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Thoracoscopic Resection of Congenital Cystic Adenomatoid Malformation in A Patient With Fused Lung Fissure Using Hookwire

--Manuscript Draft--

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Abstract:	Thoracoscopic resection is being used more commonly for the treatment of congenital cystic adenomatoid malformation (CCAM) in neonates and infants. However in the rare case of CCAM with congenital absence of lung fissure, thoracoscopic lobectomy cannot be performed safely. Moreover if the lung lesion is deep-seated and cannot be visualized on the pleural surface, wedge resection may result in residual lesion. Here we reported our approach of thoracoscopic resection under hookwire guidance to tackle this problem.
Response to Reviewers:	Dear Innovations: Technology and Techniques in Cardiothoracic and Vascular Surgery editors, We thank the reviewers for their constructive comments on our manuscript entitled, “An alternative approach to thoracoscopic resection of congenital cystic adenomatoid malformation in patient with fused lung fissure using hookwire”. We have replied to the reviewer’s queries in a point-by-point fashion and made the necessary corrections in the revised manuscript. We would be grateful if our manuscript could be reviewed again for consideration of publication in Innovations: Technology and Techniques in Cardiothoracic and Vascular Surgery. Response to reviewer 1: 1)The experience from other authors were addressed in the discussion section and references were added as per suggestion. 2)The potential complications of CT-guided hookwire insertion were further discussed and elaborated. 3)We acknowledge that this technique may be more applicable to lesions with favourable characteristics and advised readers to exercise their clinical judgement cautiously. 4)We would like to change the title to “Thoracoscopic resection of congenital cystic

	adenomatoid malformation in a patient with fused lung fissure using hookwire" as suggested. Other editorial comments were edited accordingly. We hope once again that this revised version can clarify queries from the reviewers. Your kind consideration of our manuscript for publication would be greatly appreciated. Yours sincerely, Dr. CT Lau

FINAL

Case Report

Title: Thoracoscopic resection of congenital cystic adenomatoid malformation in a patient with fused lung fissure using hookwire

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FINAL

Abstract:

Thoracoscopic resection is being used more commonly for the treatment of congenital cystic adenomatoid malformation (CCAM) in neonates and infants. However in the rare case of CCAM with congenital absence of lung fissure, thoracoscopic lobectomy cannot be performed safely. Moreover, if the lung lesion is deep-seated and cannot be visualized on the pleural surface, wedge resection may result in residual lesion. Here we reported our approach of thoracoscopic resection under hookwire guidance to tackle this problem.

Key words: Thoracoscopy, Minimal invasive, Congenital cystic adenomatoid malformation, Hookwire.

Introduction

With more routine antenatal ultrasonography screening and increasing awareness of clinicians, the diagnosis of congenital cystic adenomatoid malformation (CCAM) has increased dramatically in the recent decade. CCAM is a lung lesion characterized by the cystic dilatation of terminal respiratory bronchioles. This multicystic lung mass is the result of abnormal embryological development of the lower respiratory tract and does not participate in gaseous exchange. Surgical resection is usually recommended due to the risk of recurrent chest infection and malignant transformation.

Thoracoscopic surgery is now commonly performed in many pediatric thoracic conditions. The benefits of thoracoscopic resection of CCAM over conventional open approach have been well demonstrated in previous studies [1, 2]. Traditionally, lobectomy has been considered as the gold standard for complete removal of the lesion to avoid residual disease. However, thoracoscopic lobectomy can be even more technically demanding in patients with fused lung fissure, and pneumonectomy may also be required in multilobar disease. Wedge resection has been suggested by some authors to tackle these problems, but its application has been limited

to small, well confined lesions with favorable anatomical location due to the risk of post-operative air leak and residual disease [3, 4].

Herein, we reported our technique to perform thoracoscopic wedge resection of a poorly demarcated CCAM lesion with fused lung fissure under hookwires guidance.

Case Report

A one-year-old girl with antenatally diagnosed cystic left lung lesion was referred to our unit for further management. Computed tomography (CT) of thorax showed a CCAM lesion at the apicoposterior area of the left upper lobe, extending inferiorly to involve the apical area of the left lower lobe, measuring 3.6 x 2.5 x 3.7 cm (figure 1). The left oblique fissure was fused at the superoposterior section where the lesion was located. No systemic arterial supply or venous drainage was observed.

Elective operation with CT guided hookwires insertion followed by thoracoscopic excision was subsequently arranged. Patient was put under general anesthesia and intubated before transferring to the CT suite. Since

the lesion was located on the posterior side, patient was put into a prone position on the CT table for hookwire insertion from the back (figure 2A and 2B). Two 18-gauge, 120 mm long spiral wire needles (Somatex, Rietzneuendorf, Germany) were inserted to the superior and inferior borders of the lesion under CT guidance respectively (figure 3A and 3B).

Patient was then transferred to the operation theatre with the hookwires in situ. The patient was put into the lateral decubitus position with single lung ventilation using bronchial blocker. A 5mm port was put at the fifth intercostal space at the anterior axillary line for insertion of the telescope to determine the anatomy. Pneumothorax was created with carbon dioxide infusion at low pressure (4 mmHg) and low flow (1 L/min) to compress the lung. Thoracoscopic view confirmed the left oblique fissure was completely fused, and the CCAM lesion was not visible on the lung surface. The two hookwires inserted under CT guidance were in-situ and helped to mark the superior and inferior borders of the CCAM lesion (figure 4). Two additional ports, 5 mm and 12 mm, were inserted at the sixth and seventh intercostal space as working ports respectively. Wedge resection of CCAM was carried out with endoscopic stapler (Endo GIA stapler, Covidien, Mansfield, MA USA).

The two hookwires served as marker of resection margin and direction of stapling planes where they converge behind the CCAM lesion. The resected lung specimen, measuring 5 x 2 cm, was delivered via the 10 mm port (figure 5). Intra-operative leak test confirmed air tightness of the staple line. 16 French chest drain was inserted via one of the 5 mm ports at the end of procedure.

Pathology of the specimen confirmed the diagnosis of CCAM with clear resection margin. Patient recovered well after the operation with no complication including bleeding, pneumothorax or wound infection. Chest drain was removed the next day. She was discharged two days after the operation. Subsequent follow-up imaging showed no residual lesion and patient remained asymptomatic thereafter.

Discussion

Since the first report of thoracoscopy in children by Rodgers et al in the 1970s, there had been a dramatic growth in the application of this minimal invasive approach [5, 6]. CCAM is one of the most common elective conditions for thoracoscopic surgery in infants. Lobectomy was

conventionally the treatment of choice due to the low risk of prolonged air leak and residual lesion. Since children can usually tolerate lobectomy well, some authors now even advocate thoracoscopic lobectomy in early infancy, with the theoretical benefit of compensatory lung growth [1, 7].

Fused lung fissure poses a specific difficulty for minimal invasive thoracic surgeons. Cadaveric studies suggested that it was not such a rare phenomenon with prevalence of 10 to 20% [8]. There had been reports of using energy device and blunt dissection of the lung parenchyma along the presumed anatomical fissure until the pulmonary vessels were exposed for a proper lobectomy [1, 9]. However, Gomez-Caro et al commented that this approach was associated with higher risk of persistent air leak and inadvertent injury to underlying vessels in their study [10]. This was because minor air leak from fragile lung parenchyma was often encountered even if the latest tissue sealing device was employed. The use of endoscopic stapler to transect the fused fissure may reduce air leak but carry an even bigger risk of damaging major structures located deep inside the lung parenchyma if it was applied empirically.

Fissureless technique involving the division of pulmonary vessels and bronchus at the hilum before dissecting the fused lung fissure using stapler has been well established in adults. This approach was associated with less post-operative air leak and shorter hospital stay. Nonetheless, this approach can be technically difficult if not impossible in small infants where the thoracic space is very limited. In addition, it carries the risk of residual lesion with recurrent chest infection because there is no guarantee of complete CCAM excision, especially if the lesion was located close to the interlobar area or crossed the imaginary dissection plane. In these cases of multilobar disease pneumonectomy may seem inevitable.

Wedge resection of CCAM has come to light in recent years as a rescue of multilobar disease. Advocators have extended its use to include lung-sparing surgery for solitary CCAM lesions [3, 4, 11]. The benefits of wedge resection include preservation of healthy lung parenchyma as well as larger airways, which was not possible in case of lobectomy. Johnson et al reported the largest series of thoracoscopic wedge resection in 2011 and confirmed its safety and feasibility. Rothenberg et al proposed thoracoscopic segmentectomy as a better alternative since the dissection was based on

anatomical segmental plane [11]. Yet residual lesions remained to be a concern and only highly selected group of patients were recruited in both studies. The two authors agreed that ideally the lesions should be small, focally confined to a particular segment and peripherally located. Visual cues and colour change on the pleural surface were also essential in facilitating a complete resection.

CT guided hookwire insertion is a well described technique to localize solitary pulmonary nodule in adult [12]. Experience in children were limited to marking the position of pulmonary nodules using single hookwire without defining the resection margin clearly [13, 14]. We believe with our application of hookwire as guidance for thoracoscopic CCAM resection help to circumvent the current obstacle. Even in the face of a large, deep-seated lung lesion with poor surface demarcation the hookwires can still guide us to complete a clear resection, while at the same time preserving normal lung parenchyma and larger airways. Despite the proposed advantage, this technique of hookwires guided thoracoscopic resection is not without its own limitations. Firstly, its application may be limited by the body size of patients since a 12 mm port and at least 4 cm intrathoracic space is required for

proper use of an endoscopic stapler. The selection of suitable surgical candidate can also be difficult because pre-operative CT scan may not be always diagnostic of fused lung fissure. Besides, hookwire insertion per se can lead to complications including pneumothorax, hemothorax and possibly marker dislodgement, which had all been well documented [12]. The potential problem of residual lesion, although minimized, required a larger scale study with more patients to truly eliminate. Thus application on lesion with unfavorable features had to be decided cautiously.

In conclusion, hookwires guided thoracoscopic resection of CCAM in patient with congenital absence of lung fissure is feasible and safe, even for deep-seated lesions.

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Figures

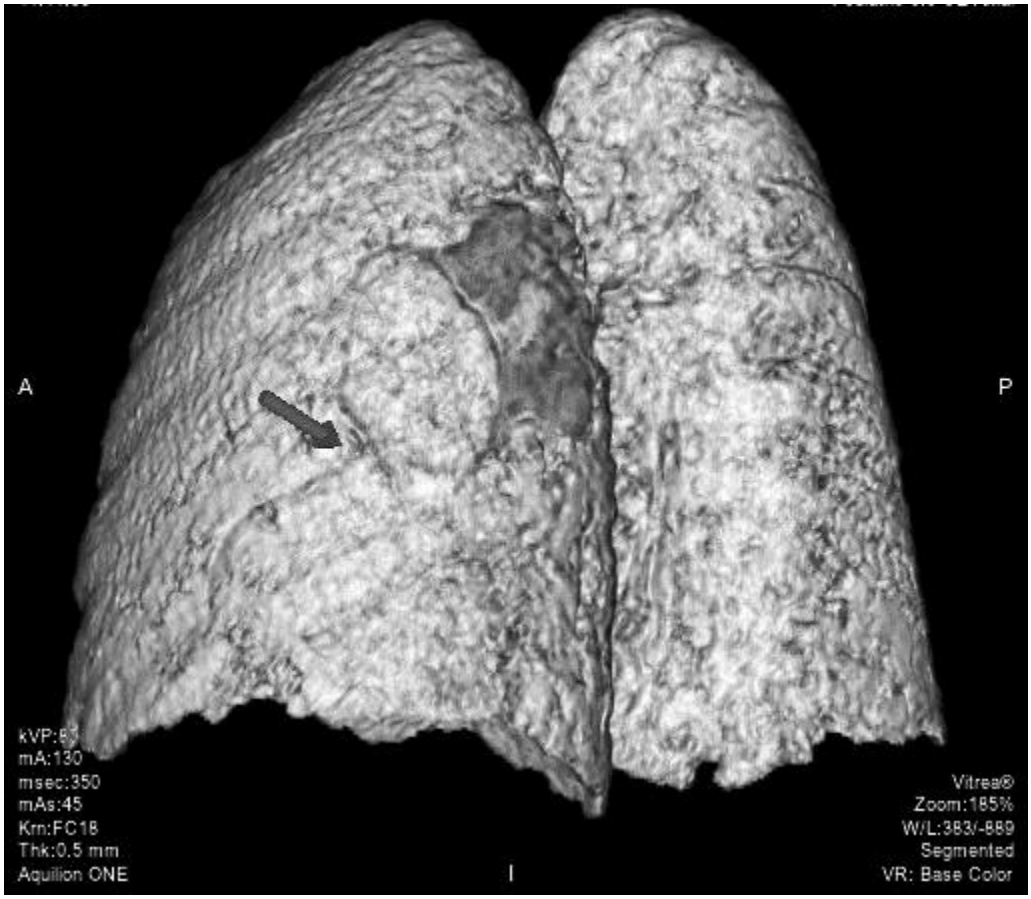

Figure 1: 3D reconstructed CT thorax image. Arrow pointing to the fused left oblique fissure. Dark area signifies the lesion.

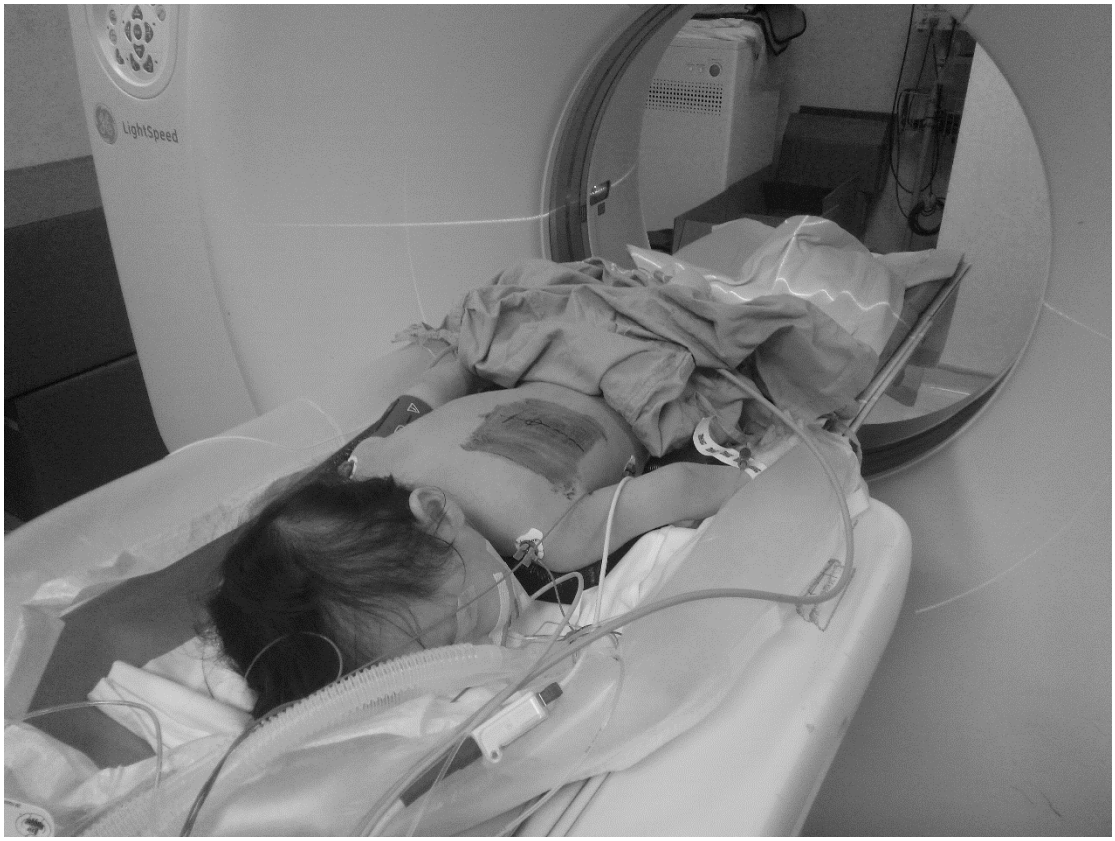

Figure 2A: Hookwire insertion point marked under CT guidance.

Figure 2B: External view after hookwires inserted.

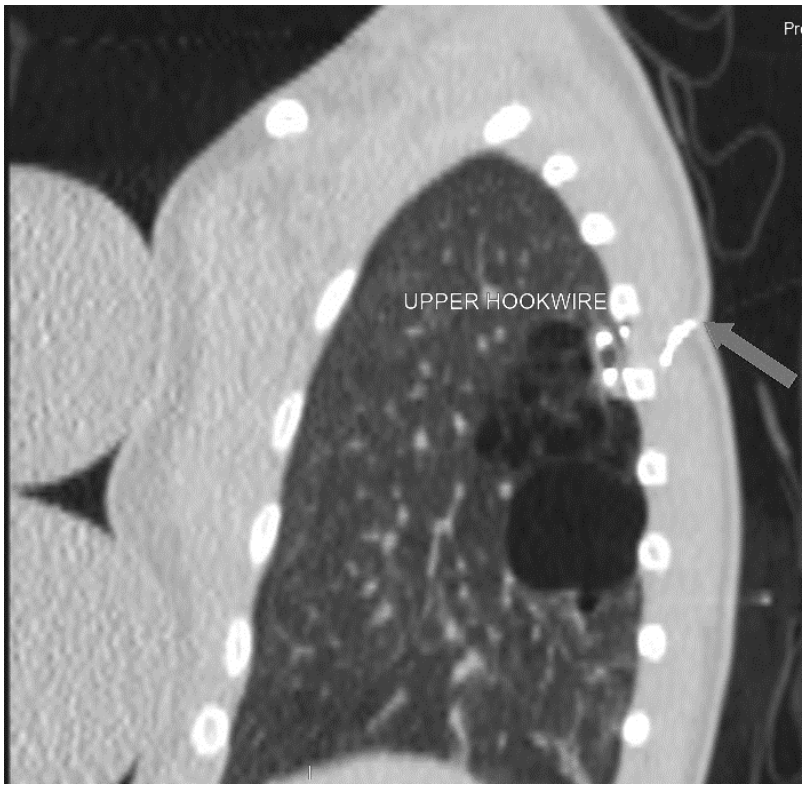

Figure 3A: Sagittal view CT image with arrow pointing to the superior hookwire.

Figure 3B: Sagittal view CT image with arrow pointing to the inferior hookwire.

Figure 4: Thoracoscopic view of the left lung. Note the fused left oblique fissure and that the CCAM lesion was not visible on the surface. The two hookwires marked the superior and inferior border of the lesion.

Figure 5: Resected lung specimen with the two hookwires in-situ.