



Anesthetic management of pediatric patients with large posterior mediastinal tumor – a systematic review and two case reports

Carlos Darcy A. Bersot^{1,2} , Heloísa Leal Pires³ , Lucas Ferreira Gomes Pereira^{2,4} ,
Rafael Mercante Linhares⁵ , Vitório Moscon Puntel⁶ , Vinicius Caldeira Quintão⁴ ,
Bruno José Martini Santos⁶ , Anna Carolina Fontoura Machado⁶ ,
Rodrigo Zillig Gac dos Santos⁶ , José Eduardo Guimarães Pereira¹ 

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ABSTRACT

Anesthetic management of mediastinal masses in pediatric patients is challenging for the anesthesiologist. There is abundant literature available on anesthesia management of anterior mediastinal masses. Posterior masses traditionally have been suggested to carry a low risk of anesthetic implications. However, our patient with a posterior mediastinal mass experienced hemodynamic and respiratory collapse typically associated with anterior mediastinal masses. The objective of this review was to evaluate the anesthetic management of children submitted to posterior mediastinum tumor resection and the respective postoperative outcomes. A systematic review was performed following PRISMA guidelines. Electronic databases were searched using multiple combinations of keywords to identify all case reports of patients undergoing mediastinal mass operation. Information such as clinical characteristics, perioperative management and patients' outcomes were abstracted and analyzed. Six case reports encompassing six patients aged from 2.5 months to 10 years old were included. Three of them were on the right side, located in the paravertebral space, and one of them presented intraspinal involvement. One had an esophagus mucocele, and the remaining two were on the left side, one presenting main left bronchus compression, and the other located in the paravertebral space. General anesthesia was used in all cases. Muscle relaxants were administered to all patients during anesthesia induction and controlled respiration was maintained for all of them. Only one case used succinylcholine as a muscle relaxant agent. The remaining five cases were all conducted using non-depolarizing agents. Extracorporeal circulation was not utilized in any of the cases. There were no patients experiencing cardiac arrest, or death during surgery, and all patients were discharged from the hospital. Perioperative management of patients undergoing mediastinal mass operations is challenging. It is imperative that a preoperative multidisciplinary discussion, well-planned anesthetic management, and pre-determined protocols for emergency situations are adequate to ensure patient safety.

KEYWORDS

Mediastinal neoplasm; ganglioneuroma; pediatric anesthesia

¹ Universidade Federal de São Paulo, Escola Paulista de Medicina, Programa de Pós-graduação em Medicina Translacional, São Paulo, São Paulo, Brasil

² Hospital BP Mirante, Departamento de Anestesiologia, São Paulo, São Paulo, Brasil

³ Hospital Unimed Volta Redonda, Departamento de Anestesiologia, Volta Redonda, Rio de Janeiro, Brasil

⁴ Universidade de São Paulo, Faculdade de Medicina, Hospital das Clínicas, Disciplina de Anestesiologia, São Paulo, São Paulo, Brasil

⁵ Hospital Municipal Miguel Couto, Departamento de Anestesiologia, Rio de Janeiro, Rio de Janeiro, Brasil

⁶ Hospital Unimed Volta Redonda, Departamento de Cirurgia Torácica, Volta Redonda, Rio de Janeiro, Brasil



INTRODUCTION

Posterior mediastinum tumors are a group of tumors that encompass schwannomas, neuroblastomas, and ganglioneuromas. Primary thoracic neuroblastoma represents approximately 14% of possible locations⁽¹⁾. It originates from the thoracic sympathetic ganglion chain and grows into the posterior mediastinum. Despite the proximity, airway displacement has rarely been reported⁽²⁾.

Ganglioneuromas are rare and categorized as neurogenic tumors, which are the most common tumors occupying posterior mediastinal masses in children⁽³⁾. They are benign and generally asymptomatic⁽⁴⁾. However, approximately 10% of patients with neurogenic tumors experience growth of the paravertebral mass through the intervertebral foramina with a significant portion of the tumor located intraspinally⁽⁵⁾.

Removal of these neoplasms risks potential spinal cord injury owing to the presence of the tumor adjacent to and within the spinal canal. The most common surgical approach is a postero-lateral thoracotomy, which provides good exposure and control of the mediastinum and ensures a safe tumor resection. Video-assisted thoracoscopic (VAT) surgery is a valuable approach for smaller lesions, and the patient benefits from minimal incisions and a lower rate of postoperative pain⁽⁶⁾. Integral to the minimally invasive approach is the use of one-lung ventilation (OLV). OLV technique has evolved significantly over the past 25 years, particularly with the widespread availability and routine use of several bronchial blockers⁽⁷⁻⁹⁾. Additionally, there has been a significant increase in the understanding and appreciation of the potential contributions of regional anesthetic techniques beyond local infiltration and neuraxial approaches to improving postoperative pain control.

Non-cardiac thoracic surgery in pediatric patients requires age or device-specific strategies to achieve one-lung ventilation. Due to the rarity of these cases, even in large children's hospitals, there are few, if any, prospective trials to guide the clinician in choosing an appropriate technique for one-lung ventilation. As a result, much of the available literature on this topic is based on case reports or smaller case series⁽⁸⁻¹²⁾. When thoracotomy is performed, anesthesia commonly involves general anesthetic drugs associated with different analgesia strategies. In contrast, in the field of thoracic surgery, relevant anesthetic techniques in children are advancing rapidly.

It is crucial to be aware of the potential complications, such as cardiac and respiratory arrest, which require immediate attention. Airway management may present challenges, so it is essential to allocate and prepare resources accordingly. Finally, neurophysiological

monitoring provides an additional layer of safety when caring for pediatric patients.

The objective of this review was to evaluate the anesthetic management of children submitted to posterior mediastinum tumor resection and the respective postoperative outcomes.

METHODS

The Cochrane Handbook for Intervention Reviews⁽¹³⁾ guided our choice of method. This review was registered in PROSPERO (International Prospective Register of Systematic Reviews) under CRD42023453781. We report our results following the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-analysis) statement⁽¹⁴⁾ (Appendix 1 – PRISMA Checklist).

Eligibility criteria

We considered all case reports about anesthetic management of children under 19 years old presenting with posterior mediastinum tumors and submitted to thoracotomy for tumor resection. The primary goal of this review was to evaluate the airway management technique of children submitted to posterior mediastinum tumor resection. Secondary outcomes were the following: pain management, hemodynamic monitoring, neurological monitoring, and anesthetic technique.

Eligible studies reported one or more of the outcomes listed above. The inclusion criteria were children under 19 years of age, posterior mediastinal mass, undergoing thoracotomy for tumor resection, and pulmonary ventilation. Exclusion criteria were tumor location other than the posterior mediastinum, patients aged 19 or over, and not submitted to thoracotomy for tumor resection.

Data source and searches

The search was performed in the following electronic databases: the Cochrane Central Register of Controlled Trials (CENTRAL, 2023)⁽¹⁵⁾ and PubMed (OvidSP, 2013 to 2023)⁽¹⁶⁾.

The databases were searched for published case reports, with the following filters: last ten years, humans, and under 19 years old. The last search was conducted on July 4, 2023. No restrictions were placed on language or publication status.

The search was conducted using multiple combinations of the following keywords: "mediastinum tumor" and "anesthesia" (Appendix 2 – search strategy). In addition, a search was conducted in PubMed using multiple combinations of the keywords "ganglioneuroma" and "anesthesia."

Selection of studies

All authors participated in conceptualization, data curation, formal analysis, investigation, methodology, project administration, resources, supervision, validation, visualization, and writing. Using standardized screening forms, two reviewers (JEGP, LFGP), independently screened all titles and abstracts identified by the literature search, obtained full-text articles of all potentially eligible studies, and evaluated these studies for eligibility. Reviewers resolved the disagreement through discussion.

RESULTS

We identified a total of 61 results in Cochrane and 2149 in PubMed. The additional search retrieved ten citations. After independent screening by title, and then by abstract, we obtained full-text copies of 15 articles. After duplicate removal, all 15 studies were potentially eligible for inclusion in the review.

Nine did not fulfill our eligibility criteria and were excluded (Appendix 1 - PRISMA flowchart). Therefore, we included six studies^(15,17-21) with a total of six participants in this review.

Ages ranged from 1 year and five months old^(14,18) up to 14 years old^(17,19). The patients presented respiratory symptoms in four cases⁽¹⁷⁻²⁰⁾. In the other two cases^(14,21), the patients had neurological symptoms, such as opsoclonus, ataxia, weakness, and diabetes insipidus.

OLV was the management choice in one case⁽¹⁸⁾, using a Fogarty catheter in a four-year-old patient. The other 5 cases^(14,17,19-21) were managed with endotracheal intubation.

Two case reports did not describe analgesia strategies^(17,19). Hosking et al.⁽¹⁸⁾ opted for an epidural catheter directly placed by the surgeon during the operation. Intravenous metamizole, ketorolac, and morphine were the strategy options for Maranhão et al.⁽¹⁴⁾ Rastogi et al.⁽²⁰⁾ used intercostal nerve block (ICNB), and Puri et al.⁽²¹⁾ opted for surgical site infiltration with local anesthetic associated with intravenous morphine and paracetamol.

Extubation occurred at the 20th hour in one case⁽¹⁹⁾, 48 hours in another⁽²⁰⁾, and on the 20th day in another one⁽²¹⁾. The remaining three cases did not describe when extubation took place.

Discharge from the hospital was not specified in the two case reports^(17,20). Hosking et al.⁽¹⁸⁾ reported discharge from the hospital on the 6th postoperative day, Maranhão on the⁽¹⁴⁾ 17th, Ahmed on the⁽¹⁹⁾ 10th and Puri et al.⁽²¹⁾.

After presenting the systematic review results and the data summarization in Table 1 above, we will describe the

case reports we conducted and highlight the challenging points of anesthetic management.

Case report 1

A four-year-old patient with no comorbidities, ASA 1, presented to the emergency department with upper airway infection symptoms. After an X-ray, a left posterior mediastinum mass was noted. Further investigation of the mass with magnetic resonance imaging (MRI) and computed tomography (Figure 1), a guided biopsy of the tumor of the thorax was performed, and a ganglioneuroma was diagnosed. Laboratory exams were routine.

Elective surgery was scheduled, and the patient was admitted to the hospital on the day of the surgery and taken to the surgical theater; standard monitors were placed: non-invasive blood pressure, SpO₂, EKG and EtCO₂. The patient was then pre-oxygenated via face mask, followed by inhalational induction to facilitate peripheral venous puncture. After the peripheral catheter (22G) was inserted, inhalational anesthetics were discontinued, and total intravenous anesthesia (TIVA) started with Propofol infusion using an open TCI system with the Schnider model, targeted for 4ng/dl and remifentanyl TCI, targeted for 3 ng/dl.

Intubation was performed under direct laryngoscopy, and a cuffed 4.0 orotracheal tube was introduced to the right main bronchus, and the position was confirmed by bronchoscopy. A vesical catheter was introduced. Ultrasound-guided (USG) the internal left jugular vein, which was cannulated with a double-lumen 5Fr catheter. The choice for the central line placement was made to keep the site on the same side as the thoracotomy.

The neurophysiologist then monitored the patient for motor and somatosensory evoked potentials, followed by patient positioning at the right lateral decubitus. Ringer Lactate solution was infused following an initial 10 ml/Kg bolus and subsequently kept at 100 ml/hr.

Tumor resection was performed by thoracoscopy and extraction of the tumor from the cavity was performed by video-assisted minimally invasive thoracotomy. The resection was uneventful and blood loss was kept to a minimum. The surgical team performed ropivacaine 0,5% into the intercostal nerve blocks at the incision of the intercostal spaces, port sites, and drain sites associated with tramadol and metamizole.

Neuromonitoring helped guide anesthesia depth and provided information regarding the integrity of the nervous system. Before the closure of the surgical site, the orotracheal tube cuff was deflated and retracted until the non-dependent lung expanded upon ventilation and the orotracheal tube cuff reinflated.

Table 1. Study Characteristics related to the description of intervention and outcome

Author	Country	Patients included age	Description of tumor and symptoms	Airway management strategy	Analgesia strategy	Measured outcomes
Hosking et al. ⁽¹⁸⁾	USA	4 years old	Posterior mediastinal mass adjacent to thoracic vertebrae T3-7 with spinal canal involvement Recurrent right upper lobe pneumonias.	Endotracheal intubation One lung ventilation accomplished by inflation of Fogarty catheter.	Epidural catheter directly placed by surgeon – 1,6 mg of Morphine.	No changes in amplitude or latency of the MEPs occurred during excision of the tumor. Patient discharged on the 6 th postoperative day.
Maranhão et al. ⁽¹⁴⁾	Brazil	1 year and 5 months old	Posterior mediastinal mass occupying the right paravertebral region at the level of the thoracic segments T3-T6. Opsoclonus, myoclonus, ataxia, and irritability.	Endotracheal intubation. Controlled ventilation with a tidal volume of 8ml/kg.	Intravenous Metimazole 30 mg/kg; Ketoprofen 2 mg/kg; Morphine 0,1 mg/kg.	Discharge from ICU on the 4 th day; and left the hospital on the 17 th postoperative day.
Tewari et al. ⁽¹⁷⁾	India	14 years old	Esophageal mucocele extending from D1-D7 vertebrae. Breathlessness.	Awake fiber-optic endotracheal intubation.	Not specified.	x
Ahmed et al. ⁽¹⁹⁾	Pakistan	10 years old	Left-sided mass, measuring 13.0×14.0×14.5 cm. Cough	Endotracheal intubation.	Not specified	Extubated after 20h and discharged from hospital after 10 days.
Rastogi et al. ⁽²⁰⁾	India	2.5 month	Posterior mediastinum mass. Cough.	Endotracheal intubation. Hand assisted/ PCV	ICNB	Extubated after 48h.
Puri et al. ⁽²¹⁾	India	9 years old	Left paravertebral Ganglioneuroma extending from T8-T12. Central Diabetes Insipidus, precocious puberty, autonomic dysfunction, retinal hemorrhages, palpitations, hypersomnolence and weakness of both lower limbs.	Endotracheal intubation. PCV - 14 cmH2O	Surgical site infiltration with Ropivacaine and intravenous Morphine 10 mg and Paracetamol 650 mg.	Extubated on the 20 th postoperative day and discharged home on the 29 th postoperative day.

After re-expansion, the non-dependent surgical sites were closed, and the patient awakened with no neurological deficits. Extubation occurred in the operating room and was performed after oropharyngeal aspiration; reflexes were recovered and breathing resumed. All vital signs

were within the normal range and the patient was taken to the ICU as per institutional protocol.

Admission into the ICU was uneventful. Drainage was below 150ml in the first 24 hours and the patient was discharged to the ward on the first postoperative day.

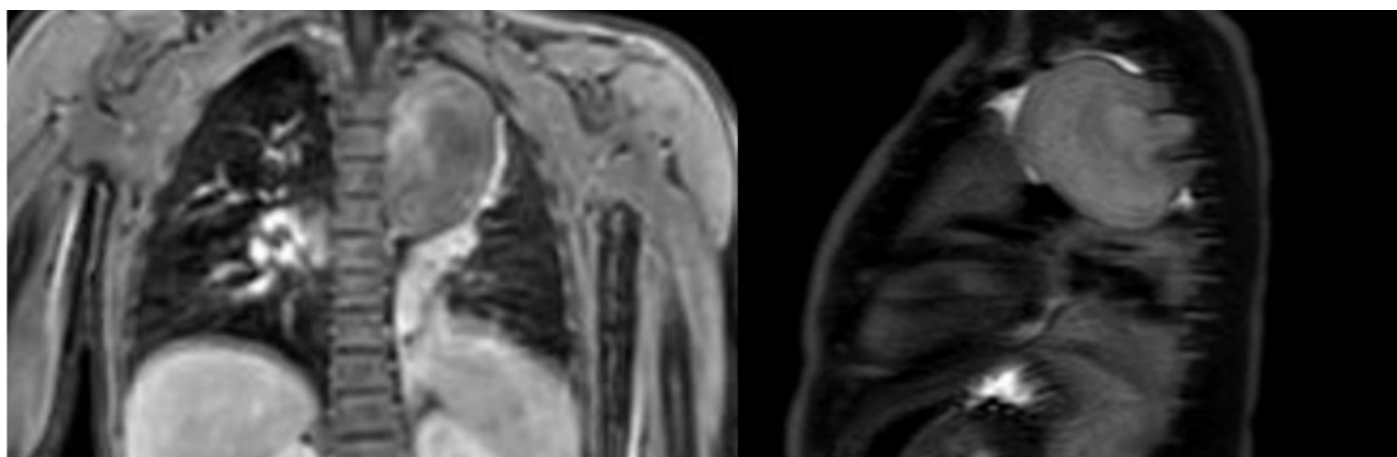


Figure 1. Contrast-enhanced chest MRI in coronal (A) and sagittal (B) reconstructions, demonstrating a mass lesion in the mid-posterior mediastinum (6.2 × 4.6 × 4.2 cm) of heterogeneous texture and discrete heterogeneous contrast enhancement. The lesion is closely related to the large mediastinal vessels, right main bronchus, vertebral bodies, right atrium, diaphragm, and esophagus. There are no direct signs of invasion of these structures.

Total drainage volume was 40ml for the subsequent 24h. The thorax drain was removed, and discharge from the hospital occurred on the third postoperative day.

Case report 2

A 4-year-old patient with no comorbidities, ASA 1, arrived in the emergency department with cough and fever symptoms. Upon examination, blood oxygen saturation was low. An X-ray revealed pneumonia. Antibiotics were started with a resolution of clinical symptoms but no total radiological resolution. Further investigation was then performed with a CT scan-guided biopsy, diagnosing a ganglioneuroma in the right posterior mediastinum. Laboratory exams were routine (Figure 2).

Elective surgery was scheduled, and the patient was admitted to the hospital on the day of the surgery; standard monitors were placed: non-invasive blood pressure, SpO₂, EKG, and EtCO₂. The patient was pre-oxygenated under a face mask, followed by inhalational induction to facilitate peripheral venous puncture. After the peripheral catheter (22G) was inserted, inhalational anesthetics were discontinued, and total intravenous anesthesia (TIVA) started with Propofol infusion using a TCI system with the Schnider targeted for 4ng/dl and remifentanil TCI targeted for 3 ng/dl.

Intubation was performed under direct laryngoscopy, and a cuffed 4.0 orotracheal tube was introduced to the left main bronchus, whose position was confirmed by bronchoscopy. A vesical catheter was also introduced. An ultrasound-guided (USG) internal right jugular vein was cannulated with a double-lumen 5Fr catheter. The choice for the central line placement was made to keep the site on the same side as the thoracotomy.

The patient was then monitored by the neurophysiologist for motor and somatosensory evoked potentials followed

by patient positioning at the right lateral decubitus. Ringer Lactate solution was infused following an initial 10 ml/Kg bolus and subsequently kept at 100 ml/hr.

Tumor resection was performed by thoracoscopy and extraction of the tumor from the cavity was performed by video-assisted minimally invasive thoracotomy. The resection was uneventful and blood loss was kept to a minimum. The surgical team performed ropivacaine 0,5% intercostal nerve blocks at the incision of the intercostal spaces, port sites, and drain sites associated with metamizole and tramadol.

Neuromonitoring helped guide anesthesia depth and provided information regarding the integrity of the nervous system. Before closing the surgical site, the orotracheal tube cuff was deflated and retracted until the non-dependent lung expanded upon ventilation, at which point it was reinflated.

After re-expansion, the non-dependent surgical sites were closed, and the patient awakened with no neurological deficits. Extubation occurred in the operating room after oropharyngeal aspiration, where reflexes had been recovered and breathing resumed.

All vital signs were within normal range and the patient was taken to the ICU as per institutional protocol. Admission into the ICU was uneventful, and the patient was discharged to the ward on the third postoperative day. Drainage was minimal, and the drain was removed on the second postoperative day. The patient was discharged from the hospital on the fifth postoperative day. The patient visited the doctor's office one month postoperatively for a follow-up consultation. There were no complaints and no restrictions, and regular activities were resumed.

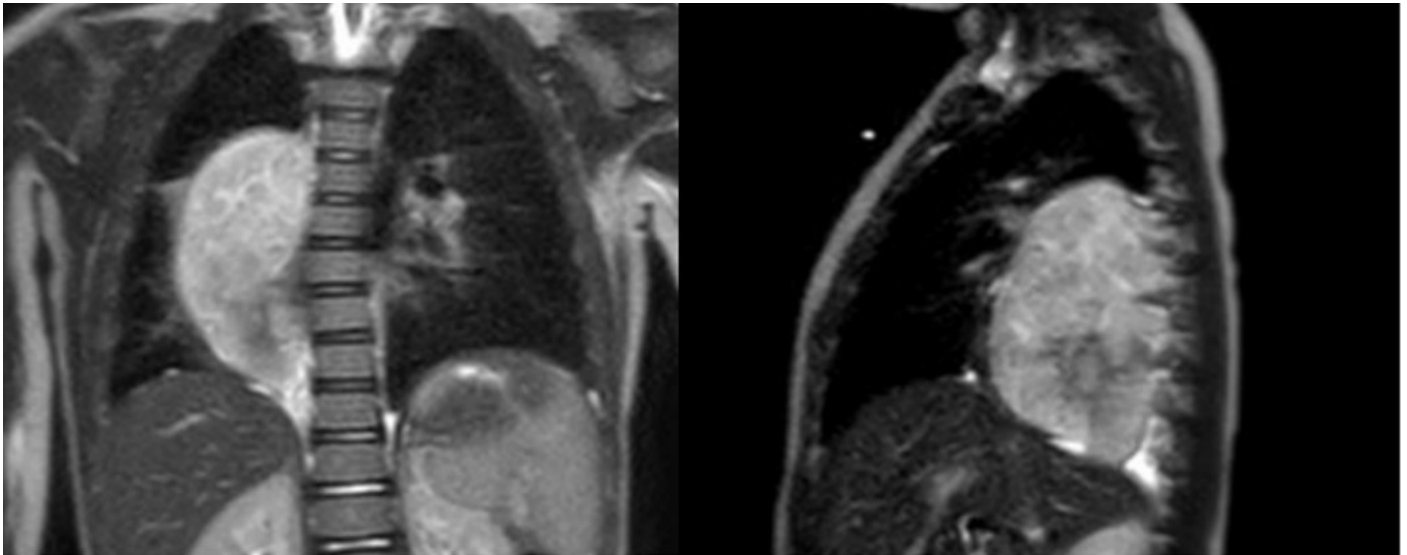


Figure 2. Contrast-enhanced chest MRI in coronal (A) and sagittal (B) reconstructions, demonstrating a mass lesion in the mid-posterior mediastinum (6.0 × 3.9 × 3.8 cm), of heterogeneous texture, with discrete heterogeneous contrast enhancement. The lesion is closely related to the large mediastinal vessels, right main bronchus, vertebral bodies, right atrium, diaphragm, and esophagus. There are no direct signs of invasion of these structures.

DISCUSSION

This article presents two case reports with detailed anesthetic management of children submitted to VAT and OLV, along with a description of similar cases found in a systematic literature review. It provides valuable information from our long-term experience and six other cases from the literature.

Anesthesia technique

We opted for total intravenous anesthesia (TIVA), necessary for motor and somatosensory evoked potential monitoring during surgery, and for hypoxic vasoconstriction reflex preservation. We decided to establish a central line due to a possible significant blood volume loss and the subsequent necessity of vasoactive drug infusion for volume and blood product administration if deemed necessary.

Ventilation strategy

OLV was the choice in both cases to provide the optimal surgical field for the tumor resection. This was achieved by main bronchus intubation using an endotracheal tube one size smaller than the usual choice for the same patient size and age. That was due to the unavailability of bronchial blockers at the hospital for the procedure.

Although there were no adverse events in our cases, we understand it would have been more efficient if we had used bronchial blockers. They offer the advantages of possible suction and CPAP of the non-dependent lung, and they prevent the anesthetic team from having to manipulate the orotracheal tube during the procedure.

In addition, they provide less risk of tracheal tube obstruction and injury to the main bronchus from the tracheal tube.

Among the techniques utilized by other authors for airway management, only Hosking et al.⁽¹⁸⁾ used OLV as the technique of choice. In his case, the patient's age was very similar to ours. This is the only other case report in which neuromonitoring was applied to guide surgical resection.

Analgesia

We provided analgesia by intercostal nerve block (ICNB) using a multimodal analgesia strategy involving methimazole and tramadol. The results were excellent, with no complaints of pain during the postoperative period and prompt ventilation recovery without ventilatory restrictions due to pain for both of our patients. The surgical team's option to adopt a pigtail catheter thoracic drain and Heimlich valve facilitated pain control.

Similar to our cases, only Rastogi et al.⁽²⁰⁾ adopted ICNB as the technique of choice for analgesia. However, the author does not describe the multimodal strategy being applied alongside the blocks. Furthermore, there is no information on the analgesia outcomes for all case reports, making it difficult to infer how successful the different strategies worked out.

Strengths and limitations

A major strength of this study was the systematic literature review, which brought information on similar cases published so far.

The limitations of this study are the small number of cases in this specific population, the heterogeneity of analgesia strategies across different studies, and the absence of data regarding their efficacy. Therefore, it was difficult to evaluate the success of different analgesia strategies compared to each other. It was also hard to draw further conclusions regarding OLV strategies because, besides our own, there was only one other case report in the literature.

CONCLUSION

Perioperative management of patients undergoing mediastinal mass operations is challenging. Preoperative multidisciplinary discussion, well-planned anesthetic management, and predetermined protocols for emergency situations are all vital to ensure patient safety. The anesthesiologist plays an important role in perioperative care by providing neurological monitoring, OLV, TIVA, and a strong multimodal analgesia strategy to guarantee the safety and quality of the patient's assistance care.

In this context, the anesthesiologist must not only provide anesthesia and analgesia care but should also communicate and integrate between the surgical and anesthetic teams. OLV was safely achieved by utilizing an endotracheal tube one size smaller than the usual option for children's age and size, directed to the main bronchus, and checked with a fiberscope.

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This study was carried out at the Universidade, Cidade, Estado [Hospital Unimed Volta Redonda, Departamento de Anestesiologia, Volta Redonda, Rio de Janeiro, Brasil.

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Ethics statement: The authors confirm that the statements of written informed consent from legally authorized representatives/parents/guardians are available. The manuscript is based on a dataset that has been the basis of another manuscript.

Conflict of interest: None.

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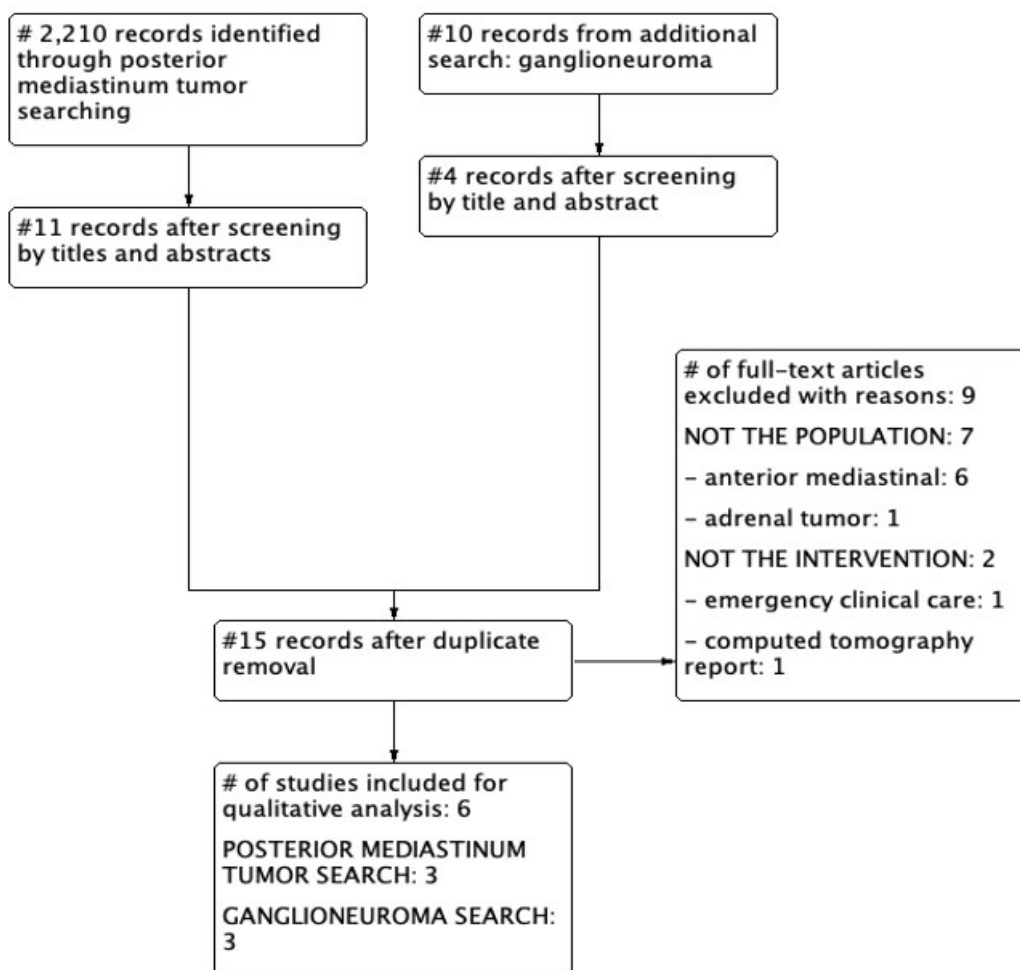
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Correspondence

Carlos Darcy Alves Bersot
UNIFESP, Paulista School of Medicine, Postgraduate in Translational Medicine
Rua Templários, 275, Jardim Anália Franco, São Paulo, SP, Brasil
Phone: +55 (11) 97777-4994
carlosbersot@gmail.com

APPENDIX 1. PRISMA FLOWCHART



APPENDIX 2. SEARCH STRATEGY

SEARCH 1: Filters: humans, under 19 years old, case reports, last 10

Years

SEARCH STRATEGY

MEDIASTINAL TUMOR

Mediastinal Neoplasm OR Neoplasm, Mediastinal OR Mediastinum Neoplasm
OR Neoplasm, Mediastinum OR Cancer of Mediastinum OR Mediastinum
Cancers OR Mediastinal Cancer OR Cancer, Mediastinal OR Mediastinal
Cancers OR Mediastinum Cancer OR Cancer, Mediastinum OR Cancer of the
Mediastinum OR Thoracic Neoplasms OR Neoplasm, Thoracic

AND

Anesthesia OR anesthetics

SEARCH 2: filters: case reports, under 19 years old

Ganglioneuroma search

Ganglioneuroma OR Ganglioneuromas OR Gangliocytoma OR
Gangliocytomas

AND

Anesthesia