



CASE REPORT

Anesthetic management for thoracic surgery in Rubinstein–Taybi syndrome



E. Blazquez^{a,*}, D. Narváez^a, A. Fernandez-Lopez^a, L. Garcia-Aparicio^b

^a Anesthesiology, Critical Care and Pain Management Department, Hospital Universitario Virgen Macarena, University of Seville, Spain

^b Hospital Sant Joan de Déu, University of Barcelona, Spain

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KEYWORDS

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Abstract Rubinstein–Taybi syndrome (RTS) is a chromosomopathy associated to molecular mutations or microdeletions of chromosome 16. It has an incidence of 1:125,000–700,000 live births. RTS patients present craniofacial and thoracic anomalies that lead to a probable difficult-to-manage airway and ventilation. They also present mental retardation and comorbidity, such as congenital cardiac defects, pulmonary structural anomalies and recurrent respiratory infections, which increase the risk of aspiration pneumonia. Cardiac arrhythmias have been reported after the use of certain drugs such as succinylcholine and atropine, in a higher incidence than in general population. There is an increased risk of postoperative apnea-hypopnea in these patients.

We report the anesthetic management in a RTS patient undergoing emergent thoracic surgery due to oesophageal perforation and mediastinitis. Lung isolation was achieved with a bronchial blocker guided with a fiberoptic bronchoscope and one-lung ventilation was performed successfully.

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PALABRAS CLAVE

Síndrome de Rubinstein–Taybi;
Cromosopatías;
Manejo de la vía aérea;
Ventilación unipulmonar;
Perforación esofágica

Manejo anestésico en cirugía torácica en el síndrome de Rubinstein-Taybi

Resumen El síndrome de Rubinstein-Taybi es una enfermedad de baja incidencia (1:125.000–700.000 RN vivos) asociada a mutaciones o microdeleciones del cromosoma 16. Los pacientes afectados presentan frecuentemente anomalías craneofaciales y torácicas que condicionan una vía aérea y ventilación dificultosas. Asimismo, asocian retraso mental y comorbilidades, entre las que cabe destacar cardiopatías congénitas, infecciones respiratorias de repetición y enfermedad por reflujo gastroesofágico, que aumenta el riesgo de broncoaspiración. En estos

* Corresponding author.

E-mail addresses: eva_bg@msn.com, evablazquezgomez@hotmail.com (E. Blazquez).

pacientes se ha descrito una mayor incidencia de arritmias tras la administración de fármacos, como la succinilcolina y atropina, así como una mayor incidencia de apnea e hipopnea en el postoperatorio.

Presentamos el manejo anestésico de un paciente afecto de síndrome de Rubinstein–Taybi sometido a cirugía torácica urgente por perforación esofágica y mediastinitis, con exclusión pulmonar llevada a cabo con bloqueador bronquial.

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Introduction

The Rubinstein–Taybi syndrome (RTS), first described in 1963, has an incidence of 1:125,000–700,000 live births.¹ It is associated with microdeletions or pathogenic variants in the chromosome 16 and the genes *CREBBP* (50% of affected individuals) and *EP300* (3–8%). The most commonly described phenotype is a patient with short stature, mental retardation, craniofacial and skeletal anomalies, recurrent respiratory infections and congenital cardiac defects.^{1,2} Despite the low prevalence of RTS, these patients may require surgery to correct orthopedic, ophthalmic, dental or heart anomalies.¹ Most of the anesthetic implications of RTS stem from a difficult airway and ventilation. The anesthetic management is also conditioned by the common presence of lung disease and structural or functional heart disease.¹

Case report

A 26-year-old female who had the medical history of RTS required emergent thoracic surgery due to mediastinitis secondary to esophageal perforation after endoscopic removal of food bolus impaction. She revealed predictors of difficult intubation: Mallampati scoring class IV, 5 cm interincisor gap, mandibular hypoplasia, thyromental distance <6.5 cm, 80° head extension, short neck, high arched palate and obesity (BMI >25). Chest CT showed esophageal perforation, esophageal-pleural fistula, right hydropneumothorax, compressive atelectasis of the lower lobe, and pneumomediastinum (Fig. 1).

In the operating room the patient was hemodynamically stable, breathing spontaneously with a SaO₂ of 97% on room air. Due to the predicted difficult airway and the lack of cooperation of the patient we performed an inhalational induction with FiO₂ 100% and sevoflurane 86.4% maintaining spontaneous ventilation until we achieved an end-tidal concentration of 4%. We started remifentanyl continuous intravenous infusion 0.05 mcg/kg/min. A diagnostic direct laryngoscopy using Macintosh blade size 4 revealed Cormack III anatomy. We opened the patient's mouth and anesthetized the oropharynx with lidocaine 10% spray. To maintain the depth of anesthesia during the procedure we placed a nasopharyngeal tube and connected it to FiO₂/Air 100%/4 lpm and sevoflurane 4%. With the videolaryngoscope GlideScope® (Verathon Medical, Bothell, WA, USA) we improved the visualization to Cormack grade II. Surgery



Figure 1 Intubated patient with visible retrognathia and short neck. Bronchial blocker placed outside the tracheal tube.

required one-lung-ventilation (OLV). The materials available to achieve the OLV were double lumen tubes (DLT) sizes 37–41F and Coopdech bronchial blocker (BB) 9F, 60 cm, 9 mm ID (Daiken Medical Co., Ltd.). Pediatric material was not available and we were in an emergent clinical situation. We opted for a BB. The patient's narrow trachea only permitted a #6.5 tracheal tube (TT), but the diameter of the TT did not allow passing both fiberoptic bronchoscope (FOB) and BB at the same time. We selected a non-conventional way to insert the BB in this situation. We applied 1% lidocaine as airway topical anesthesia by a "spray-as-you-go" technique via the working channel of the FOB. We exposed the glottis via videolaryngoscopy and inserted the BB guiding it with the Olympus FOB OD 3.7 mm and directing it to the right main bronchus. Then we placed a #6.5 standard TT and administered rocuronium 1 mg/kg. We took out the nasopharyngeal tube and connected the TT to mechanical ventilation. Thus, we accomplished the placement of the BB outside the TT (Fig. 2).

Surgery was performed by a posterolateral right thoracotomy and consisted of a mediastinal drain and esophageal closure (Fig. 3). We used remifentanyl, sevoflurane and rocuronium boluses for maintenance of anesthesia. Volume controlled ventilation was programmed with a tidal volume 7 ml/kg, respiratory rate 14/min, time ratio 1:2, 5 mmHg PEEP, reaching a peak pressure of 22 mmHg and plateau pressure of 10 mmHg. Recruitment maneuvers were

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