless, all individuals with a baseline BMI of 35 kg/m^2 or greater that met criteria regardless of age were included in the meta-analysis. If this group was not included the LSG cohort would be too small for analysis.

Statistical analysis was severely limited by the small number of patients, especially at the 7- and 10-year follow-up periods. The GB group had the fewest number of individuals with PWS and thus any conclusions for this group at any follow-up period may be confounded by selection bias and may not be representative of the actual PWS population.

Risk of bias

There were no randomized studies of MBS of individuals with PWS identified. The risk of bias for each article was assessed using the Cochrane Risk of Bias in Non-Randomized Studies of Interventions (ROBINS-I) tool for mortality outcomes within the first year following MBS and separately for outcomes related to change in BMI [48].

First, a low risk of bias was identified in all publications reporting the outcome of postoperative mortality at 1 year following MBS in PWS (Appendix Table 3). Second, a moderate risk of bias was identified for the outcome of BMI change in publications included in the meta-analysis (Appendix Table 3). This is primarily due to confounding variables such as the inconsistent reporting of use of rhGH, the inconsistent reporting of co-morbidities, the lack of reporting of race and ethnicity, missing data, and the variation of mean age in each group.

Conclusion

Despite growth hormone treatment, pharmacotherapy, and aggressive lifestyle and behavioral modification, severe obesity is still the major cause of rapid deterioration and death in patients with PWS [1]. The current data on MBS in patients with PWS show that procedures performed today such as the LSG, GB, and BPD with or without DS can safely provide rapid weight loss and alter the natural progression of weight gain seen in these patients. It may indeed be time to take a new look at MBS for patients with PWS [49].

However, the poor quality and paucity of the articles in MBS as treatment for severe obesity in PWS highlights the need for high quality, prospective data collection. Although there are registries for PWS such as the Italian National Registry and the Global Prader-Willi Syndrome Registry, to our knowledge there is no registry that tracks long-term outcomes following MBS for patients with PWS [44,50]. A PWS surgical registry would facilitate more rigorous and standardized data collection including demographics, genetics, operation performed with details of any variation, baseline and yearly follow-up weight and co-morbidities, standardized diagnosis of co-morbidities, timing of complications, revisions, changes in behavior such as the level of hyperphagia, and quality of life indicators. Future longitudinal studies should include these elements as well. Ethically, it is important for these patients to be followed for decades and not just years. MBS operations on patients with PWS are infrequent enough that enrolling each patient with PWS is feasible. Such a registry would be useful in prospectively obtaining data to determine the optimal care for the patients with PWS. It may be reasonable to collaborate with the developers of the Global PWS Registry to include a surgical component as described here.

Acknowledgments

The authors acknowledge Alexandra Short for her assistance in the librarian-mediated search, Tom Wasser, Ph.D., for statistical support, and Lisa Barrow, R.N., C.N.S., for acquisition of MBSAQIP 2020 data.

Disclosures

The authors have no commercial associations that might be a conflict of interest in relation to this article.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at https://doi.org/10.1016/j.soard.2023.01.017.

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