Cholecystectomy under thoracic epidural anaesthesia for patient with severe intra-pulmonary shunt

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Abstract
Pulmonary arteriovenous malformations are common in patients with hereditary haemorrhagic telangiectasia and can be associated with significant hypoxia and intra-pulmonary shunt. We present a case of a young man with a known 57% calculated shunt requiring abdominal surgery and the multidisciplinary decisions required in the preoperative period to minimize post-operative complications.

Introduction
Hereditary haemorrhagic telangiectasia (HHT) is a rare autosomal dominant disorder, associated with a high risk of morbidity and mortality due to the development of pulmonary and brain arteriovenous malformations [1]. We share our experience of open cholecystectomy under thoracic epidural anaesthesia in a man with HHT and intra-pulmonary arteriovenous malformations (pAVMs) with a calculated shunt fraction of 57%. Our patient’s brittle cardiopulmonary condition placed limitations on the type of procedures that could be safely performed. We detail measures taken to enable safe surgery in a patient whose complexity may otherwise deter anaesthetists and surgeons from operating.

Case Report
A 34-year-old man presented with recurrent acute cholecystitis in 2018, ultimately requiring cholecystectomy, despite multiple endoscopic retrograde cholangiopancreatography procedures with stent insertions and stone removal. The acute infective episode occurred on a background of HHT, with associated innumerable large pAVMs, gastrointestinal telangiectasia, and recurrent epistaxis. The size and physiology of pAVMs were known at the time of surgical work up; at least eight on the right with the largest measuring 30.8 mm and six on the left, with the largest measuring 35.2 mm (Fig. 1). There were no other parenchymal pathologies on imaging. A shunt study demonstrated a calculated shunt fraction to be 57.1% despite recent coiling, with resting hypoxaemia (alveolar partial pressure of oxygen (PaO2): 40–50 mmHg; Fig. 2). Over the previous eight years, embolization had been performed in five sessions, with a total of 21 coils, resulting in no improvement in oxygen saturations. An echocardiogram demonstrated preserved left ventricular size and function (ejection fraction: 61%), mild right ventricular dilatation with preserved function and normal pulmonary pressures (right ventricular systolic pressure: 29 mmHg). He was frail and underweight at 57 kg (body mass index (BMI): 17.6 kg/m²) and had secondary polycythaemia (haemoglobin: 211 g/L). Pulmonary function tests showed forced expiratory volume in 1 sec (FEV₁) 3.25 L (76% predicted), forced vital capacity (FVC) 4.75 L (86% predicted), and diffusion capacity of the lung for carbon monoxide (DLCO) 11.7 mL/mmHg/min (34% predicted). He had no past history of smoking. A 6-min walk test
demonstrated significant oxygen desaturation from baseline 79% to 61%, with a reduced distance achieved (429 m, predicted distance of 801 m). Preoperative computed tomography with contrast excluded significant spinal arteriovenous malformations (AVMs).

A multidisciplinary discussion ensued, including the patient and his family, senior surgical, intensive care, respiratory physician, anaesthetic, and interventional radiology clinicians, to evaluate the preoperative and post-operative anaesthetic and surgical options for cholecystectomy. With standard monitoring in place, a peripheral intravenous cannula and a radial arterial line for invasive blood pressure monitoring were inserted. A thoracic epidural was inserted at T7–T8, using a 16-G Tuohy needle with loss of resistance to air. Dermatomal testing confirming numbness from T4 to T10 was performed with ice and surgical pinch test confirmed surgical site cover. Fentanyl and lignocaine 2% with 1:200,000 adrenaline were titrated through the epidural, totalling 100 mcg and 12 mL, respectively. Light sedation was achieved with propofol, using the Marsh model, 0.5—1.3 mcg/mL. To further prevent changes in pulmonary or peripheral vascular resistance, we aimed for pulse pressure variation of 10%, normothermia, and avoidance of further hypoxaemia, with 70% air:30% oxygen mixture delivered via high-flow nasal cannulae at 20–30 L/min. A total of 0.5 L of Hartmann’s solution was administered intravenously throughout the operation. An arterial blood gas intraoperatively confirmed the expected low PaO2 of 45.5 mmHg.

Intraoperative oxygen saturations ranged between 80% and 88%, with the patient spontaneously breathing. Open cholecystectomy was performed uneventfully over 2 h. A transverse abdominis plane block using 40 mL of 0.2% ropivacaine concluded the case.

The patient was electively transferred to the intensive care unit (ICU) post-operatively, with a 0.2% ropivacaine infusion administered through the epidural for analgesia. The patient was discharged from ICU the following day and was discharged home from the ward on post-operative day 12; the delay was due to the formation of a wound haematoma during the post-operative course. Analgesic requirements included paracetamol and 24 h of oxycodone patient-controlled analgesia (PCA). The epidural infusion was continued post-operatively at a rate of 6–8 mL/h and removed on post-operative day 4. The PCA was then replaced with regular oxycodone/naloxone (10/5 mg oral twice daily) and as needed oral oxycodone.

| Arterial Blood Gas Measurements |
|--------------------------------|
| Hb  | 196.0 g/L |
| pH  | 7.39     |
| PaCO₂ | 35 mmHg |
| PaO₂  | 46 mmHg |

Cumulative Results

| Date      | PaO₂ | Shunt |
|-----------|------|-------|
| 24/07/2018| 46.4 | 57.1% |
| 12/07/2017| 47.8 | 56.3% |

Calculated Values

|                        |        |
|------------------------|--------|
| P\text{C'O}_2         | 669 mmHg |
| Sc\text{O}_2           | 99.99%  |
| Cc\text{O}_2           | 28.3 g/dL |
| CvO₂                   | 16.7 g/dL |
| CaO₂                   | 21.7 g/dL |
| SaO₂                   | 82.04%  |

Figure 1. Chest X-ray posteroanterior (PA) showing location of multiple pulmonary arteriovenous malformation (pAVM) coiling.

Figure 2. Shunt study results. CaO₂, oxygen content arterial blood; Cc\text{O}_2, oxygen content end capillary blood; CvO₂, oxygen content mixed venous blood (estimated as CaO₂ – 5 g/dL); Hb, serum haemoglobin; PaCO₂, alveolar partial pressure of carbon dioxide; PaO₂, alveolar partial pressure of oxygen; P\text{C'O}_2, estimated from calculated PaO₂ (ideal alveolar air equation); SaO₂, oxygen saturation; Sc\text{O}_2, calculated oxygen saturation (100 Y/(Y + 23,400), where Y = PO₂/Y + 150 x PO₂).
Discussion

pAVMs are uncommon in the general population; however, they are present in ~50% of patients with HHT [1]. The majority of pAVMs are asymptomatic and are only identified when they enlarge to cause intra-pulmonary shunt with associated hypoxaemia, haemoptysis, or paradoxical emboli. Other conditions less commonly associated with intra-pulmonary shunt include hepatic cirrhosis, complication of congenital heart disease, or can be idiopathic.

There is limited literature detailing anaesthetic considerations of patients with significant intra-pulmonary shunt. Anaesthesia is known even in healthy individuals to worsen intra-pulmonary shunting through atelectasis with resulting hypoxic vasoconstriction. In the general population, laparoscopic surgery is well documented to increase intra-abdominal pressure and reduce functional residual capacity, particularly at induction of pneumoperitoneum; this increases the risk of atelectasis and hypoxia [2].

In more prevalent conditions associated with hypoxaemia, such as severe chronic obstructive pulmonary disease, there is evidence that regional anaesthesia is associated with lower morbidity and complications compared with general anaesthesia [2]. The specific benefits of epidural anaesthesia include avoiding the requirement for airway instrumentation, reduced surgical blood loss, reduced incidence of thromboembolic, cardiac and pulmonary complications, and obtunding surgical stress response [2]. Thoracic epidurals have also been shown to reduce the incidence and duration of post-operative ileus associated with open abdominal surgery [3].

Operative outcomes in HHT have been reported in a case series of 74, in which only three major complications were encountered. However, the majority of the operations were related to nasal cauterization and none of the patients had significant hypoxaemia [4]. Other case reports of HHT with significant pAVMs have detailed principles of optimizing preoperatively with percutaneous image-guided embolization to reduce the shunt burden [5]. Fortunately, this had been performed in our case prior to the requirement for urgent surgery.

The aim of intraoperative management in this case was to utilize epidural anaesthesia and open surgery to retain the pre-existing ventilation-perfusion ratio. Post-operatively, epidural analgesia was used to avoid the significant reduction in vital capacity and functional residual capacity associated with open cholecystectomy incision pain. This minimizes the risk of post-operative atelectasis and pulmonary infection, while simultaneously reducing opioid administration and avoiding significant respiratory depression.

Location for epidural was also paramount. Serious adverse events have occurred from neuroaxial anaesthesia without prior imaging to exclude cervical or thoracic spinal AVs [4]. T7–T8 interspace was chosen on the basis that lower total volumes of local anaesthetic would be required than if trying to achieve a similar block from a lower thoracic level. The block was sufficient for adequate surgical conditions, with the surgeon confirming that muscle tone was similar compared with general anaesthesia.

In conclusion, for appropriately chosen patients with HHT and significant intra-pulmonary shunt, where spinal AVs have been excluded, epidural anaesthesia in combination with open surgery can offer an effective and safe alternative to general anaesthesia and laparoscopic surgery for cholecystectomy. This should be done in the setting of multidisciplinary care and with consideration given to intensive care requirements.

Disclosure Statement

Appropriate written informed consent was obtained for publication of this case report and accompanying images.

References

1. Guttmacher AE, Marchuk DA, and White RI. 1995. Hereditary hemorrhagic telangiectasia. N. Engl. J. Med. 333(14): 918–924.
2. Hausman MS, Jewell ES, and Engoren M. 2014. Regional versus general anesthesia in surgical patients with chronic obstructive pulmonary disease: does avoiding general anesthesia reduce the risk of postoperative complications? Anesth. Analg. 120:1404–1412.
3. Nimmo SM. 2004. Benefit and outcome after epidural analgesia. Contin. Educ. Anaesth. Crit. Care Pain. 4(2):44–47. https://doi.org/10.1093/bjaceaccp/mkh014
4. Weingarten TN, Hanson JW, Anusionwu KO, et al. 2013. Management of patients with hereditary hemorrhagic telangiectasia undergoing general anesthesia: a cohort from a single academic center’s experience. J. Anesth. 27(5):1–7.
5. Sharma P, Kochar P, Sharma S, et al. 2017. A case of pulmonary arteriovenous malformation: role of interventional radiology in diagnosis and treatment. Ann. Transl. Med. 5(17): 345. https://doi.org/10.21037/atm.2017.06.23.