Case Report
pISSN 2383-9309 ▮ eISSN 2383-9317
J Dent Anesth Pain Med 2026;26(2):175-180 ▮ https://doi.org/10.17245/jdapm.2026.26.2.175

General anesthesia management for oral surgery in a patient with plastic bronchitis associated with Fontan circulation: a case report

Kaho Noda[1], Yukiko Nishioka[2], Hitoshi Higuchi[2], Fumika Hashimoto[1], Saki Miyake[1], Takuya Miyawaki[1]

[1]Dental Anesthesiology and Special Care Dentistry, Okayama University Graduate School of Medicine, Dentistry and Pharmaceutical Sciences, Okayama, Japan
[2]Department of Dental Anesthesiology, Okayama University Hospital, Okayama, Japan

Plastic bronchitis is a rare condition in which mucus plugs obstruct the bronchi, potentially leading to fatal respiratory failure. It has been reported in some patients with congenital heart disease following Fontan surgery. We report the general anesthesia management of a 22-year-old female patient with Fontan circulation and type II plastic bronchitis that was controlled using regular intravenous heparin injections. In this case, concerns existed regarding airway obstruction by bronchial plugs and hemodynamic instability specific to the Fontan circulation. During endotracheal intubation, the absence of mucus plugs was confirmed using a flexible bronchoscope. Intraoperatively, ventilation was managed at low pressure to avoid an increase in intrathoracic pressure caused by high positive-pressure ventilation. Additionally, fluid overload was avoided to prevent elevations in the central venous pressure. Consequently, perioperative management can be safely performed without any respiratory or circulatory complications. As treatment outcomes improve, the number of dental and oral surgical procedures in adult patients with congenital heart disease is expected to increase. Therefore, knowledge of congenital heart disease and its sequelae, such as plastic bronchitis, is essential to perform appropriate risk assessment and management.

Keywords: Anesthesia, General; Plastic Bronchitis; Fontan Procedure; Oral Surgical Procedures.

INTRODUCTION

Plastic bronchitis (PB) is a rare respiratory condition in which highly viscous mucus plugs obstruct the bronchi like a “cast.” Patients with PB present with wet cough and dyspnea, which can result in fatal airway obstruction [1]. Among pediatric patients with congenital heart disease, PB develops in approximately 4% who undergo Fontan surgery, a functional hemodynamic repair surgery performed for cyanotic congenital heart disease [2]. With advances in surgical and medical treatments, pediatrics life expectancy with heart disease has increased [3]. Consequently, administering general anesthesia for non-cardiac procedures, including dental and oral surgical procedures, in patients with PB are expected to increase.

However, reports on anesthesia in post-Fontan patients with history of PB remain scarce. We report general anesthesia management during oral surgery in an adult patient with Fontan circulation and PB.

Received: December 17, 2025 • Revised: February 25, 2026 • Accepted: February 28, 2026
Corresponding Author: Yukiko Nishioka, Department of Dental Anesthesiology, Okayama University Hospital, Okayama, Japan
Tel & Fax: +81-86-235-6721 E-mail: de421038@s.okayama-u.ac.jp

CASE REPORT

The patient was a 22-year-old woman (height: 122.7 cm, weight: 23.9 kg) who required extraction of the two lower left molars due to inflammation. However, due to neurodevelopmental disorders and dental anxiety, conventional treatment is difficult, and the procedure is scheduled under general anesthesia. The patient was diagnosed with hypoplastic left-sided heart syndrome at birth. At 12 days, 5 months, and 7 months of age, the patient underwent a modified Norwood surgery, percutaneous transluminal angioplasty for aortic stenosis, and Glenn surgery. At 1 year and 11 months of age, the patient underwent Fontan surgery. At the age of 2 years, she developed symptomatic epilepsy. At the age of 3 years, she developed pneumonia and experienced multiple symptom relapses. At 4 years of age, she was diagnosed with PB based on expectoration of mucus plugs and chest computed tomography (CT) scans (Fig. 1). Treatment was challenging; however, her symptoms improved after intravenous heparin administration and fluid restriction. Heparin was administered intravenously three times daily via a central venous catheter. Following regular intravenous heparin administration, the patient developed osteoporosis secondary to long-term heparin use and received intravenous bisphosphonates for limited period. The patient's PB condition remained stable. However, she experienced three episodes of mucus plug expectoration, with the most recent occurring six months before surgery. Preoperative examinations revealed no findings that could interfere with the surgery (Tables 1 and 2). Her peripheral oxygen saturation (SpO_2) was approximately 92% on room air.

After entering the operating room, the patient was highly anxious and uncooperative while lying in bed. Therefore, while wearing an SpO_2 monitor, midazolam 0.13 mg/kg was administered intravenously via her venous line while she remained seated in a wheelchair. After adequate sedation was achieved, she was assisted on the bed, an electrocardiogram monitor, a blood pressure cuff, and capnography were applied, and propofol 1.3 mg/kg was administered intravenously, confirming loss of consciousness. Oxygen was supplied at 6 L/min via a face mask, during which end-tidal carbon dioxide was

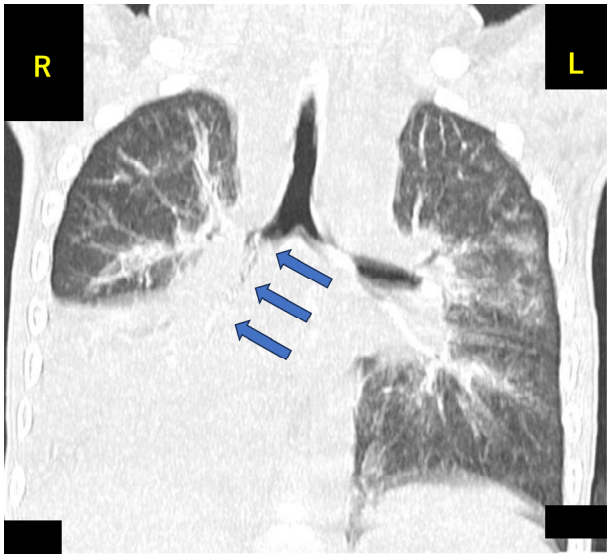

Fig. 1. CT image demonstrating PB that developed at age 4. An intraluminal soft-tissue density lesion suggestive of a mucus plug extended from the right main bronchus to the right lower lobe bronchus (blue arrow). L, left; R, right.

Table 1. Findings from various preoperative examinations

Examination	Findings	Normal range at our hospital
Complete blood count	Within normal limits	-
Serum biochemistry	APTT: 38.7 sec (prolonged); PT: 42 % (low); ALT: 36 U/L (elevated); γ-GTP: 207 U/L(elevated)	APTT: 24.0-34.0 sec; PT: 73-118 %; ALT: 7-23 U/L; γ-GTP: 9-32 U/L
Electrocardiography	Sinus tachycardia Heart rate: 114 bpm	-
Pulmonary function test	Restrictive ventilatory impairment (%VC 42.1%)	%VC > 80%
Chest radiography	No abnormal findings	-

APTT, activated partial thromboplastin time; PT, prothrombin time; ALT, alanine aminotransferase; γ-GTP, γ-glutamyl transpeptidase, %VC, percent vital capacity.

continuously monitored within the range of 20–25 mmHg and spontaneous respiration was maintained without airway obstruction. General anesthesia was induced with continuous intravenous administration of remifentanil at 0.3 μg/kg/min, inhalation of 3% sevoflurane, and a bolus of 0.67 mg/kg rocuronium. Before endotracheal intubation, the airway was examined using a flexible bronchoscope to confirm the absence of mucus plugs or secretions (Fig. 2A), followed by nasal intubation using a video laryngoscope. After intubation, normal breath sounds were auscultated bilaterally and no abnormalities were observed in the capnogram waveform. From the initiation of manual ventilation until the completion of endotracheal intubation, SpO_2 remained stable at approximately 99% under 100% oxygen. After intubation, mechanical ventilation was set in pressure-controlled mode with an inspiratory pressure of 11–12 cmH_2O and a positive end-expiratory pressure (PEEP) of 4 cmH_2O; end-tidal carbon dioxide was maintained at 30–36 mmHg, FiO_2 was set to 0.4, and SpO_2 was maintained at 96%–100%. Additionally, because of the patient's lack of cooperation, it was not possible to secure a venous line or administer prophylactic antibiotics for infective endocarditis before entering the operating room. Therefore, surgery was initiated only after the intravenous administration of cefazolin sodium following endotracheal intubation. During surgery, anesthesia was maintained with 1.5% sevoflurane and remifentanil at 0.1 μg/kg/min. Upon completion of surgery, the trachea was rechecked using a flexible bronchoscope (Fig. 2B). Sugammadex 2.1 mg/kg was administered intravenously and the endotracheal tube was removed once adequate spontaneous breathing and eye opening were achieved. The SpO_2 remained at approximately 95% in room air. Intraoperative parameters are summarized in Table 3. Surgery and anesthesia duration was 33 and 102 min, respectively. After returning to the ward, the patient's postoperative course remained uneventful. The patient was discharged 2 days postoperatively.

Table 2. Preoperative transthoracic echocardiographic findings

Parameter	Value / Unit
RVEDD	40.1 mm / 44.1 mm/m^2 (Indexed for BSA: 0.91 m^2)
RVESD	29.3 mm / 32 mm/m^2 (Indexed for BSA: 0.91m^2)
EF	53.1%
FS	26.9%
PV	17.5 mm
TV	22.4 mm
TR	moderate
AR	mild
Aortic arch flow velocity	1.4 m/s
SVC flow velocity	0.26 m/s
IVC flow velocity	0.14 m/s

Echocardiography revealed a mildly enlarged systemic right ventricle indexed to BSA, within acceptable limits, with preserved ejection fraction. Moderate TR was present, without significant vascular stenosis.
RVEDD, right ventricular end-diastolic diameter; BSA, body surface area; RVESD, right ventricular end-systolic diameter; EF, ejection fraction; FS, fractional shortening; PV, pulmonary valve; TV, tricuspid valve; TR, tricuspid regurgitation; AR, aortic regurgitation; SVC, superior vena cava; IVC, inferior vena cava.

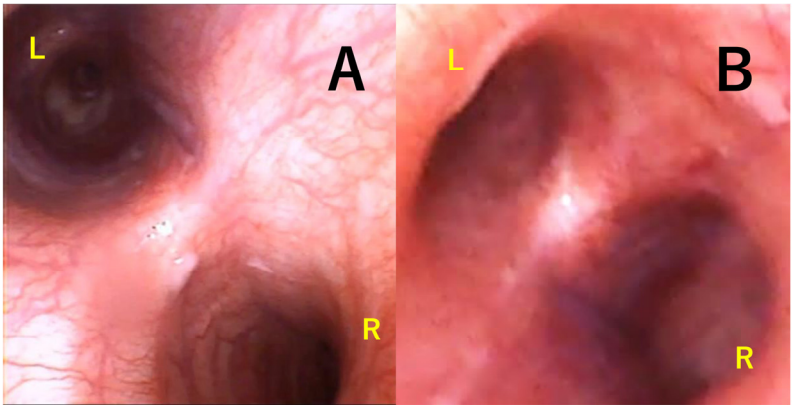

Fig. 2. Condition of the trachea. (A) Before intubation. (B) Before extubation. In either case, no mucus plug or secretion are presented. L, left; R, right.

Table 3. Values of various parameters in the operating room

	Time elapsed in the operating room (minutes)										
	0	10	20	30	40	50	60	70	80	90	100
SpO_2 (%)	93	99	99	96	99	99	99	99	99	99	100
HR (bpm)	-	129	122	113	108	106	105	105	104	106	97
BP (mmHg)	-	85/63	74/54	71/51	76/48	71/47	73/50	77/51	71/46	70/47	79/49
$EtCO_2$ (mmHg)	-	24	35	30	31	32	33	36	36	36	49
Ppeak (cmH_2O)	-	-	-	16	16	16	16	16	16	16	-
PEEP (cmH_2O)	-	-	-	4	4	4	4	4	4	4	-
RR (min^{-1})	-	20	39	12	10	10	10	10	10	10	15
		×	T			◎				◎	T×

SpO_2, peripheral oxygen saturation; HR, heart rate; BP, blood pressure; $EtCO_2$, end-tidal carbon dioxide; Ppeak, peak airway pressure; PEEP, positive end-expiratory pressure; RR, respiratory rate. ×, start and end of anesthesia; ◎, start and end of surgery; T, intubation and extubation.

DISCUSSION

The primary anesthetic concern in patient was respiratory failure and circulatory collapse risk from mucus plug obstruction occur due to PB. There are two types of PB: type I, from inflammatory reactions, such as asthma or infection, and type II, associated with congenital heart disease, such as after Fontan surgery [1]. Type II is considered to result from impaired lymphatic drainage into the thoracic duct due to elevated central venous pressure in Fontan circulation, leading to lymphatic fluid leakage into the bronchi and subsequent mucus plug formation [1]. The pathogenesis of PB remains largely unclear. Unlike typical chronic bronchitis or asthma, β-agonists were reportedly ineffective [4,5]. Although treatments such as steroids, heparin, urokinase, and tissue plasminogen activator (tPA) have shown efficacy [2,6], no consistently effective pharmacological therapies were established [7]. PB associated with congenital heart disease is considered prone to severe progression. There are reports of pediatric patients who developed PB following congenital heart surgery, including cases complicated by cardiac arrest as cases successfully managed with extracorporeal membrane oxygenation [8]. Lymphatic embolization is a novel and effective surgical treatment option [1,2].

In patients with PB and Fontan circulation, mechanical ventilation may further elevate central venous pressure due to increased intrathoracic pressure from high-positive-pressure ventilation, potentially accelerating mucus plug formation. Moreover, positive pressure ventilation may propel mucus plugs distally into the peripheral airways [9], posing ventilatory failure risk due to peripheral bronchial obstruction. The patient was diagnosed with PB at age 4 and received various treatments, including steroids and inhaled tPA; however, these were ineffective. Intravenous heparin administration resulted in symptom improvement. Mucus plugs in PB are believed to form when excessively activated eosinophils undergo specialized cell death, extracellular trap cell death (ETosis), releasing highly viscous reticular DNA from their nuclei [10]. Heparin was suggested to promote DNA degradation by deoxyribonuclease (DNase), thereby potentially reducing mucus viscosity and hydrophobicity. This mechanism is believed to underlie the therapeutic effects of intravenous heparin on PB [11]. However, in this case, mucus plug formation occurred multiple times despite regular heparin administration; therefore, we considered the aforementioned risks to be elevated during general anesthesia.

Furthermore, in Fontan circulation, systemic venous return flows directly into the pulmonary arteries; therefore, hemodynamics are largely determined by pulmonary blood flow and pulmonary vascular resistance. Maintaining adequate pulmonary blood flow and low pulmonary vascular resistance is essential for preserving

optimal Fontan circulation [12]. Morgan et al. [13] reported that increased intrathoracic pressure reduces venous return by decreasing the pressure gradient between the peripheral veins and heart, leading to reduced cardiac output. In Fontan circulation, the pulmonary vascular bed is directly connected to the systemic circulation, increasing ventricular afterload. Combined with reduced venous return due to elevated intrathoracic pressure, causing circulatory failure. Therefore, during positive-pressure ventilation in patients with Fontan circulation, it is essential to prevent atelectasis, hypoxemia, and hypercapnia, while avoiding excessive inspiratory pressure and PEEP to maintain low intrathoracic pressure.

Considering the above, manual ventilation at low airway pressure was performed after anesthesia induction. During endotracheal intubation, the trachea was carefully examined using a flexible bronchoscope to confirm the absence of mucus plugs or secretions. During surgery, respiratory management was guided by continuous end-tidal carbon dioxide monitoring to prevent hypoventilation, with ventilation parameters set to low-pressure mode (respiratory rate, 10–12 breaths/min; peak airway pressure < 16 cmH_2O). Prior to extubation, the trachea was re-examined using a flexible bronchoscope to confirm the absence of mucus plugs or secretions.

Fluid management is equally important. Patients with Fontan circulation require adequate preload [14]; however, excessive fluid administration may increase the central venous pressure, potentially promoting mucus plug formation. Therefore, we carefully avoided fluid overload and administered vasopressors to counteract hypotension during the procedure. Stable perioperative respiratory and hemodynamic management was achieved without the development of hypoxemia during or after surgery.

In our case, no perioperative respiratory complications related to mucus plugs occurred. However, previous reports indicated that when respiratory failure due to mucus plugs develops during mechanical ventilation, spontaneous expectoration tends to improve the condition [9]. Therefore, during general anesthesia, prepare conventional bronchodilators and reversal agents, such as sugammadex and naloxone, is important. Furthermore, although PB was generally well-controlled in this case, procedure postponement should be considered if active mucus plug expectoration is observed immediately preoperatively. Finally, thorough preoperative evaluation and multidisciplinary consultation are essential when administering general anesthesia to patients with PB.

In conclusion, we successfully administered general anesthesia in a patient with PB and Fontan circulation. As outcomes improve, adult patients undergo dental and oral surgical procedures, increasing demand for non-cardiac surgery general anesthesia. Given PB is a rare disease, awareness of this condition as a postoperative complication of Fontan surgery remains limited, and undiagnosed cases may exist [15]. Therefore, when administering general anesthesia to patients after Fontan surgery, anesthetic management should consider the potential coexistence of PB. Adequate knowledge of congenital heart disease and its complications is essential for appropriate risk assessment and perioperative management.

AUTHOR ORCIDs

Kaho Noda: https://orcid.org/0009-0006-5977-9612
Yukiko Nishioka: https://orcid.org/0000-0002-6264-5694
Hitoshi Higuchi: https://orcid.org/0000-0002-8600-7793
Fumika Hashimoto: https://orcid.org/0009-0007-0914-2311
Saki Miyake: https://orcid.org/0000-0002-9222-7872
Takuya Miyawaki: https://orcid.org/0000-0002-6378-2623

AUTHOR CONTRIBUTIONS

Kaho Noda: Writing - original draft, Writing - review & editing
Yukiko Nishioka: Conceptualization, Writing - original draft, Writing - review & editing
Hitoshi Higuchi: Conceptualization, Writing - review & editing
Fumika Hashimoto: Data curation, Writing - review & editing
Saki Miyake: Data curation, Writing - review & editing
Takuya Miyawaki: Conceptualization, Supervision, Writing - review & editing

DECLARATIONS OF INTERESTS: The authors declare no conflict of interest.

FUNDING: Not applicable.

STATEMENT INFORMED CONSENT AND INSTITUTIONAL REVIEW BOARD APPROVAL: Written informed consent was obtained from the patient's guardian to report the case. This case report was approved by the Okayama University Graduate School of Medicine, Dentistry, and Pharmaceutical Sciences, and the Okayama University Hospital Ethics Committee (approval No. KEN 2602-001).

ARTIFICIAL INTELLIGENCE DECLARATION: The authors used AI-assisted tools solely for language editing and grammar correction. All scientific content, interpretations, and conclusions were developed entirely by the authors.

REFERENCES

1. Patel N, Patel M, Inja R, Krvavac A, Lechner AJ. Plastic bronchitis in adult and pediatric patients: a review of its presentation, diagnosis, and treatment. Mo Med 2021; 118: 363-73.
2. Dori Y, Keller MS, Rome JJ, Gillespie MJ, Glatz AC, Dodds K, et al. Percutaneous lymphatic embolization of abnormal pulmonary lymphatic flow as treatment of plastic bronchitis in patients with congenital heart disease. Circulation 2016; 133: 1160-70.
3. Triedman JK, Newburger JW. Trends in congenital heart disease: the next decade. Circulation 2016; 133: 2716-33.
4. Muller AF, Mithal A. A case of plastic bronchitis. J R Soc Med 1988; 81: 360.
5. Hess NR, Piercecchi C, Desai N, Fisher MR, Lee EH, Force SD. Successful thoracic duct ligation for plastic bronchitis in an adult. Ann Thorac Surg 2017; 103: e539-40.
6. Coen M, Daniel L, Serratrice J. An adult case of plastic bronchitis: a rare and multifactorial disease. J Thorac Dis 2018; 10: E16-9.
7. Colaneri M, Quarti A, Pozzi M, Gasparini S, Carloni I, de Benedictis FM. Management of plastic bronchitis with nebulized tissue plasminogen activator: another brick in the wall. Ital J Pediatr 2014; 40: 18.
8. Brogan TV, Finn LS, Pyskaty DJ Jr, Redding GJ, Ricker D, Inglis A, et al. Plastic bronchitis in children: a case series and review of the medical literature. Pediatr Pulmonol 2002; 34: 482-7.
9. Nakamoto H, Kayama S, Harada M, Honjo T, Kubota K, Sawamura S. Airway emergency during general anesthesia in a child with plastic bronchitis following Fontan surgery: a case report. JA Clin Rep 2020; 6: 6.
10. Yoshida M, Miyahara Y, Orimo K, Kono N, Narita M, Ohya Y, et al. Eosinophil extracellular traps in the casts of plastic bronchitis associated with influenza virus infection. Chest 2021; 160: 854-7.
11. Miyabe Y, Fukuchi M, Tomizawa H, Nakamura Y, Jikei M, Matsuwaki Y, et al. Aggregated eosinophils and neutrophils characterize the properties of mucus in chronic rhinosinusitis. J Allergy Clin Immunol 2024; 153: 1306-18.
12. Liang F, Senzaki H, Kurishima C, Sughimoto K, Inuzuka R, Liu H. Hemodynamic performance of the Fontan circulation compared with a normal biventricular circulation: a computational model study. Am J Physiol Heart Circ Physiol 2014; 307: H1056-72.
13. Morgan BC, Martin WE, Hornbein TF, Crawford EW, Guntheroth WG. Hemodynamic effects of intermittent positive pressure respiration. Anesthesiology 1966; 27: 584-90.
14. Luppes VAC, Willems A, Hazekamp MG, Blom NA, Ten Harkel ADJ. Fluid overload in pediatric univentricular patients undergoing Fontan completion. J Cardiovasc Dev Dis 2023; 10: 156.
15. Caruthers RL, Kempa M, Loo A, Gulbransen E, Kelly E, Erickson SR, et al. Demographic characteristics and estimated prevalence of Fontan-associated plastic bronchitis. Pediatr Cardiol 2013; 34: 256-61.