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CASE REPORT

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# Anesthesia for bronchoscopy interventional therapy in a patient with Gorham-Stout disease, lung cancer, and right lung atelectasis: a case report

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## Abstract

**Background** Gorham-Stout disease (GSD) is an extremely rare disease of unknown etiology, characterized by painless and progressive bone resorption that may affect multiple bones throughout the body. GSD primarily involves the maxillofacial region, leading to facial disfigurement and reduced joint stability, thereby increasing the risk of challenging tracheal intubation. Limited cases have been reported on the co-occurrence of GSD in the maxillofacial region with lung cancer and right lung atelectasis, particularly regarding anesthesia management for bronchoscopy interventional therapy in such patients.

**Case presentation** This report presents a successful case of a patient with maxillary GSD and right lung atelectasis secondary to lung cancer who underwent bronchoscopy interventional therapy under general anesthesia. The perioperative course was uneventful, with no complications observed.

**Conclusion** Anesthesia management is critical in the surgical treatment of patients with GSD. Airway management poses unique challenges, necessitating thorough preoperative evaluation and implementation of strategies to address potential intubation difficulties. Additionally, vigilance for intraoperative complications is essential.

**Keywords** Gorham-Stout disease, Lung cancer, Right lung atelectasis, Bronchoscopy interventional therapy, Anesthesia management

## Background

Gorham-Stout disease (GSD) is a rare disorder of unknown etiology, characterized by painless, progressive resorption of bone [1]. It can affect multiple bones throughout the body, primarily involving the mandible, scapula, pelvis, femur, and skull. While maxillofacial GSD most commonly involves the mandible, it may also extend to other maxillofacial bones [2]. Meanwhile, lung cancer ranks second in global cancer incidence rates and first in cancer mortality [3]. Nearly 80% of lung cancer patients are diagnosed at an advanced stage, where treatment options are relatively limited [4].

Bronchoscopy has emerged as an indispensable therapeutic modality in the management of lung cancer [5]. This approach can alleviate symptoms such as dyspnea and hemoptysis through techniques including tumor resection, laser therapy, and stent implantation, thereby slowing disease progression and improving the patient's

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quality of life [6]. However, bronchoscopy airway interventions typically require general anesthesia and may be associated with complications such as hypoxemia and cardiac arrhythmia [7]. Consequently, meticulous anesthesia management and vigilant monitoring are essential to ensure safe and efficient execution of the procedure.

We report a successful airway management and anesthesia strategy during bronchoscopy intervention in a male patient in his 50s with maxillary GSD complicated by lung cancer and right lung atelectasis.

## Case presentation

A 59 -year-old, 68 kg male patient was admitted to the Department of Respiratory and Critical Care Medicine with a one-month history of worsening cough and chest tightness and a diagnosis of right lung squamous cell carcinoma. Ten years prior, the patient developed left maxillary swelling, facial asymmetry, malocclusion, and missing teeth in the left upper and lower jaws. Pathological examination revealed proliferating thick-walled capillaries beneath the maxillary epithelium, dilated capillaries between bone trabeculae, and bone trabecular resorption, consistent with massive osteolysis, confirming GSD (Fig. 1). The patient received no regular treatment. Two years earlier, a chest computed tomography (CT) scan for recurrent cough and chest tightness identified a right lower lobe mass and obstructive pneumonia. Pathological analysis confirmed poorly differentiated squamous cell carcinoma, and the patient underwent regular radiotherapy and chemotherapy with stable pulmonary lesions on follow-up.

Preoperative evaluations included: (1) Imaging: Enhanced chest CT demonstrated right pneumothorax with hydropneumothorax, a right main bronchus mass, complete right lung atelectasis, mediastinal shift to the right, and right pleural effusion (Fig. 2). (2) Laboratory tests: Blood biochemistry showed hypoalbuminemia (34.2 g/L), hyperglycemia (8.0 mmol/L), and elevated C-reactive protein (140.5 mg/L). Arterial blood gas analysis revealed hypoxemia [$PaO_2$ (partial pressure of oxygen in arterial blood)of 58 mmHg]. (3) Physical examination: Karnofsky Performance Status score was 60, and Borg dyspnea score was 3. Breath sounds were diminished in the right lung with dullness to percussion; the left lung exhibited coarse breath sounds. Other ancillary examinations showed no significant abnormalities.

Surgical planning: Multidisciplinary consensus determined the need for rigid bronchoscopic tumor resection to relieve right lung atelectasis and dyspnea. Given the procedure's duration and stimulation, general anesthesia was required. Bronchial artery embolization was performed preoperatively to minimize intraoperative bleeding risk.

Preoperative anesthesia assessment: The patient presented with severe maxillary GSD-related craniofacial deformity, accompanied by restricted cervical spine mobility, severely limited mouth opening (approximately one fingerbreadth), and a thyromental distance of two fingerbreadths. Combined with a Mallampati class IV airway, these anatomical features strongly indicated anticipated challenges in both tracheal intubation and mask ventilation. On admission, the patient was placed in a semi-recumbent position with nasal cannula oxygen (5 L/min), maintaining pulse oxygen saturation($SpO_2$) approximately 90%. According to the latest American Society of Anesthesiologists guidelines for managing difficult airways [8], emergency airway equipment was prepared, including various airways, laryngeal mask airways,

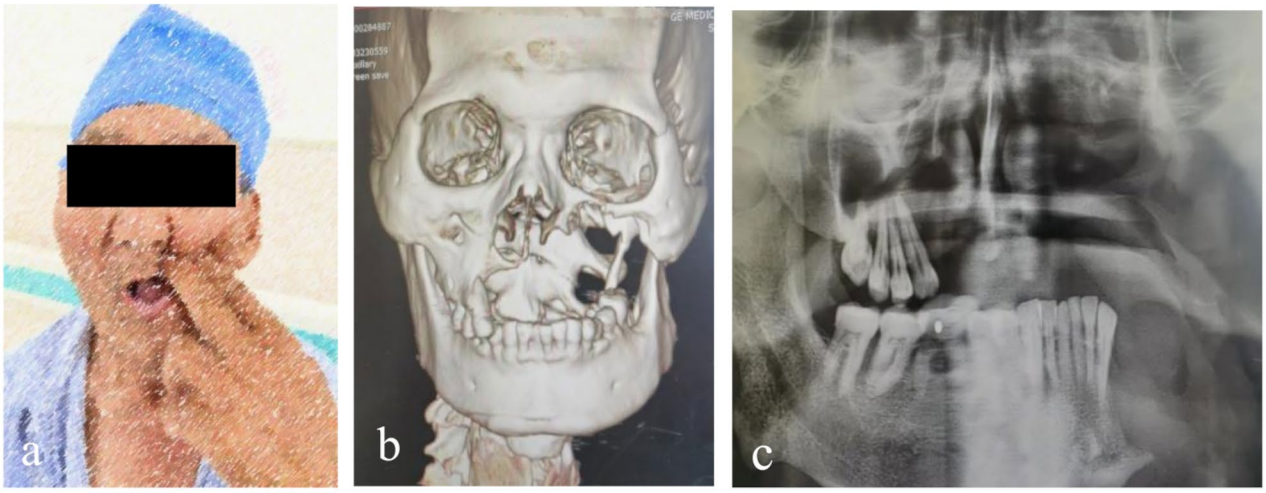


**Fig. 1** Clinical and Radiological Features of Facial Deformities in GSD. **a**: Facial deformity in GSD, with facial asymmetry, occlusal disorder, and loss of upper and lower jaw and teeth on the left side. **b**: Three-dimensional reconstructed CT of the face: collapse of the left side of the face. **c**: X-ray of the maxillofacial region: most of the maxilla is missing, and part of the left mandible is missing

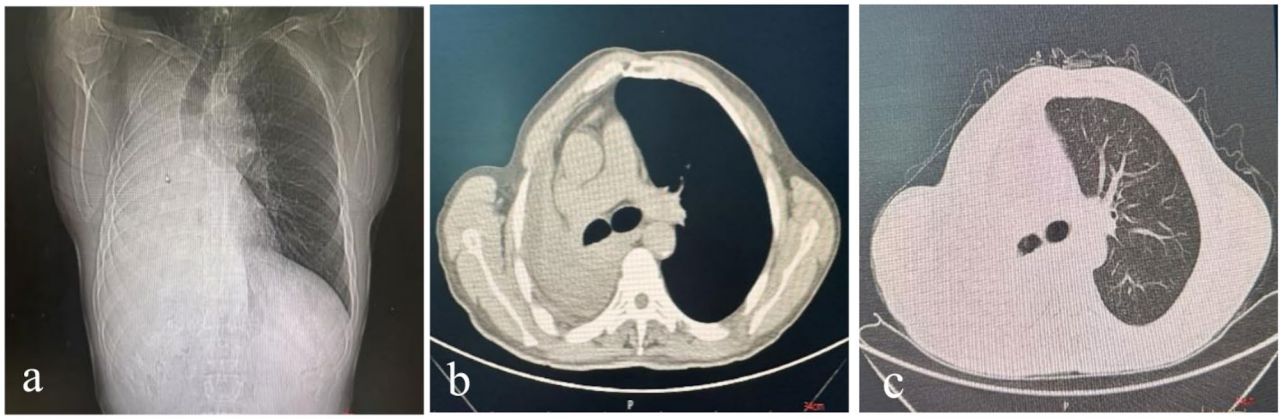


**Fig. 2** Preoperative Enhanced Chest CT Findings. **a**: Plain film: Occupation of the right main bronchus and right lung, leading to total atelectasis of the right lung. **b**: Mediastinal window: Total atelectasis of the right lung, rightward shift of the mediastinum, and right pleural effusion. **c**: Lung window: Occupation of the right main bronchus and right lung, thickening of the right main bronchial wall, lumen obstruction, atelectasis of the right lung, and increased and disordered markings in the left lung

endotracheal tubes, video laryngoscopy, bronchoscopes, surgical airway kits (cricothyrotomy, tracheostomy), and advanced cardiopulmonary support devices like extracorporeal membrane oxygenation. No significant abnormalities were detected in other organ systems during preoperative evaluation.

Anesthesia management: Upon arrival in the bronchoscopy suite, the patient was positioned semi-sitting. Oxygen was administered via a face mask [fraction of inspired oxygen ($FiO_2$) = 100%, flow rate 5 L/min], and continuous monitoring revealed the following vital signs: heart rate (HR) 102 bpm, non-invasive blood pressure (NIBP) 100/68 mmHg, respiratory rate (RR) 26 bpm, $SpO_2$ 89%. Pre-induction management included topical anesthesia with 2% lidocaine applied to the oropharynx and cricothyroid membrane puncture for airway topical anesthesia. Concurrently, dexmedetomidine 40 μg was administered intravenously via slow infusion. After achieving mild sedation with stable spontaneous breathing, bronchoscopy confirmed intact nasopharyngeal and main airway structures. Anesthesia induction was initiated with ciprofol 0.4 mg/kg and remifentanil 0.5 μg/kg. Following loss of consciousness and confirmation of adequate mask ventilation, rocuronium 0.6 mg/kg was administered as a muscle relaxant. A rigid bronchoscope was then inserted and connected to a high-frequency jet ventilator (Twinstream®, Austria) with initial ventilator settings were normal frequency jet ventilation with a driving pressure (DP) of 0.9 bar, RR of 18 bpm, and $FiO_2$ of 80%.

At 1 min, $SpO_2$ dropped below 85%, prompting an increase in DP to 1.1 bar and RR to 20 bpm. After another minute, $SpO_2$ remained below 90%. The ventilation mode was switched to normal frequency combined with high-frequency jet ventilation (DP of 0.6 bar, RR of 300 bpm, $FiO_2$ of 100%), which stabilized $SpO_2$ above 90%. Anesthesia was maintained with remifentanil at 0.2 $ug{\cdot}kg^{-1}{\cdot}min^{-1}$ and propofol at 3 $mg{\cdot}kg^{-1}{\cdot}h^{-1}$.

Continuous NIBP, HR, $SpO_2$, and bispectral index were recorded. The 80-minute procedure involved 500 mL crystalloid infusion and 50 mL blood loss. Postoperative blood gas analysis showed $PaO_2$ of 150 mmHg and partial pressure of carbon dioxide in the artery ($PaCO_2$) of 55 mmHg. Reversal with sugammadex 130 mg and doxapram hydrochloride 20 mg restored spontaneous breathing. Upon achieving an Aldrete score ≥ 9, the patient was transferred to the ward.

The operation proceeded smoothly. The tumor obstructing the right main bronchus (Fig. 3a) was resected using electrocautery snare, $CO_2$ cryotherapy, argon plasma coagulation, and high-frequency electrocautery (Fig. 3b). During resection, the patient experienced a sudden onset of supraventricular tachycardia (SVT), characterized by a regular RR interval and normal QRS morphology on the electrocardiogram, with a peak HR of 200 bpm. The NIBP remained stable at 100/55 mmHg. The endoscopist was immediately instructed to cease the procedure, and 20 mg of esmolol was administered to control the HR, reducing it to 130 bpm. The HR was further stabilized to around 110 bpm, allowing the surgery to continue. Post-procedure, the right middle bronchial lumen was notably wider, with the openings to the right upper and middle lobes visible. However, the right lower lobe opening remained completely obstructed by the tumor. The left main bronchus and its branches were clear and unobstructed (Fig. 3c).

The patient recovered without complications, with improved breathing and cough. Follow-up CT showed partial re-expansion of the right lung (Fig. 4). The patient was discharged on the fifth postoperative day.

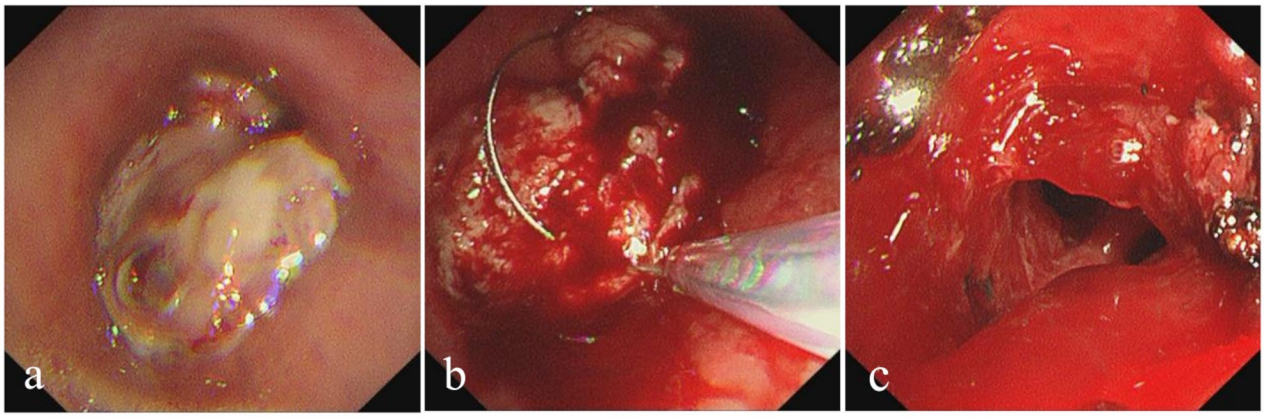


**Fig. 3** Intraoperative Bronchoscopy Findings. **a**: Preoperative: New growth in the lumen of the right main bronchus, completely obstructing the lumen. **b**: Intraoperative: Tumor removal using electrocautery snare, $CO_2$ cryotherapy, argon knife, and high-frequency electric cautery. **c**: Postoperative: Significant widening of the middle section of the right bronchial lumen, with exposure of the openings of the right upper lobe and right middle lobe; the opening of the right lower lobe remains completely obstructed by the tumor

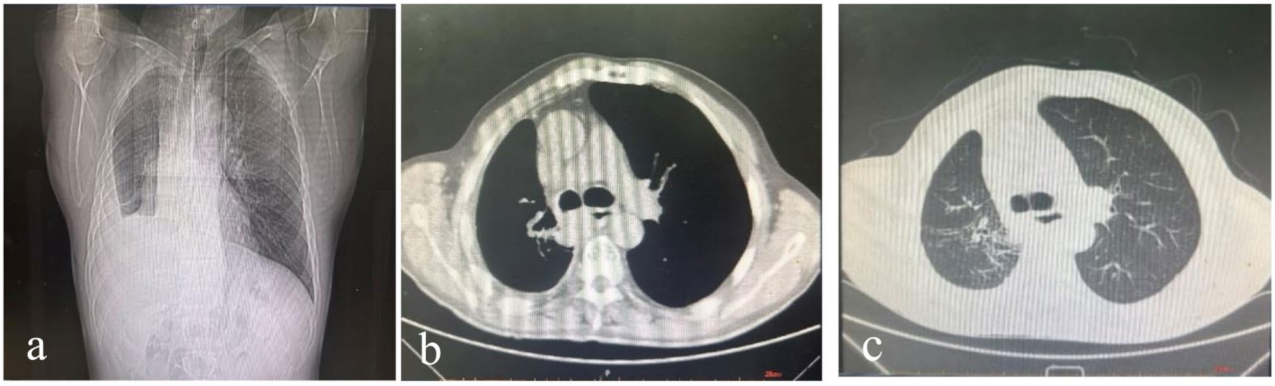


**Fig. 4** Postoperative Enhanced Chest CT Findings. **a**: Plain film: Re-expansion of the right upper and middle lobes, with persistent obstruction of the right lower lobe bronchus, and atelectasis of the right lower lobe. **b**: Mediastinal window: Re-expansion of the right upper and middle lobes, rightward shift of the mediastinum, and right pleural effusion. **c**: Lung window: Re-expansion of the right upper and middle lobes, with a mass lesion in the right hilum and subhilar region, primarily of soft tissue density, containing air bronchograms, and increased and disordered markings in the left lung

## Discussion and conclusions

GSD, also known as vanishing bone disease, is an extremely rare, idiopathic bone disorder, with approximately 300 reported cases to date, about 60 of which involve the craniofacial region [9]. GSD most commonly affects bones such as the mandible (15%), ribs (12%), femur (11%), pelvis (10%), scapula (10%), and humerus (8%). Clinical manifestations vary depending on the affected bone. In the craniofacial region, symptoms may include pain, tooth mobility, fractures, and facial deformities [10]. Spinal involvement can lead to neurological symptoms, paraplegia, and even death, while thoracic involvement can cause chylothorax, leading to respiratory failure and subsequent death [11, 12].

Airway management in GSD patients is particularly challenging. A retrospective study by Professor R. Scott Herd found that 64% of patients with primary osteolysis syndrome had difficult intubation, and 27% required prolonged mechanical ventilation postoperatively [13]. The success of airway interventional therapy hinges not only on meticulous anesthesia management but equally on synergistic collaboration between endoscopists and anesthesiologists. The inherent challenge of shared airway management during bronchoscopy procedures introduces multifactorial risks, encompassing mechanical trauma, bronchospasm, laryngospasm, hypoxemia, hypercapnia, and hemodynamic perturbations [14–16]. Additionally, the presence of atelectasis reduces ventilated lung tissue volume, which in turn decreases effective alveolar ventilation and increases the risk of ventilation-perfusion mismatch, further predisposing the patient to hypoxemia and hypercapnia.

The anesthesia management of such patients poses significant challenges, requiring special attention to the following points:

1. Preoperative Evaluation: This patient, due to GSD, presents with severe facial deformity, limited neck extension, and significant restriction in mouth opening, all of which indicate a difficult airway. Additionally, tumor compression has led to one-lung atelectasis and subsequent respiratory failure. The combination of one-lung ventilation and a difficult airway further complicated intraoperative management. Therefore, a comprehensive assessment should be performed, taking into account the location and extent of the lesions associated with the patient's GSD, as well as any comorbidities and underlying conditions.
2. Anesthesia Method: Under general anesthesia, tumor resection using a rigid bronchoscope was the preferred method to alleviate the patient's breathing difficulties. However, given the patient's condition, there was a high risk of difficult intubation and mask ventilation. To prevent potential ventilation difficulties post-anesthesia, it was essential to prepare various tools for difficult airway management. A combination of adequate topical anesthesia and a method that preserves spontaneous breathing should be used [17]. Only after confirming the absence of mask ventilation difficulties should general anesthetics be administered, followed by the placement of the rigid bronchoscope.
3. Intraoperative Management: Given the patient's poor lung function and complete atelectasis of one lung, ensuring adequate oxygenation was critical. To avoid hypercapnia, normal frequency jet ventilation should be the first choice. If maintaining oxygenation becomes challenging, switching to combined normal frequency and high-frequency jet ventilation may be necessary [18]. Also, studies have proven that the use of high-frequency jet ventilation during bronchoscopy can effectively improve intraoperative hypoxemia [19]. A Spanish study analyzing 208 fiberoptic bronchoscopy procedures in an intensive care unit found that SVT was one of the most common complications, with an incidence of about 3.8% [20]. SVT can result from various factors, including hypoxemia, hypercapnia, acidosis, hypotension, electrolyte imbalance, surgical stimulation, hypothermia, shock, adrenergic stimulation (e.g., insufficient anesthesia), use of arrhythmogenic drugs, myocardial ischemia, and emotional stress [21, 22]. The patient developed SVT during the operation. It was considered that this may be due to the strong stimulation from the bronchoscopy treatment, or the sympathetic nerve excitation or elevated circulating catecholamines induced by the use of electrocautery and other procedures by the endoscopist, leading to SVT. Additionally, the patient's rightward mediastinal shift and the change in ventilation/perfusion ratio during lung re-expansion could have triggered arrhythmia. Postoperative blood gas analysis showed a slight increase in $PaCO_2$. Research indicates that mild hypercapnia does not cause cognitive dysfunction or postoperative delirium [23, 24]. The patient reported significant improvement in dyspnea upon awakening, with no signs of re-expansion pulmonary edema or significant hypercapnia.

GSD is a rare disease that can affect multiple organs and tissues, with a reported mortality rate of up to 16% [25]. While there is no evidence of liver, kidney, muscle dysfunction, or malignant hyperthermia in GSD patients [26], careful preoperative assessment is essential. Anesthetic management should focus on potential respiratory complications, metabolic imbalances, and skeletal abnormalities that may impact airway management and positioning. Sahoo et al. [27] reported a 21-year-old male with clavicular GSD undergoing spinal decompression and fusion, emphasizing the importance of airway management and intraoperative positioning. Yildiz et al. [28] described a pediatric case of GSD with chylothorax, necessitating one-lung ventilation with a bronchial blocker, illustrating the complexity of airway and respiratory management in thoracic involvement. Huang et al. [29] detailed a case of postoperative respiratory failure in a GSD patient requiring prolonged mechanical ventilation, underscoring the need for careful extubation planning and postoperative respiratory support. Collectively, these cases emphasize the necessity of thorough preoperative assessment, individualized anesthetic planning, and vigilant postoperative monitoring to optimize perioperative outcomes in GSD patients.

This case report still has some limitations. First, there is limited literature on the anesthesia management of patients with GSD, especially cases involving concurrent lung cancer and complete atelectasis of one lung. Secondly, further research is warranted to explore the long-term outcomes of GSD patients undergoing anesthesia and to investigate anesthesia management strategies across a variety of surgical procedures in this patient population. As a single case report, the generalizability of anesthetic management is limited. However, we believe that this case provides valuable insights into the specific challenges of anesthetic management in patients, and will build a more comprehensive understanding of this area.

The occurrence of maxillofacial GSD combined with lung cancer and unilateral lung atelectasis is relatively rare. Effective anesthesia management is crucial for airway intervention surgery in these patients, as airway management presents significant challenges. Intraoperative complications, such as hypoxemia and arrhythmias,

must not be overlooked. Therefore, developing a tailored anesthesia management strategy is essential to ensure the smooth execution of the surgical procedure.

**Abbreviations**

| | |
|---|---|
| GSD Gorham | Stout disease |
| CT | Computed tomography |
| PaO< | Subscript>2</Subscript> partial pressure of oxygen in arterial blood |
| FiO< | Subscript>2</Subscript> fraction of inspired oxygen |
| HR | Heart rate |
| NIBP | Non-invasive blood pressure |
| RR | Respiratory rate |
| SpO< | Subscript>2</Subscript> pulse oxygen saturation |
| DP | Driving pressure |
| PaCO< | Subscript>2</Subscript> partial pressure of carbon dioxide in arterial blood |
| SVT | Supraventricular tachycardia |

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**Author contributions**
Yuxue Yao, Hong Li have drafted the work. Mingyuan Yang, Qinghao Cheng substantially revised the draft. Mingyuan Yang, Qinghao Cheng, Yuxue Yao have approved the submitted version and have agreed both to be personally accountable for the author's own contributions and to ensure that questions related to the accuracy or integrity of any part of the work.

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**Data availability**
No datasets were generated or analysed during the current study.

## Declarations

**Ethics approval and consent to participate**
Not applicable.

**Consent for publication**
Written informed consent for publication of this paper was obtained from the patient's family.

**Competing interests**
The authors declare no competing interests.

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