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Anesthesia management for patients with Prader-Willi syndrome undergoing bariatric surgery: a single-center retrospective case series study

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Abstract

Background Prader-Willi syndrome (PWS) is a rare neurodevelopmental disorder resulting from abnormalities on chromosome 15q11.2-q13. These genetic anomalies pose significant challenges in anesthetic management when PWS patients undergo bariatric surgery.

Methods We present five instances of anesthetic management in three PWS patients who underwent bariatric surgery under general anesthesia supplemented with nerve block techniques.

Results Obesity, sleep apnea, airway ventilatory dysfunction, and hypotonia were the primary challenges faced by PWS patients in our study. We implemented specific strategies, primarily including the reverse Trendelenburg position, gradually deepening sedation, multimodal analgesia and perioperative progressive respiratory exercises. Only in case 1a, respiratory obstruction occurred during mask ventilation, which was resolved through the use of a nasopharyngeal ventilation tract. Additionally, delayed awakening was observed in case 1a postoperatively, with the spontaneous breathing showing minimal recovery following the administration of neostigmine and atropine. Extubation of the tracheal tube was performed on the first postoperative day. Upon her second admission (case 1b), we administered sugammadex as the neuromuscular blockade reversal agent, which facilitated successful tracheal extubation ten minutes post-procedure.

Conclusions We advocate the use of sugammadex as the neuromuscular blockade reversal agent, the implementation of neuromuscular monitoring, progressive respiratory exercises, and multimodal analgesia in PWS patients undergoing bariatric surgery.

Keywords Prader-willi syndrome, Anesthesia, Airway management, Bariatric surgery

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Introduction

Prader-Willi syndrome (PWS) is a rare neurodevelopmental disorder resulting from the absence of imprinted genes at 15q11.2–q13, typically due to paternal deletion of this region (65–75% of individuals), maternal uniparental disomy 15 (20–30%), or an imprinting defect (1–3%) [1]. The prevalence of PWS is estimated to be between 1 in 10,000 and 30,000 individuals [2]. A hallmark clinical feature of PWS is a progressive alteration in eating behaviors that correlates with age [3]. Affected individuals often develop obesity secondary to hypothalamic dysfunction-related hyperphagia and gain weight easily due to decreased satiety and limited physical activity [4, 5].

Despite increasing fat masses, lung volume remains stable in weight-gaining adults with PWS [6]. However, the risk of pulmonary complications, such as respiratory failure, hypoxia, and apnea, is markedly elevated in morbidly obese patients, particularly those with obstructive sleep apnea (OSA) [7]. The incidence of difficult tracheal intubation has been reported to be 13–24%, whereas difficult face mask ventilation is observed in 79% of obese patients [8, 9]. Reduced lung compliance and functional residual capacity (FRC) lead to increased work of breathing, oxygen consumption, and carbon dioxide production, thereby significantly impairing tolerance to hypoxia [6]. A high incidence of atelectasis even 24 h after general anesthesia predisposes patients to arterial desaturation in the postoperative period [10]. Patients with PWS exhibit hypotonia, characteristic orofacial dysmorphic features (e.g., poor dentition, micrognathia, palatal abnormalities), viscous saliva and airway secretions [11–13]. Consequently, PWS patients with obesity pose a greater challenge in perioperative airway management compared to other obese patients.

Furthermore, PWS patients are also susceptible to cardiovascular complications, hypothalamic dysfunction (e.g., temperature dysregulation, endocrine dysfunctions), a high pain threshold, mental retardation, temper tantrums, compulsive traits, short stature, and skin picking [3, 11, 12, 14]. These factors further complicate perioperative anesthesia management. Here, we report a case series involving PWS patients who underwent bariatric surgery under general anesthesia combined with nerve block, aiming to provide a reference for the anesthesia management of such cases.

Method

Patient collection

This study was a single-center retrospective analysis conducted at our institution. The Institutional Review Board approved this study (approval number: 2024-S-146). Informed consent was obtained from all patients and their guardians. All patients included in the study were diagnosed with PWS through genetic testing. We collated data from three cases of PWS patients who underwent successful bariatric surgery. Two of these patients underwent surgery twice, resulting in a total of five instances of perioperative anesthesia management. These cases were sequentially designated as case 1a, case 1b, case 2a, case 2b, and case 3. Case 1b was readmitted due to postoperative Gastroesophageal Reflux Disease (GERD) following case 1a, and case 2b was readmitted due to weight regain (WR) following case 2a.

Data collection

Patient data were extracted from the case information system (Union Digital Medical Record browser, version 2012.4) and the surgical anesthesia information management system (Docare Anesthesia Clinical Information System, version 5.0). The data extraction included demographics (including gender, age, height, weight, and diagnosis), auxiliary examination results, preoperative anesthesia evaluations, anesthesia management records, postoperative complications, and patient figures.

Results

Preoperative evaluation and therapy

All patients presented with obesity, OSA, hypotonia, intellectual developmental disorder, and metabolic syndrome. Some individuals also had hypertension, diabetes, bronchitis, arrhythmia, and other diagnoses. The demographic characteristics and clinical manifestations are detailed in Table 1. Preoperative evaluation revealed that all patients exhibited airway ventilatory dysfunction, distinctive orofacial dysmorphic features, a shortened and thickened neck, Mallampati class III, and short stature. The specifics of pulmonary function assessments and polysomnography are outlined in Table 1. Prior to surgery, patients underwent lung function training and continuous positive airway pressure (CPAP) therapy. The CPAP settings were adjusted to a pressure of 12 mmH_2O, with an oxygen concentration of 30%, and a sustained inflation time of 10 min. CPAP was initiated upon hospital admission and continued until the resolution of hypercapnia. All patients received prophylactic antithrombotic therapy with low molecular weight heparin (LMWH). The preoperative status of all patients is presented in Fig. 1.

Monitoring

In the operating room, electrocardiogram, oxygen saturation, invasive blood pressure (in the radial artery) and bispectral index (BIS) were continuously monitored. However, arterial puncture catheterization in case 1a was unsuccessful due to difficulties in puncture and limited vascular selection. Case 1a experienced challenges with

Table 1 Demographic characteristics and clinical features

Patient	Sex	Age (years)	Height (m)	Weight (kg)	BMI (kg/m^2)	Pulmonary function tests	Polysomnography	Other diagnoses	Challenge during anesthesia management
Case1a	female	16	141	97	48.79	Moderate restricted ventilation dysfunction, small airway ventilation dysfunction	Mild sleep apnea syndrome	Bronchitis, diabetes, arhythmia, abnormal liver function, hiatal hernia, hypoplasia of uterus, hyperuricemia, hyperlipidemia	Hypotonia, airway secretions, small airway ventilatory dysfunction, poor blood glucose control
Case1b	female	18	141	73.9	37.7	Severe airflow obstruction in the large and small airways	Moderate sleep apnea syndrome	Reflux esophagitis, gallstone with cholestasis, mild regurgitation of tricuspid valve, sinus arrhythmia, hypoplasia of uterus	Hypotonia, airway ventilatory dysfunction
Case2a	female	22	154	137	57.8	Moderate restricted ventilation dysfunction, small airway ventilation dysfunction	Moderate sleep apnea syndrome	Restrictive ventilation dysfunction, gallstone, hyperuricemia, sinus tachycardia	Hypotonia, restrictive ventilatory dsyfunction, small airway ventilatory dysfunction
Case2b	female	23	154	139	57.85	Moderate restricted ventilation dysfunction, small airway ventilation dysfunction	Moderate sleep apnea syndrome	Diabetes, heart enlargement, liver insufficiency, sleep disorders, hyperuricemia, gallstones	Hypotonia, poor blood glucose control, moderate pulmonary arterial hypertension, small airway ventilatory dysfunction
Case3	male	17	160	120	46.8	Mild restrictive ventilatory dysfunction	Moderate sleep apnea syndrome	Bronchitis, diabetes, hypertension, left atrial enlargement, reflux esophagitis, hyperlipidemia, lung nodules and cyst	Hypotonia, airway secretions, poor blood glucose control, small airway ventilatory dysfunction

venipuncture, including failed peripheral vein puncture, necessitating central venous catheterization (CVC).

Anesthesia induction and artificial airway establishment

Patients were positioned in a reverse Trendelenburg position (with the head elevated and feet lowered) to improve ventilation conditions and reduce the risk of reflux aspiration. Concurrently, they were administered a high flow of oxygen (6–10 L/min) via a face mask. Prior to anesthesia induction, the oropharyngeal or nasopharyngeal ventilatory tract was prepared. A loading dose of 0.5 μg/kg dexmedetomidine was administered within 10 min, based on standard weight (female: standard weight = height-105 cm, male: standard weight = height-100 cm) with vigilant monitoring of respiratory function. Ten mg of dexamethasone, 2 mg of midazolam, 1 mg/kg of lidocaine, 50 mg of flurbiprofen axetil, and 0.1–0.3 μg/kg of sufentanil were administered, with dosing based on standard weight.

As sedation levels increased, only case 1a experienced ventilation obstruction which was resolved through the use of a nasopharyngeal ventilation tract. The controlled plasma infusion concentrations of propofol and remifentanil were set to 1–4 μg/ml and 2–6 ng/ml respectively, adjusted according to the hemodynamics and the depth of anesthesia. Concurrently, 0.6 mg/kg rocuronium was intravenously administered to complete anesthesia induction, with dosing based on adjusted weight, (adjusted weight = standard weight + 0.4× (actual weight-standard weight)).

Tracheal intubation was successfully performed using a video laryngoscope. The post-intubation status of the patients is shown in Fig. 2. The dimensions of the tracheal tube are detailed in Table 2. Mechanical ventilation was managed using lung-protective strategies, including low tidal volume (6–8 ml/kg based on standard weight), intermittent pulmonary recurrent ventilation, individualized positive end expiratory pressure (PEEP), and low inspired oxygen concentration (< 60%).

Maintenance of anesthesia and intraoperative details

Following endotracheal intubation, bilateral transverse abdominal fascia plane (TAP) and rectus sheath nerve blocks were performed under ultrasound guidance.

Fig. 1 PWS patients before surgery
Note Severe obesity, orofacial dysmorphic features (micrognathia, narrow bifrontal diameter, almond-shaped palpebral fissures, downturned corners of the mouth), a short and thickened neck, short stature were present

Sufentanil or esketamine, along with rocuronium were administered prior to incision, with dosing based on standardized weight. Anesthesia was maintained using a combination of intravenous agents and inhalational anesthesia. BIS was maintained between 40 and 60 through real-time adjustment of anesthetic agents. Prior to gastric resection, patients were administered 0.15 mg/kg of oxycodone for adequate analgesia (based on standardized weight) and 5 mg of tropisetron. The details of the surgical procedure are outlined in Table 2.

Anesthesia recovery

After surgery, patients were repositioned to the reverse Trendelenburg position. They were monitored for spontaneous breathing and recovery of consciousness. And the neuromuscular blockade reversal agent was administered. After their consciousness and autonomous respiration were recovered completely, case 1b, case 2a, case 2b, case 3 all successfully extracted the tracheal tube. Case 1a experienced delayed awakening, with autonomous respiration recovering 150 min after entering the PACU, characterized by shallow and rapid breathing (respiratory rate of 38 breaths per minute). Administration of neostigmine and atropine did not significantly improve her spontaneous breathing. She was admitted to the intensive care unit (ICU) with endotracheal intubation. On the first postoperative day, her consciousness and breathing fully recovered, and the tracheal tube was successfully removed. During her second surgery (case 1b), sugammadex was used as the neuromuscular blockade reversal agent, facilitating successful tracheal extubation 10 min post-surgery.

All patients received aggressive chest physiotherapy and other respiratory adjuncts for pulmonary toilet upon returning to the ward. These measures included external diaphragmatic pacing, nebulizer inhalation, and CPAP therapy, aimed at preventing atelectasis and pulmonary infections. All patients had an uneventful postoperative course, free of complications, and were discharged successfully.

Discussion

For PWS patients combined with obesity, airway management poses a significant challenge for anesthesiologists. However, obesity alone or BMI is not a predictive factor for difficult intubation in obese patients. Instead, factors such as decreased mandibular mobility, male gender, OSA, snoring, and a thickened neck are predictive of a difficult airway [15].

Some studies have recommended conducting sleep breathing monitoring before surgery [16, 17]. During the preoperative evaluation, we conducted a comprehensive assessment of airway conditions, including mouth opening, Mallampati classification, mandibular mobility, neck circumference, OSA, hypotonia, cervical mobility, respiratory infections, airway secretions, and dental condition. The reverse Trendelenburg position is advocated as the standard position for patients with obesity, as it can significantly improve conditions for intubation [18] and provide the longest safe apnea period [19, 20]. During induction, patients may face a risk of airway obstruction and difficult ventilation. In our study, airway patency was evaluated by gradually increasing sedation levels in conjunction with a manual positive pressure ventilation test. Only case 1a experienced airway obstruction during mask ventilation which was resolved after the application of a nasopharyngeal ventilatory tract.

Additionally, the accumulation of lipophilic volatile anesthetics in adipose tissue can lead to prolonged emergence from anesthesia, potentially exceeding 2–4 h [21]. When considering the use of inhaled anesthetics for morbidly obese patients, it is advisable to prioritize anesthetics with relatively lower lipid solubility, such as sevoflurane or desflurane [22]. Numerous studies have suggested the use of multimodal analgesia to reduce the total quantity of opioid use, thereby preventing postoperative respiratory complications [6, 7, 18]. We applied TAP

Fig. 2 PWS patients during surgery
Note Severe obesity, orofacial dysmorphic features, a short and thickened neck, microcheiria and skin picking were present

Table 2 Intraoperative ventilation, surgical details and antagonistic muscle relaxants condition

Patient	Mask ventilation	Tracheal intubation	Date of surgery	Procedure	Agonist drug	Tracheal extubation
Case1a	Difficult The nasopharyngeal ventilation tract was used	7.5 enhanced tracheal tube	June 15, 2018	Laparoscopic sleeve gastrectomy and esophageal hiatal hernia repair and abdominal wall plastic surgery	1 mg neostigmine 0.5 mg atropine	Bring the tracheal intubation to the ICU
Case1b	Not difficult	7.5 enhanced tracheal tube	November 18, 2019	Laparoscopic gastric bypass and cholecystectomy	100 mg sugammadex	Ten minutes after the procedure, the tracheal tube was removed
Case2a	Not difficult	8.0 enhanced tracheal tube	April 27, 2022	laparoscopic sleeve gastrectomy and jejunojejunostomy	2 mg neostigmine 0.5 mg atropine	Fifteen minutes after the procedure, the tracheal tube was removed
Case2b	Not difficult	8.0 enhanced tracheal tube	November 8, 2023	laparoscopic gastric volume reduction and double-channel anastomosis	1 mg neostigmine 0.5 mg atropine 0.5 mg flumazenil	Seventeen minutes after the procedure, the tracheal tube was removed
Case3	Not difficult	7.5 enhanced tracheal tube	January 11, 2023	Laparoscopic sleeve gastrectomy and jejunojejunostomy and bowel arrangement	2 mg neostigmine 0.5 mg atropine 0.25 mg flumazenil	Twelve minutes after the procedure, the tracheal tube was removed

and rectus sheath block, along with PCIA, to provide multimodal analgesia. To our knowledge, no anesthesiologists have reported using this combined anesthesia method in patients with PWS in the published literature.

Proactive measures to enhance respiratory status should be implemented throughout the perioperative period. In our study, all patients underwent CPAP to improve their ventilation status before surgery. Nixon et al. recommended CPAP for the treatment of OSA [23]. Adequate pre-oxygenation with 100% oxygen was essential prior to anesthesia induction. Some researchers have suggested that using a CPAP of 10 cm H_2O can achieve higher PaO_2 levels after intubation and reduce atelectasis formation [7]. Besides, other researchers have recommended incentive spirometry or chest physiotherapy to improve pulmonary function and decrease complications after surgery [24, 25].

All patients had hypotonia associated with a higher risk of reflux aspiration and delayed recovery. The obese patients had limited lung reserve, which may exacerbate the consequences of aspiration [26]. The reverse Trendelenburg position, securing the airway promptly and gastrointestinal decompression can reduce the morbidity and mortality associated with inhalation of gastric contents [26].

The use of Neuromuscular blocking agents (NMBAs) should be carefully titrated in PWS patients with hypotonia. Studies have indicated that the effects of nondepolarizing NMBAs are prolonged in these patients, particularly in infants [27]. There have been case reports of delayed recovery in these patients, leading to decreased or no use of NMBAs due to this issue [28–31]. However, several reports have demonstrated the safe use of various nondepolarizing NMBAs, including pancuronium, atracurium, vecuronium, and rocuronium, without evidence of prolonged effects [26, 32–34]. Some cases have reported that good reversal with NMBAs is possible [35, 36].

Delayed recovery of muscle strength occurred in case 1a, and the improvement was not appreciable following the administration of neostigmine and atropine. Consequently, for her second surgery, we opted for sugammadex as a reversal agent for NMBAs. At 10 min postoperatively, she regained muscle strength, and the tracheal tube was successfully removed. Neostigmine functions by inhibiting cholinesterase competitively and is not suitable for profound blocks due to the ceiling effect [37]. In contrast, sugammadex can encapsulate and inactivate unbound aminosteroid muscle relaxants, exhibiting superior effects in reversing muscle relaxation and reducing the risk of residual paralysis [38]. The advantage of sugammadex is more than just shortening neuromuscular recovery duration regardless of the degree of the blockade [39]. Research has indicated that immediate reversal of neuromuscular blockade can be achieved using sugammadex at a dose of 2 mg/kg + 40% (based on standard weight) [40], regardless of the depth of neuromuscular blockage [41].

Due to the unavailability of specific equipment at our institution, neuromuscular blockade monitoring was not conducted for any patients. A review article emphasized the importance of monitoring neuromuscular function when administering NMBAs, both in anesthesia and intensive care unit. Monitoring should be objective and quantitative, rather than clinical, qualitative, and subjective [42]. However, a cross-sectional survey on the status of current management of NMBAs and monitoring in China revealed that only 10.2% of anesthesiologists reported the use of neuromuscular function monitors and 6.59% of respondents indicated their use of relevant monitors in the operating room in China [43]. If neuromuscular monitoring equipment is available, it is highly recommended for use. Furthermore, it is advised that endotracheal extubation should only be performed after a comprehensive assessment of the patient's consciousness and muscle strength has been conducted.

Innovation and limitations

In this study, we introduce a novel series of anesthetic management strategies for PWS patients undergoing bariatric surgery, highlighting the innovation of using sugammadex as a reversal agent for neuromuscular blockade and the implementation of multimodal analgesia to mitigate the risks in the perioperative period. However, our study is not without limitations. Due to the unavailability of specific equipment at our institution, neuromuscular blockade monitoring was not conducted for any patients.

Conclusion

Given the low prevalence of PWS and the associated inexperience, patients with PWS undergoing bariatric surgery present significant anesthesia challenges. The risk of developing a difficult airway should be carefully assessed during the preoperative evaluation. The oropharyngeal or nasopharyngeal ventilatory tract should be prepared before anesthesia induction. Additionally, we recommend the use of sugammadex as a neuromuscular blockade reversal agent, along with the concurrent implementation of neuromuscular monitoring, aggressive respiratory function exercises, and multimodal analgesia to minimize opioid use. We advocate for the removal of the endotracheal tube only after a comprehensive assessment confirms the complete recovery of patient consciousness and muscle strength.

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Author contributions

A was responsible for ethical approval, data collation, writing and revision of the article. B assisted with the revision of the article. C assisted with data collation, and obtained informed consent. D assisted with data collation, and obtained informed consent. E was responsible for the revision of the article. F assisted with the revision of the article.

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Data availability

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

The institutional review board of the Third People's Hospital of Chengdu approved this study, the approval number: 2024-S-146. We obtained informed consent from the patients and guardians of the patients at the time of admission.

Consent for publication
We obtained informed consent from the patients and guardians of the patients for publication at the time of admission.

Competing interests
The authors declare no competing interests.

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