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Case Report

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Anesthesia management in double balloon enteroscopy for a patient with hereditary hemorrhagic telangiectasia (Osler-Weber-Rendu)

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ABSTRACT

This case report discusses the anesthesia management of a 70-year-old male with hereditary hemorrhagic telangiectasia, also known as Osler-Weber-Rendu disease, who underwent double balloon enteroscopy for the treatment of gastrointestinal angiodysplasia. The patient, with a history of anemia requiring frequent blood transfusions, was managed with deep sedation using a combination of midazolam, dexmedetomidine, and propofol during the procedure. The sedation approach was tailored to the patient's comorbidities and preference against general anesthesia. The procedure was completed successfully without complications, and the patient was discharged the following day. The use of dexmedetomidine and propofol ensured stable hemodynamy and airway security, making it an effective sedation strategy for complex gastrointestinal procedures in elderly patients. The report highlights the importance of appropriate sedation protocols, particularly for patients with multiple comorbidities, and emphasizes the need for further research on sedation depth monitoring to optimize anesthetic care.

Keywords: Anesthesia, double balloon enteroscopy, hereditary hemorrhagic telangiectasia, Osler-Weber-Rendu

INTRODUCTION

Hereditary hemorrhagic telangiectasia (HHT), also known as Osler-Weber-Rendu disease, is a rare, autosomal dominant, multisystemic disorder characterized by mucocutaneous telangiectasias and arteriovenous malformations in various internal organs. With an average global prevalence of 1 in 5.000 to 8.000, HHT[1] primarily affects the nasal mucosa, gastrointestinal (GI) tract, lungs, liver, and central nervous system, causing arteriovenous shunts.

HHT can lead to severe anemia and high-output heart failure due to recurrent massive nosebleeds and GI bleeding. Selective cauterization of GI telangiectasias can help control anemia but does not prevent recurrent bleeding.[2] Anesthesia management under sedation is crucial for protecting the patient's physical and psychological well-being, preventing adverse events, and enhancing the effectiveness of the procedure. The appropriate technique and drug selection should be made to partially reduce the patient's level of consciousness, ensure immobility, and manage pain and stress, while closely monitoring the depth of sedation. Effective anesthesia management not only increases the success of the surgical intervention but also ensures the patient's perioperative well-being. In this case presentation, we aim to share the anesthesia management under deep sedation of a patient who underwent cauterization of GI angiodysplasia foci due to recurrent transfusion needs caused by anemia.

CASE

A 70-year-old male patient, weighing 83 kg with a BMI of 32 kg/m^2 presented to the anesthesia clinic for double balloon enteroscopy (DBE). He had a history of melena and had been receiving periodic blood transfusions, with the need for transfusion increasing over the past few months. The patient had been diagnosed with Osler-Weber-Rendu syndrome, and his jejunal angiodysplasia lesions had been previously treated with argon plasma coagulation (APC) during two prior DBE procedures.

The patient's diagnosis was confirmed during a family screening following the hemorrhagic brain hemorrhage of his 22-year-old son, who had widespread arteriovenous malformations in the brain. Until four years ago, the patient had no symptoms related to the syndrome. There was no known angiodysplasia in areas other than the GI tract. Additionally, he had suffered a myocardial infarction 9 years ago and had undergone coronary artery bypass surgery. The patient had a 50 pack/year smoking history, with chronic obstructive pulmonary disease for the last 20 years as an ex-smoker. He had experienced ischemic cerebrovascular events after the bypass surgery and had been started on 75 mg clopidogrel.

Preoperative evaluation revealed no specific findings on body surface. His blood pressure was 116/61 mmHg, and his heart rate was 71 bpm. Peripheral oxygen saturation (SpO_2) in room air was 93%. Lab results showed a hemoglobin of 9.2 g/dl, hematocrit 29.6, platelet count 153.000/µL, INR 1.06, serum creatinine (sCrea) 0.87 mg/dl, ALT 45 U/L, AST 55 U/L, with no notable abnormalities other than anemia. The ECG showed negative T waves between leads T3 and T5. Transthoracic echocardiogram revealed left ventricular hypertrophy and a 55% ejection fraction. The estimated procedure duration was 2 hours.

The case was a procedure as an elective outpatient surgery. The patient was evaluated as ASA III and informed about the procedure and anesthesia, and consent was obtained. Due to the patient's comorbidities and preference against general anesthesia, deep sedation was planned for anesthesia management.

Upon entering the operating room, the patient was positioned supine, and monitoring included a 5-lead ECG, pulse oximeter, non-invasive blood pressure cuff, capnography, and a temperature probe. SpO_2, heart rate, and ECG were continuously monitored, and noninvasive blood pressure was measured every 5 minutes. A 21-gauge catheter was placed for peripheral venous access. The patient received oxygen via a procedural oxygen mask (POM) at 4 L/min throughout the procedure. The patient was sedated with 0.07 mg/kg of IV midazolam, 1 mcg/kg of dexmedetomidine in 10 minutes, and a propofol infusion at 75 mcg/kg/min. Only the loading dose of dexmedetomidine was administered, and the infusion dose was not given as it was necessary. On the 15th minute, 10 mg of Hyoscine-N-butyl bromide IV was administered as requested by the gastroenterologist. The procedure advanced approximately 450 cm into the duodenum, jejunum, and ileum, where 84 APC treatments were applied to the angiodysplasia lesions, more densely located in the jejunum (Figure). At the end of the procedure, the patient's blood pressure was 117/60 mmHg, and his heart rate was 77 bpm. Oxygen was administered via mask at 2 L/min, and SpO_2 was 96%. He was transferred to the recovery unit.

During the procedure was applied deep sedation. The patient was spontaneously breathing but did not any move. Blood pressure remained within normal limits. SpO2 ranged from 90- 93%, and end-tidal CO_2 was between 35-45 mmHg. Respiratory rate ranged from 12-18 breaths per minute. The procedure was completed within 75 minutes, and a total of

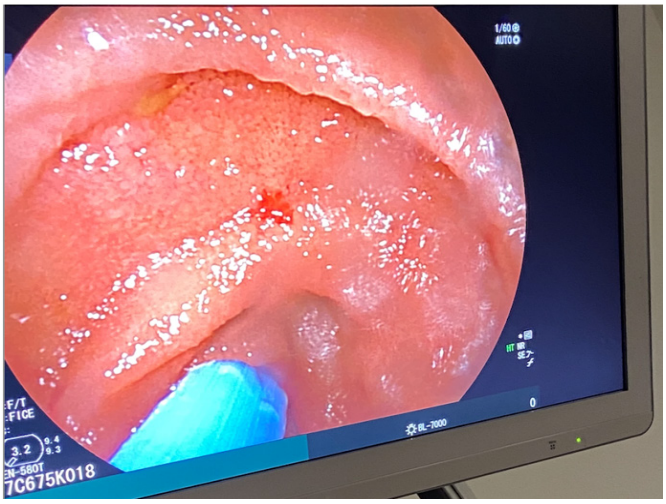

Figure. The angiodysplasia lesion

750 ml of Ringer's lactate was administered. The patient was transferred to the gastroenterology department and was discharged on the first postoperative day without issues.

DISCUSSION

This case describes our anesthesia management experience for a 70-year-old male patient with HHT who underwent double-balloon enteroscopy to treat GI telangiectasias that caused anemia and required frequent blood transfusions.

In HHT, lesions should be treated in a multidisciplinary manner. Treatments for lesions in the skin, mucosa, GI tract, and liver are symptom-driven, while vascular malformations in the lungs and brain should be treated even without symptoms due to the potential life-threatening complications. Common clinical findings include GI and nasal bleeding, which result in anemia and the need for transfusions. Pulmonary vascular malformations can cause dyspnea, heart failure, stroke, and brain abscesses. Cerebral vascular malformations can lead to hemorrhagic stroke and seizures.[3] Our patient's son was diagnosed after experiencing a hemorrhagic brain hemorrhage.

GI bleeding is common in HHT patients. A study by Kjeldsen et al.[4] showed that 25% of HHT patients over 60 years old had significant GI bleeding.

Similarly, our patient, who had no symptoms until his later years, presented with anemia and frequent blood transfusions in the recent years. Mora-Lujan et al.[5] reported that patients with GI involvement were more likely to require emergency room visits and hospital admissions, with telangiectasias larger than 3 mm requiring more frequent transfusions. Our patient also required transfusions every 15 days. Although upper GI endoscopy was normal, there were widespread telangiectasias in the duodenum, jejunum, and ileum. The size and number of telangiectasias made it difficult to assess their exact characteristics.

The APC procedure for 84 angiodysplasia lesions was completed without any complications under deep sedation, with an anesthesia duration of 75 minutes. A study by May et al.[6] demonstrated that DBE with APC is effective in treating

bleeding foci in the small intestine, and long-term follow-up data showed a significant increase in hemoglobin levels and a reduction in the need for transfusions. Many publications support APC as an effective treatment for angiodysplasia lesions.[7,8]

During invasive procedures, the sedation level and airway management are tailored based on patient safety, comorbidities, the type and duration of the procedure, and the complexity. The European Society of GI Endoscopy recommends that DBE can be performed under conscious sedation, deep sedation, or general anesthesia, with the choice based on the complexity of the procedure, clinical factors, and local protocols.[9]

Given the numerous lesions in our patient, deep sedation was necessary for this challenging procedure. Goudra et al.[10] found that most elective ERCP procedures could be performed without endotracheal intubation (intubation rate <1%), and intubation was reserved for cases with aspiration risks. A study by Thamson et al.[11] indicated that children might have lower sedation success or higher rates of general anesthesia. Our adult patient, who had previously undergone similar procedures twice, adapted well to the sedation process, aided by preprocedural counseling and administration of 0.07 mg/kg IV midazolam to reduce anxiety.

Multimodal anesthesia agents should be used based on the type and duration of the procedure. A variety of medications, including benzodiazepines (midazolam), opioids (fentanyl and meperidine), propofol, and dexmedetomidine (DEX), are available for conscious sedation during endoscopic procedures.[12]

In our case, a combination of dexmedetomidine (80 mcg loading dose) and propofol infusion (75 mcg/kg/min) provided adequate sedation depth. Dexmedetomidine allowed stable hemodynamics and deeper sedation without excessive propofol, which can lead to respiratory depression. We did not use an infusion of dexmedetomidine beyond the loading dose to avoid its side effects in elderly patients. In line with findings by Chen et al.[13], the combination provided stable sedation depth, airway security, and hemodynamic stability with reduced propofol consumption.

Concerns regarding delayed awakening in elderly patients treated with dexmedetomidine were mitigated in this case, as the patient regained adequate consciousness and spontaneous breathing within 5 minutes after stopping the propofol infusion, allowing for smooth transfer to the recovery unit.

Propofol combined with an opioid is a common sedation protocol; however, high doses or prolonged use of propofol may lead to respiratory depression, hypoxemia, and hypotension.[14] In the study conducted by Tuzlalı and colleagues[15], it was demonstrated that propofol infusion provides rapid recovery and is effective in preserving postoperative cognitive functions. In this case, we aimed to benefit from these effects of propofol. Dexmedetomidine alone, while effective for sedation, may cause hypotension and bradycardia and is not as effective as propofol for sedation.[16] In this case, we avoided continuous infusion of dexmedetomidine to prevent hypotension.

Dexmedetomidine-based conscious sedation is a safe option for long-duration GI endoscopy, with elderly patients requiring careful monitoring. Despite potential side effects like bradycardia and hypotension, dexmedetomidine has advantages in maintaining respiratory strength and airway patency during sedation.[17] Dexmedetomidine, recognized for its neuroprotective properties, may help reduce the incidence and severity of postoperative cognitive disorders.[18] However, more clinical studies are needed to assess its risks and benefits.

In future studies, it would be helpful to measure the target plasma concentration of propofol and sedation depth using BIS monitoring for more accurate results.

CONCLUSION

In this case, deep sedation during APC treatment for multiple telangiectasia foci was successfully managed according to the standard protocols recommended in the literature. However, measuring propofol plasma concentrations and sedation depth would lead to more accurate results and better optimization of sedation strategies.

ETHICAL DECLARATIONS

Informed Consent

The patient signed and free and informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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