



OPEN ACCESS

# Intrapulmonary mature cystic teratoma presenting with haemoptysis treated with right middle lobectomy in a low-resource setting

Vijay Anand Ismavel ,<sup>1</sup> Ann Miriam<sup>2</sup>

<sup>1</sup>Surgery, Makunda Christian Leprosy and General Hospital, Karimganj, Assam, India  
<sup>2</sup>Anaesthesia, Makunda Christian Leprosy and General Hospital, Karimganj, Assam, India

Correspondence to  
Dr Vijay Anand Ismavel;  
ivijayanand@yahoo.in

Accepted 4 July 2022

## SUMMARY

A teenage girl presented to our remote rural hospital with history of massive haemoptysis. A CT scan showed a spherical lesion in the right lung. Exploratory thoracotomy was performed with a provisional diagnosis of pulmonary hydatid cyst. Intraoperative findings were consistent with intrapulmonary teratoma and this was confirmed by a histopathological diagnosis of mature teratoma. Four years later, the patient presented again with recurrence of haemoptysis and a right middle lobectomy was performed. We present a report of successful management of this patient with 20-year follow-up since the first surgery, describe the pathology of this rare condition and the procedures used to treat it. We also discuss the social and economic factors that affect decision making in the management of rare conditions in remote rural areas among poor communities that are usually referred to tertiary hospitals and suggest modifications to protocols to provide effective treatment in spite of resource constraints.

## BACKGROUND

Intrapulmonary teratoma is a rare condition and its occurrence in the right middle lobe has not been documented. We report the management of this condition in a low-resource setting. A teenage girl presented to our secondary-level remote rural hospital with haemoptysis. Initial surgery was performed with the facilities available at that time. Four years later, she had a recurrence of haemoptysis and underwent right middle lobectomy. Twenty years after the first surgery, the patient remains healthy. The case is reported for its rarity, the challenging circumstances in which the patient was treated and its successful outcome with long follow-up. Further, we discuss the social and economic factors that affect health-seeking behaviour, the ethical challenges in managing rare conditions (that are usually referred to tertiary care facilities) in a secondary level hospital with respect to the resources available to the institution and patient and suggest modifications to traditional decision-making protocols.

## CASE PRESENTATION

### First presentation

A teenage girl presented to our remote rural hospital located in Assam in northeastern India. She was studying in school; her father was a marginal farmer and mother a housewife. She had experienced small amounts of haemoptysis at home

over several months followed by a large bout, 'half a bucket' of fresh blood along with blood spilled on her clothes and the ground. She was taken to a district hospital, transfused 4 units of whole blood and referred to our hospital for further management. She was stable at initial presentation but was still coughing up small amounts of blood-stained sputum. She was afebrile.

Chest X-ray showed a rounded lesion in the lower half of the right lung. The hospital had a Picker 15 mA X-ray machine at that time which did not produce good quality images and the patient was therefore referred for a CT scan to an imaging centre 100 km away. The report of the CT scan was that there was a cystic lesion of the lung, probably a hydatid cyst (figure 1). Her blood group was O positive, other routine investigations were within normal limits.

At that time, the nearest large referral hospital was over 12 hours by road and a hospital with reliable lung surgeons was 1400 km away (2 days by train). The patient was not able or willing to travel and there was a significant risk of a life-threatening rebleed. Although lung surgery had not been done at the hospital at that time, the surgeon had experience with the pneumonostomy procedure for hydatid cysts of the lung as well as lung resections and it was proposed that the patient undergo pneumonostomy.

The hospital is situated in a remote rural part of northeast India. It started as a leprosy hospital, closed for 10 years and was reopened by the authors 7 years earlier. The authors had undergone postgraduate training in paediatric surgery and anaesthesia and returned a few months prior to the initial presentation of the patient. At that time, they were the only consultants at the hospital, the nearest telephone was 50 km away (2 hours by road), there was no internet access in the area, electricity was provided by a 15 kVA diesel generator and oxygen procured in cylinders from a major city 400 km away (12 hours by road). The hospital had two junior doctors and an outpatient volume of about 100–125 patients but inpatient volumes were low. The operating room used an ordinary 150 W incandescent bulb for lighting and an antique Bovie Spark-Gap machine for electro-cautery but the surgical instruments required for the proposed surgery, especially an adult chest retractor were available. The hospital did not have an intensive care unit or ventilators at that time but one of the authors was well trained in critical care and several



© BMJ Publishing Group Limited 2022. Re-use permitted under CC BY-NC. No commercial re-use. See rights and permissions. Published by BMJ.

**To cite:** Ismavel VA, Miriam A. *BMJ Case Rep* 2022;**15**:e248221. doi:10.1136/bcr-2021-248221



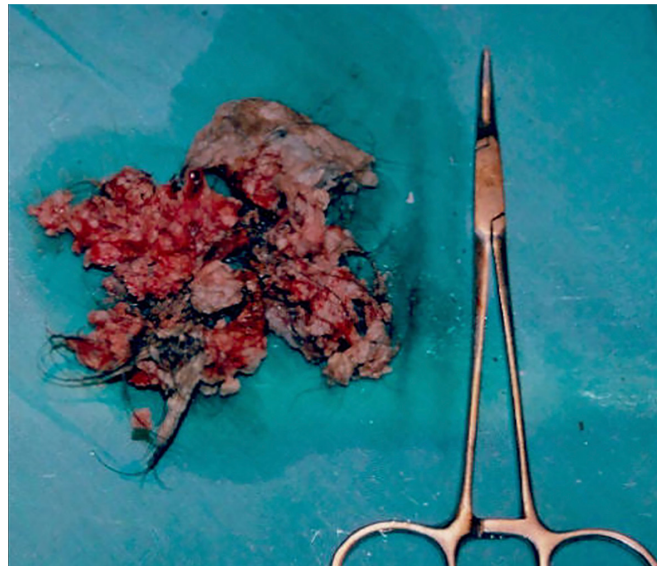
**Figure 1** CT scan image at first presentation.

sick patients had been managed successfully with hand ventilation when indicated. Considering the risk of a potentially fatal rebleed and the other constraints described above, the authors felt that they could confidently perform the pneumonostomy procedure with the facilities available after obtaining informed high-risk consent.

Anaesthesia was provided with an Epstein-Mackintosh-Oxford (EMO) apparatus using di-ethyl ether (anaesthetic ether) and atmospheric air (figure 2). The patient was induced with thiopentone 100mg intravenously and succinylcholine 100mg intravenously, followed by pancuronium 4mg intravenously, intubated with a reusable Rusch 7.0 endotracheal tube and a right posterolateral thoracotomy was done. Monitoring was done with intermittent manual sphygmomanometry, pulse oximetry and electrocardiography. A large firm mass was found in the lung with some areas that were cystic on palpation. Dense adhesions were encountered between the mass and the chest wall; when these were separated, the cystic part of the lesion was inadvertently opened causing the drainage of sebaceous material and a hair ball was removed (figure 3). Palpation within the cavity showed that part of the wall contained firm material, presumably bone and cartilage. These findings indicated that the patient had a teratoma but the team was unprepared for a major excision. The full extent of the lesion was not obvious and in the absence of facilities for frozen section and an in-house pathologist, a histopathological diagnosis could not be made during surgery. Considering these factors and the significant constraints mentioned above, it was felt that the risks of attempting a major



**Figure 2** The Epstein-Mackintosh-Oxford apparatus (image used with permission from the Wood Library-Museum of Anesthesiology, USA).



**Figure 3** Sebaceous material and hair removed at first surgery.

resection were unacceptable. Therefore, the contents of the cavity and part of the cyst wall were removed and the chest closed with a pleural drain. Muscle relaxation was reversed using neostigmine 2.5mg intravenously with atropine 1.2mg intravenously and the patient was allowed to recover from the effects of ether in the recovery room. There was no significant intraoperative bleeding and no blood was transfused during surgery. The patient made a complete recovery and was discharged on the 10th postoperative day. As there was no local histopathology service, the excised tissue was sent to a reputed institution in South India. Histopathological examination reported a mature teratoma. The hospital paid the cost of the histopathology examination. The patient was advised a second surgery to remove the remaining tumour. Since she was symptom-free, she was unwilling to undergo a second surgery at that time.

### Second presentation

The patient presented with recurrent small volume haemoptysis 4 years later (figure 4). She was married and had her first child at that time. In the intervening period, the hospital had been modernised with better equipment. Chest X-ray, performed this time with a Siemens 100 mA machine, showed a recurrent lesion in the same location that had grown in size (figure 5). The authors were still the only consultants but there were four junior doctors. The hospital used scarce resources to develop facilities for patients at the cost of facilities for staff who lived without electricity in their quarters for the first 14 years after the hospital was restarted. There was a 'high-dependency unit' with multiparameter monitors although a ventilator was yet to be installed. CT scan was not repeated as the chest X-ray clearly showed recurrence of the tumour with calcification along the borders, a histopathological diagnosis was available from the earlier biopsy and the expense of travel and another CT could be avoided. Surgery was planned and done as an elective procedure after obtaining informed consent. Anaesthesia was provided with a BOC Boyle machine using halothane, oxygen and nitrous oxide with the patient induced with thiopentone 100mg intravenously and succinylcholine 100mg intravenously, followed by pancuronium 4mg intravenously with pain relief provided by pentazocine 30mg intravenously. The patient was intubated with a Portex 24 left double lumen tube, with continuous monitoring

## Timeline

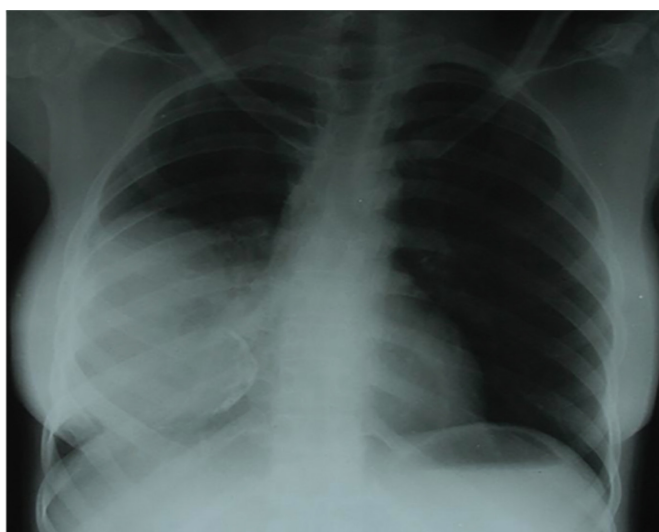
Year 0	4 Years later	20 years later
<b>Presentation and Explorative Surgery</b> <ul style="list-style-type: none"> <li>•Patient presents with history of massive haemoptysis</li> <li>•Imaging shows lesion in right lung</li> <li>•Exploratory thoracotomy and partial excision done. Histopathology - mature teratoma</li> </ul>	<b>Recurrence and Definitive Surgery</b> <ul style="list-style-type: none"> <li>•Patient presents again with recurrent haemoptysis</li> <li>•Imaging shows recurrent tumor</li> <li>•Right middle lobectomy performed with uneventful recovery. Histopathology - mature teratoma</li> </ul>	<b>Long-term Follow-up</b> <ul style="list-style-type: none"> <li>•Patient is asymptomatic</li> <li>•Seen by authors incidentally</li> <li>•Imaging shows no recurrence.</li> </ul>

**Figure 4** Timeline of presentations (graphic created by VAI).

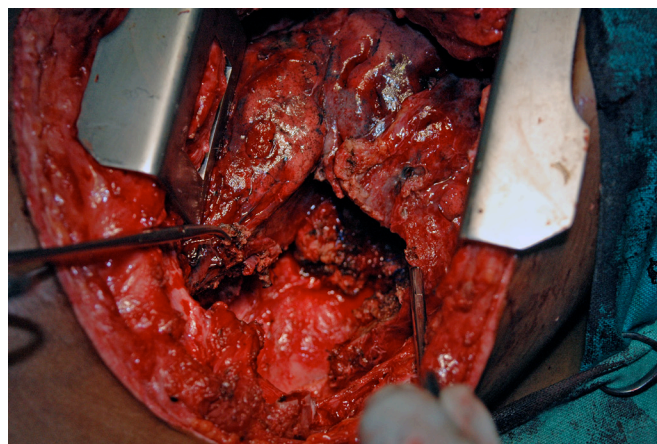
of oxygen saturation, electrocardiography, end-tidal carbon dioxide and invasive blood pressure. The chest was opened through the previous scar. Extensive adhesions were found between the chest wall and the lesion. Two overlying ribs were partially excised to provide better access. When the lung was freed from the chest wall, it was noted that the entire lesion was within the middle lobe of the lung. The horizontal and oblique fissures were dissected at the hilum until the inferior lobar bronchus was exposed. The vessels and bronchial branches entering into the right middle lobe were divided and ligated, excising the lobe. No obvious cause for haemoptysis was found during surgery. After completion of lobectomy, the double lumen tube was removed and replaced with a Portex 7.0 endotracheal tube; ventilation showed that the upper and lower lobes were inflating well with no air-leak and the chest was closed with a pleural drain (figure 6). Muscle relaxation was reversed using neostigmine 2.5 mg intravenously and atropine 1.2 mg intravenously. The patient made an unremarkable recovery and was discharged on the tenth postoperative day. A chest X-ray at follow-up about a month after surgery showed only postoperative changes and the patient was not symptomatic. Histology was reported as mature teratoma with no evidence of malignancy.

### Follow-up

Sixteen years after the second surgery, the authors met the patient incidentally and found her to be asymptomatic. She had been lost to follow-up but had no recurrence of haemoptysis



**Figure 5** Chest X-ray prior to second surgery.

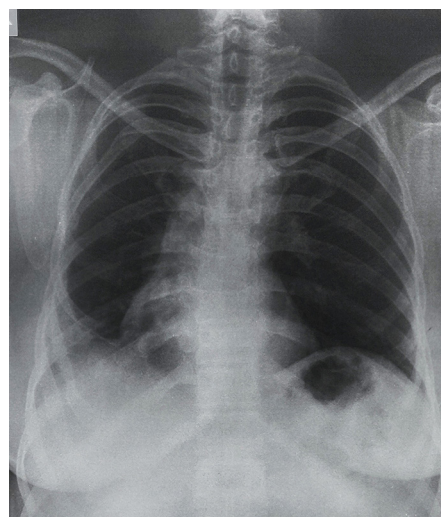


**Figure 6** Intraoperative photograph after middle lobectomy.

or any other symptoms. She was requested to visit the hospital where a chest X-ray showed some pleural thickening and post-operative changes but no recurrence of the tumour (figure 7).

### GLOBAL HEALTH PROBLEM LIST

- ▶ The correct diagnosis of rare conditions is more likely to be missed in remotely situated low-resource settings for want of adequate diagnostic facilities.
- ▶ Decision making in emergent situations has to take into account objective assessment of the facilities available in the institution, the cost and risk versus benefit of the proposed plan, logistics and the patient's informed consent.
- ▶ Many low-income and middle-income countries have low per capita national investment in healthcare resulting in large out-of-pocket expenditure (OOPE) causing catastrophic health expenditure and destitution. Further, social and economic factors cause patients from poor communities, especially those living in remote rural areas, to fear visiting hospitals with self-imposed delays and denials of treatment which contribute to poor healthcare outcomes.
- ▶ Secondary-level hospitals with limited-resources fear taking on the risks of treating patients with limited financial means. They refer to distant hospitals leaving patients with the choice of either going home untreated or incurring expenses



**Figure 7** Chest X-ray 16 years after second surgery.

that may push their families into destitution while seeking treatment in distant hospitals.

- ▶ Multiple factors lead to inequitable access to ethical medical management.
- ▶ Fear of litigation causes doctors and hospitals to 'play safe' by following rigid 'gold-standard' guidelines, which do not allow the flexibility to treat patients effectively by using services at their disposal and considering the financial means available to patients.

## GLOBAL HEALTH PROBLEM ANALYSIS

### Rural hospital facilities

The hospital had facilities to do chest X-rays using a very basic machine and this showed an ill-defined rounded opacity in the right lower zone. To understand the lesion and its relationships better, the patient was referred for a CT scan to a centre 100 km away. The reports of the scan were that there was a cystic lung lesion, probably hydatid. Bronchoscopy, angiography and better radiological facilities were possible only at a large city, 400 km and 12 hours away. Further imaging options were therefore not pursued considering the risks of bleeding in transit and the unwillingness of the patient to be referred further away.

Cystic lesions of the lung include congenital cysts, parasitic cysts and abscesses. Haemoptysis is not a common symptom with most patients having these conditions presenting with cough or fever. In this case, the CT scan was reported by a radiologist in another hospital as a lung cyst, probably hydatid. Pathognomonic findings for teratoma on CT scanning, such as the presence of teeth, were not observed. Our hospital had treated several patients with hydatid cysts of the liver, so a presumptive diagnosis of pulmonary hydatid cyst was reasonable at initial presentation to the hospital. An intrapulmonary cystic teratoma was not considered due to its extreme rarity and unusual location, the diagnosis was suspected during the initial exploratory thoracotomy and confirmed by subsequent histopathological examination.

Teratomas are most common in a gonadal location. The most common extragonadal site is the mediastinum. Intrapulmonary location for a teratoma was described in 1839 by Mohr and is very rare with less than 100 cases reported as of 2015.<sup>1</sup> They are commonly found in the upper lobes and the authors are unable to find any report of a right middle lobe location in the literature. Intrapulmonary teratomas may have varied radiological findings and have been confused with hydatid cyst of the lung.<sup>2</sup> Those with thicker walls and/or surrounded by solid areas, nodules or consolidation could be reported as cavitary lesions. They are postulated to arise from pluripotent stem cells of the third pharyngeal pouch or the ventral foregut which migrate along the developing lung bud.<sup>3</sup> Rarely, teratomas of the mediastinum and lung can occur in the same patient.<sup>4</sup> Lung teratomas are often cystic and may contain hair, bone, cartilage, sebaceous material, nervous tissue and skin. They commonly present with dry cough and sometimes with haemoptysis. Trichoptysis has been reported and when it occurs, may facilitate preoperative diagnosis.<sup>4</sup> They occur equally in males and females and present most commonly in the second and third decades of life. Malignant transformation can occur, with the most common pathology being squamous-celled carcinoma.

In this case, it is possible that erosion of surrounding vessels and bronchi led to haemoptysis. No definite cause of haemoptysis was found during surgery and bronchoscopy and angiography were not available.

### Decision making in austere circumstances

At first presentation, the patient was stable after receiving four units of blood at another hospital. Surgery is indicated for life-threatening haemoptysis in a situation in which the cause of bleeding can be treated by the intervention and the origin of the bleeding has been specifically and reliably located (strong recommendation, 1B—SEPAR guidelines).<sup>5</sup>

With a presumptive diagnosis of hydatid cyst of the lung, the pneumonostomy procedure was planned.<sup>6</sup> During the surgery, efforts were made to avoid breakage of the cyst and cyst fluid leakage because this can cause anaphylaxis; the anaesthesiologist was aware of this possibility and was prepared to manage it. Further, viable parasite present in leaked cyst fluid can cause implantation and recurrence. The risk of this can be reduced by irrigation of the cyst with scolicidal agents. In this case, the risk was reduced by packing the surrounding area with surgical swabs soaked in a scolicidal agent (10% povidone iodine). Hydatid cysts of the lung are often solitary and grow within lung tissue stretching surrounding bronchi and vessels around them. When an intrapulmonary hydatid cyst is surgically removed, weakened bronchial walls lead to multiple perforations and significant air-leak which can cause collapse of the ipsilateral lung, bronchopleural fistula, significant hospital stay and loss of lung capacity. This can be avoided by using the pneumonostomy technique where the cavity is closed around a tube which is exteriorised to the atmosphere, creating a broncho-atmospheric fistula, similar to a tracheostomy. Since air does not leak into the pleural cavity, the lung does not collapse and patients recover quickly and completely with no loss of lung tissue as the bronchial perforations heal over time. This surgery is simple and easy to perform in a low-resource setting.

At surgery, it was noted that the lesion was adherent to the chest wall and was of variable consistency on palpation. A thinned-out part of the lesion had inadvertently opened during the release of adhesions and thick sebaceous material appeared. When the cavity was opened, a hair ball was found and palpation of the internal surface showed hard, firm and soft areas. These findings indicated that this was an intrapulmonary teratoma but since frozen section facility was not available, it was not possible to have histopathological confirmation or exclude a malignant component. Since there was no intrathoracic or airway bleeding after partial removal of the tumour, it was felt that an immediate rebleed would not occur. It was therefore decided to close the chest with drainage and reoperate after confirming the diagnosis on an elective basis or refer the patient to a specialist centre. When operating in similar challenging situations, partial excision or decompression of an intrapulmonary teratoma presenting with massive haemoptysis may be sufficient to prevent an immediate re-bleed.

The EMO anaesthesia machine was used during the first surgery—this machine was used at our hospital for more than 10 years with excellent results, we stopped using it only after it developed a malfunction and anaesthetic ether was no longer available in India. It was developed for field hospitals during World War II and is a temperature compensated, calibrated, draw-over ether vaporiser, commonly used with hand-operated 'Oxford' bellows.<sup>7</sup> Ether is an effective anaesthetic agent and adequate anaesthesia can be provided with atmospheric air in most circumstances. Additional oxygen needs to be supplemented only when required. Ether anaesthesia is safe, even in non-expert hands, although it is slow in onset and recovery. Induction may be hastened by using halothane with an attached Oxford Miniature Vaporizer but this was not available at the

hospital. The other disadvantage of ether is its flammability. This is a significant risk when used with oxygen and electrocautery. In our case, oxygen was used only for preoxygenation and haemostasis was achieved primarily using ligatures and pressure with cautery used sparingly when oxygen was not in use. This technology is now considered obsolete and is not taught in modern anaesthesia training programmes. With modern anaesthesia performed by Boyle type machines that use newer anaesthetic agents and with the free availability of pressurised gases, anaesthetic ether is not manufactured in most countries. Penlon, the company that was producing the EMO machine has stopped manufacture due to absent demand. From a global health perspective, the EMO machine may still be used to provide safe, cheap, reliable anaesthesia in situations where electricity and oxygen supply are unreliable; it can be safely administered by trained anaesthesia providers and it should be considered as a realistic option in such resource limited settings. Ether is inexpensive, safe and provides excellent intraoperative pain control which extends several hours into the postoperative period and should be considered as a realistic agent for anaesthesia in situations where surgery needs to be performed under conditions of multiple major resource constraints—such as the conditions described above.<sup>8</sup>

Surgical excision provides a complete cure for benign mature teratomas of the lung and that was the procedure adopted at the second surgery. Extensive scarring and adhesions between the lung and chest wall necessitated partial excision of two overlying ribs to gain adequate exposure. Since the tumour was entirely within the right middle lobe, a right middle lobectomy was performed. Symptom-free follow-up 16 years after definitive surgery shows that simple lobectomy of the involved lobe was sufficient for complete cure of this rare condition.

### Financial pressures on patients to pay for healthcare

India has a low national per capita investment in healthcare as a percentage of Gross Domestic Product (GDP) and this results in high OOPE for patients. The resulting financial vulnerability caused by a single large healthcare expenditure is especially acute in remote rural communities and among the poor and marginalised.<sup>9</sup> Within India, healthcare accessibility and outcomes vary enormously. The Government of India has identified eight particularly vulnerable states, including Assam, which are collectively called the 'Empowered Action Group States' with under-5 mortality rate, maternal mortality rate and other healthcare indices being used as surrogate markers of healthcare access and outcomes<sup>10</sup> Healthcare outcomes in these states are enumerated in Annual Health Surveys, both nationally and for individual states such as Assam.<sup>11 12</sup> Within most states, urban areas fare better than rural areas. India's rural:urban population ratio is 72:28 but the bed availability is exactly the opposite 28:72—government healthcare facilities are often poorly equipped/staffed and private 'for profit' hospitals almost non-existent in many remote rural areas.<sup>13 14</sup> There are many challenges in attracting and retaining committed manpower in needy rural areas. Professionally, they need to manage with the facilities available to be able to provide healthcare at affordable costs and personally, they face multiple inconveniences at home which they should be willing to accept.

In this case, the income of the family rested on a sole income generating individual, the father, who was a marginal farmer. At the time of presentation, there was no provision for government-funded health insurance and all expenditure would have been out-of-pocket for the family. The fear of the resulting catastrophic

expenditure which could lead to sale of vital assets or borrowing at unsustainable terms and destitution led the family to refuse consent for distant referral. Assessment of how much the patient could pay without selling vital assets was used to provide charitable funds for both surgeries.

Other factors that affect healthcare access include lower public confidence in free or subsidised public healthcare services. This leads families to spend beyond their capacity to finance healthcare needs by going to private healthcare providers.<sup>15</sup> Many families live in chronic poverty and a sudden expenditure is coped with by curtailing already limited family budgets for basic necessities like food and schooling. Families with limited means may become destitute as a consequence of decisions to borrow unsustainably to fund health intervention, selling off vital assets, taking children out of school and even resorting to debt bondage.<sup>16</sup> The authors have noted that when a family sells their home or other vital asset to finance healthcare expenditure, it creates a chilling effect in the local community and this fear leads to self-imposed delays and denials of healthcare. A significant percentage of OOPE on healthcare is spent on non-medical expenses.<sup>17</sup> A realistic evaluation of these factors should encourage governments and healthcare institutions to develop strategies to close gaps in access.<sup>18</sup>

Our hospital has observed many such events where delay and refusal of treatment, failure to report for follow-up as well as refusal of referral have occurred.<sup>19</sup> The hospital has developed a number of poor-centric strategies to alleviate fear and welcome poor patients to access its services. These strategies include ability-to-pay based pricing, equal access to services to all, hyper-tailored charity, removing cultural barriers through community engagement, revised gold standard treatment protocols as well as recruitment and retention of an efficient workforce. To illustrate one of these strategies, charity is 'hyper-tailored' based on the hospital's assessment of financial distress where patients and their relatives prefer to starve in hospitals to save scarce financial resources to pay for treatment, request discharge before a patient dies as the cost of transporting a dead body is much higher or need to sell vital assets to finance healthcare—these are painful decisions that a non-poor family would not take. Assessment of how much a family can pay without selling vital assets goes into the quantum of charity provided to a family.<sup>20 21</sup>

### Medical ethics in difficult circumstances

Fear of litigation causes many hospitals to overinvestigate, refuse treatment and refer patients that they are capable of treating. In addition, tertiary care is not available in most rural areas and distant travel may exceed 1000 km, which causes significant costs besides delay in accessing care which could impact outcomes. The number of cardiothoracic surgeons available in India is inadequate to service the needs of cardiac surgery. There are even fewer who specialise in general thoracic surgery. It is extremely difficult for a patient with a lung problem to access the services of a dedicated thoracic surgeon in what would be considered reasonable travel distance. The amount reimbursed to hospitals by public health schemes for lung surgical procedures is insufficient to meet the costs of thoracic procedures discouraging hospitals from providing thoracic surgical services.<sup>22</sup> Some parts of the country, such as northeast India do not have centres which were operating on such patients at the time when this patient was treated, with referral to mainland India being expensive and requiring significant travel time. The authors have noted that when local hospitals refer to distant hospitals, many poor patients simply go home and may die or suffer complications

of untreated disease—this could have been the outcome in our case if the patient had been refused treatment. Each hospital should objectively examine the resources at their disposal and constraints under which they function to accept reasonable risks when the benefits are great.

The four modern principles of medical ethics, beneficence, non-maleficence, autonomy and justice, should be used to modify guidelines.<sup>23</sup> In this case, during the first surgery, considering the emergent situation and potential of a fatal rebleed, a simple exploratory thoracotomy, pneumonostomy or as it turned out, partial excision to stop haemoptysis was a reasonable risk to accept and therefore beneficence was exercised. However, in the presence of serious local constraints with the full extent and histopathological diagnosis unknown and contralateral lung protection unavailable, not assuming unacceptable risk and not proceeding with a lung resection was non-maleficence. Understanding the situation of the patient and respecting her wish not to be referred and operating with full informed consent after understanding risks involved is autonomy. Providing all possible access to quality treatment within the capacity of the institution in spite of severe constraints irrespective of poverty and rural residence is justice. Thus, the four pillars of ethical treatment have been employed in this case.

A definitive excision at a second sitting was performed in an elective setting with most of the constraints mentioned above resolved resulting in a successful outcome. This may be the most realistic way to treat such conditions ethically in resource-limited settings and among poor remote rural communities,

using a strategy of ‘best possible treatment at the time under the circumstances’.

### Guidelines for ethical and professional treatment in austere environments

‘Gold-standard’ guidelines and protocols assume that the hospital has access to the latest equipment, manpower and when this is not possible, the patient can be transported safely and quickly without affecting the life of the patient or the financial security of the family. This ideal scenario is considerably modified in many countries as described above. Guidelines should be revised on a case-by-case basis taking into account an objective evaluation of the constraints and assets at the disposal of the operating team as well as the financial resources available to the patient to reach ‘revised gold standards’ that are realistic in that particular scenario. In this case, the EMO machine and anaesthetic ether were used in the first operation. Although both are considered obsolete, the older, time-tested equipment and agent are realistic and possibly best choices in many parts of the developing world, including war-torn areas. Our hospital used such a protocol in the management of this patient and also trains Global Health residents, doing their MD (Global Health and Tropical Medicine) course from the Netherlands, to create and use such protocols effectively under challenging circumstances across the globe.<sup>24</sup>

**Acknowledgements** We wish to acknowledge Dr. Joseph Duerksen, visiting anaesthesiologist who donated a left double lumen tube on hearing about this patient and Dr. Kenny David, visiting orthopaedic surgeon who assisted the second surgery.

**Contributors** VAI and AM were involved in the management of the patient. VAI wrote the manuscript and created the graphic (figure 4). Both authors were involved in review of literature, editing the draft and reviewing the manuscript.

**Funding** The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

**Competing interests** None declared.

**Patient consent for publication** Consent obtained directly from patient(s)

**Provenance and peer review** Not commissioned; externally peer reviewed.

**Open access** This is an open access article distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>.

Case reports provide a valuable learning resource for the scientific community and can indicate areas of interest for future research. They should not be used in isolation to guide treatment choices or public health policy.

### ORCID iD

Vijay Anand Ismavel <http://orcid.org/0000-0002-2329-596X>

### REFERENCES

- 1 Ditah C, Templin T, Mandal R, *et al*. Isolated intrapulmonary teratoma. *J Thorac Cardiovasc Surg* 2016;152:e129–31.
- 2 Ram D, Padmanabhan R, Panchanatheeswaran K, *et al*. Intrapulmonary teratoma masquerading as hydatid cyst of the lung. *Indian J Thorac Cardiovasc Surg* 2019;35:71–3.
- 3 Präuer HW, Mack D, Babic R. Intrapulmonary teratoma 10 years after removal of a mediastinal teratoma in a young man. *Thorax* 1983;38:632–4.
- 4 Agarwal R, Srinivas R, Saxena AK. Trichoptysis due to an intrapulmonary teratoma. *Respiratory Care* 2007;53:1779–81.
- 5 Cordovilla R, Bollo de Miguel E, Nuñez Ares A, *et al*. Diagnosis and treatment of hemoptysis. *Arch Bronconeumol* 2016;52:368–77.
- 6 Anand V, Sen S, Jacob R, *et al*. Pneumonostomy in the surgical management of bilateral hydatid cysts of the lung. *Pediatr Surg Int* 2001;17:29–31.
- 7 Lo R. Epstein-Macintosh-Oxford inhaler (EMO anaesthetic apparatus). *Hong Kong Med J* 2020;26:358–9.

### Patient's perspective

I had an episode of massive coughing of blood 20 years ago and was taken to the local district hospital where four units of blood was transfused. I was referred to this hospital and was referred for a CT scan. An emergency surgery was done. After receiving the biopsy report, I was told that a second surgery was required but I was fine at that time and refused because of fear. I got married 18 months after the first surgery and after giving birth to my first son, I again started coughing out blood. X-ray showed that the tumour had returned and I agreed to a second surgery. Now, 16 years later, I am completely fine. I thank God and the hospital for saving my life.

### Learning points

- ▶ Mature teratomas of the lung should be considered in the differential diagnosis of haemoptysis when imaging shows a cystic or cavitary lung lesion.
- ▶ Partial excision of a mature cystic teratoma may be sufficient to prevent potential massive haemoptysis in an emergent setting.
- ▶ The Epstein-Mackintosh-Oxford apparatus and diethyl ether can be used safely to provide anaesthesia in resource-limited settings.
- ▶ Intrapulmonary teratomas can occur in the right middle lobe and treated successfully by right middle lobectomy.
- ▶ The social and economic conditions of the patient and resources at hand should be objectively analysed to create ‘revised gold standards’ to treat patients in resource-limited settings.

- 8 Chang CY, Goldstein E, Agarwal N, *et al.* Ether in the developing world: rethinking an abandoned agent. *BMC Anesthesiol* 2015;15:2–5.
- 9 Garg CC, Karan AK. Reducing out-of-pocket expenditures to reduce poverty: a disaggregated analysis at rural-urban and state level in India. *Health Policy Plan* 2009;24:1–13.
- 10 Kumar S, Sahu D. Socio-Economic, demographic and environmental factors – effects on under-five mortality in Empowered action group state of India: an evidence from NFHS-4.
- 11 National rural health mission, health and population policies, chapter 2, P2. Available: <https://main.mohfw.gov.in/sites/default/files/CHAPTER%202.pdf> [Accessed 8 May 2022].
- 12 Annual health survey 2012-13 Factsheet – Assam. Available: [https://hfw.assam.gov.in/sites/default/files/swf\\_utility\\_folder/departments/hfw\\_lipl\\_in\\_oid\\_3/menu/information\\_and\\_services/FACTSHEET-Assam%20-%2012%20-13.pdf](https://hfw.assam.gov.in/sites/default/files/swf_utility_folder/departments/hfw_lipl_in_oid_3/menu/information_and_services/FACTSHEET-Assam%20-%2012%20-13.pdf) [Accessed 8 May 2022].
- 13 Rural-Urban population distribution, census 2011, office of the registrar general and census commissioner, Ministry of home Affairs, government of India. Available: [https://censusindia.gov.in/census\\_data\\_2001/india\\_at\\_glance/rural.aspx](https://censusindia.gov.in/census_data_2001/india_at_glance/rural.aspx) [Accessed 21 Dec 2021].
- 14 Study on the not-for-profit Hospital model in India, NITI Aayog, July 2021, p11. Available: [https://www.niti.gov.in/sites/default/files/2021-09/Not-for-profit-HospitalReport\\_29\\_7\\_2021.pdf](https://www.niti.gov.in/sites/default/files/2021-09/Not-for-profit-HospitalReport_29_7_2021.pdf) [Accessed 8 May 2022].
- 15 Yadav R, Zaman K, Mishra A, *et al.* Health seeking behaviour and healthcare utilization in a rural cohort of North India. *Healthcare* 2022;10:757.
- 16 Flores G, Krishnakumar J, O'Donnell O, *et al.* Coping with health-care costs: implications for the measurement of catastrophic expenditures and poverty. *Health Econ* 2008;17:1393–412.
- 17 Ambade M, Sarwal R, Mor N, *et al.* Components of out-of-pocket expenditure and their relative contribution to economic burden of diseases in India. *JAMA Netw Open* 2022;5:e2210040.
- 18 Ismavel V, Miriam A. Massive abdominal tumor – more than a medical problem. *Curr Med Issues* 2017;15:249–51.
- 19 Flint C, Ismavel VijayA, Miriam A. The Makunda model: an observational study of high quality, accessible healthcare in low-resource settings. *CJGH* 2020;7:37–51.
- 20 Evans JR, Hall KL, Warford J. Health care in the developing world: problems of scarcity and choice. *N Engl J Med* 1981;305:1117–27.
- 21 Ismavel VA, Kichu M, Hechhula DP, *et al.* Right paraduodenal hernia with extensive bowel gangrene treated with staged surgery: a Bogota bag followed by resection in a low-resource setting. *BMJ Case Rep* 2021;14:239250.
- 22 Yendamuri S. Thoracic surgery in India: challenges and opportunities. *J Thorac Dis* 2016;8:S596–600.
- 23 Beauchamp TL, Childress JF. *Principles of biomedical ethics*. New York (NY): Oxford University Press, 2009: 162–4.
- 24 Ismavel VA. *Chapter – Closing the Gap – using Global Health Doctors in book "Into the World" 1st Edition*. Editor: Mathijs Botman, Publisher: Uitverij Boekschap, 2017.

Copyright 2022 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <https://www.bmj.com/company/products-services/rights-and-licensing/permissions/>  
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

#### Customer Service

If you have any further queries about your subscription, please contact our customer services team on +44 (0) 207111 1105 or via email at [support@bmj.com](mailto:support@bmj.com).

Visit [casereports.bmj.com](http://casereports.bmj.com) for more articles like this and to become a Fellow