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CASE REPORT

Minimally invasive resection of a marijuana-associated giant bulla: a case report

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Background

Smoking is known to be a causative factor for emphysema. Literature also supports the possibility that marijuana abuse increases the probability of giant apical bullae in young individuals. Surgical resection is the only cure for giant bullae, whilst also having a prophylactic role. Video-assisted Thoracoscopic Surgery (VATS) bullectomy is the new gold standard replacing thoracotomy.

Case Presentation

We present a 31-year old marijuana smoker with a history of multiple pneumothoraces who was found to have a large emphysematous bulla in the left upper lobe accompanied by a smaller bulla in the superior lingular segment. Even though he was asymptomatic at the time, VATS bullectomy was carried out in view of his multiple previous pneumothoraces. The patient recovered well after surgery, and a follow-up chest x-ray 2 weeks post-operatively showed a fully inflated lung.

Conclusion

This is the first case of giant bulla being removed via minimally invasive VATS in Malta. Apart from continuing to show the effectiveness and suitability of VATS in the treatment of this condition, we highlight the important link between marijuana and giant emphysematous bullae.

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Introduction

The first case report on giant bullous emphysema was published by Burke in 1937.[1] It is a rare manifestation of emphysema.

According to the Global Initiative for Chronic Obstructive Lung Disease (GOLD), chronic obstructive pulmonary disease (COPD) is defined as a "common, preventable, and treatable disease that is characterized by persistent respiratory symptoms and airflow limitation that is due to airway and/or alveolar abnormalities usually caused by significant exposure to noxious particles or gases".[2] The report continues to elaborate that small airway disease and emphysema are the two main constituents of COPD.

Emphysema is a pathological term, which refers to the destruction of airspace walls distal to the terminal bronchioles, with the creation of permanently enlarged airspaces but without any fibrosis.[3] In some patients, this continues to advance to bullous emphysema, which consists of multiple bullae within emphysematous lung parenchyma. By definition, bullae are air spaces which are at least 1cm in size, encompassed by walls made of visceral pleura and remnants of alveolar and interlobular septae.[4] When bullae occupy more than a third of the hemithorax they are located in, they are labelled as giant bullae.[5-6] If no complications occur, the usual development of giant emphysematous bullae is gradual enlargement, which would ultimately impair breathing. However, rare spontaneous regression of giant emphysematous bullae have been reported, with symptomatic and radiological improvement.[7]

Nowadays, the international gold standard treatment for this disease is VATS-bullectomy, replacing open surgery. We describe here the first case of a giant bulla resected in this minimally invasive way in Malta.

Case

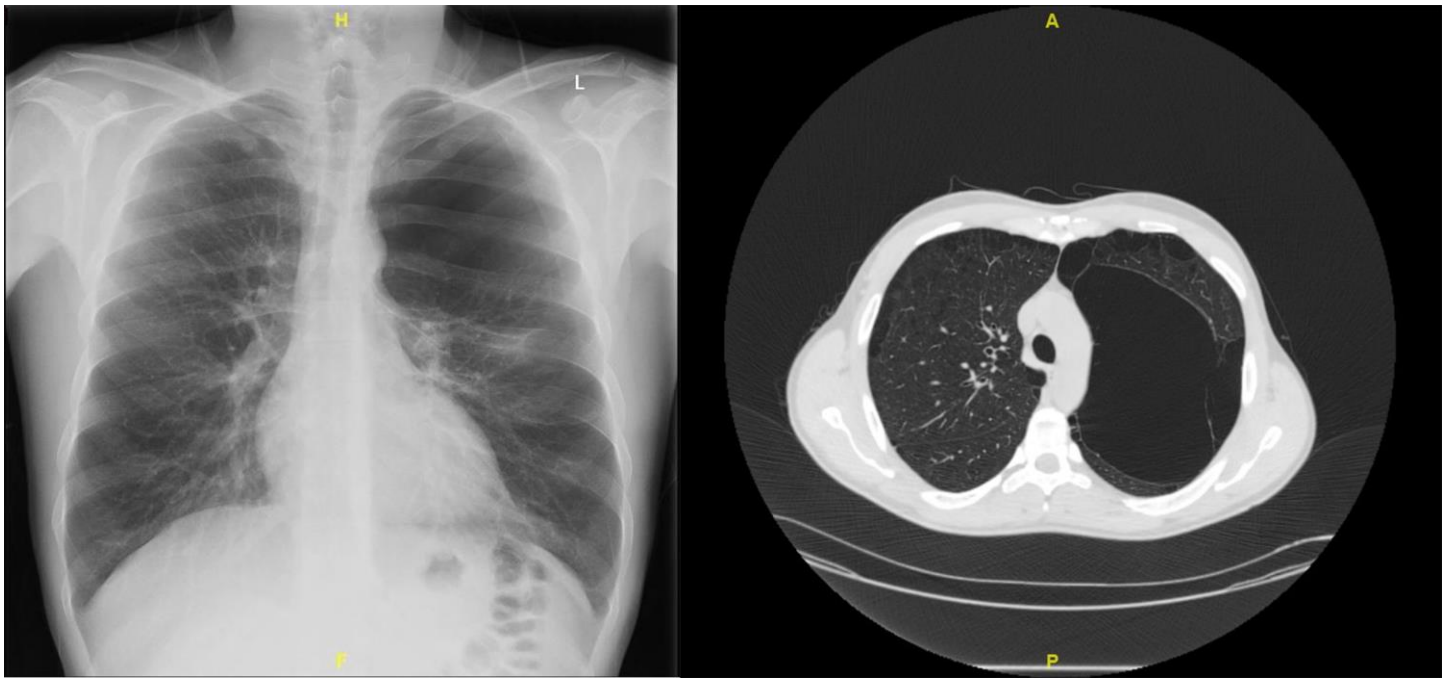
A 31-year-old patient, with a past history of right pneumothorax 3 years before, presented with a recurrent ipsilateral pneumothorax. This second pneumothorax was treated with a Seldinger insertion 12Fr intercostal chest drain for a few days. The lung re-inflated successfully, and the patient was referred electively to the cardiothoracic clinic.

His past medical history includes severe eczema requiring steroids and immunosuppressants. He is a cigarette smoker and has smoked approximately 1 packet a day for the past 17 years. He has also been smoking marijuana heavily, every day for the past 8 years. While being followed up 1 week later in the cardiothoracic clinic, he was not short of breath, and was otherwise asymptomatic. On examination, he had decreased air entry bilaterally, but was hyper-resonant over the upper 2/3 of his left lung. Pulse oximetry showed an oxygen saturation of 100% on air and his breathing rate was 12 breaths per minute. The rest of his parameters were stable.

A chest X-ray showed decreased lung markings over the upper half of the left lung (Figure 1). A CT scan of the thorax was performed. This revealed a large emphysematous bulla in the left upper lobe, about 17 centimetres by 21 centimetres in size. Another bulla, measuring 7 centimetres by 6 centimetres, was also visualised in the superior lingular segment (Figure 1).

Figure 1 Left: Chest X-ray done on admission, showing decreased lung markings over the upper half of the left lung

Right: CT scan showing the giant bulla in the left upper lobe, and the smaller bulla in the superior lingular segment.



Accompanying this, there was also a very small pneumothorax of up to 9mm in depth around the left lung, and some para-septal emphysema in the right upper lobe.

In view of his recent pneumothorax, no pulmonary function tests were done.

An elective VATS bullectomy was done, using single lung ventilation under general anaesthesia. Two port technique was used with adhesions removed between the giant bulla and the chest wall. The upper lobe giant bulla was resected using an Endo GIA™ (Covidien, Metronic, Minneapolis, Minnesota, USA) trilinear stapler. The resection was performed a centimetre beneath the origin of the bulla, to ensure that healthy lung seals well at the staple line. The second smaller bulla was resected in the same manner. A total pleurectomy was also performed to help prevent recurrence of the pneumothorax. One drain was placed but no suction was applied onto the drain, unlike routine pleurectomy operations for primary spontaneous pneumothorax. This was omitted to avoid sudden re-expansion of a chronically compressed lung with risk of staple line dehiscence. The patient mobilized the day after the procedure, his drain was removed 2 days post-operatively and he was discharged that same day.

The patient was reviewed two weeks after the surgery and all wounds were healing well (Figure 2). A repeat chest X-ray showed full re-expansion of the lung (Figure 3). The patient also stated that he was feeling quite well and was very happy with the care he had received.

Figure 2 The port through which the VATS bullectomy was carried out, 2 weeks post-surgery

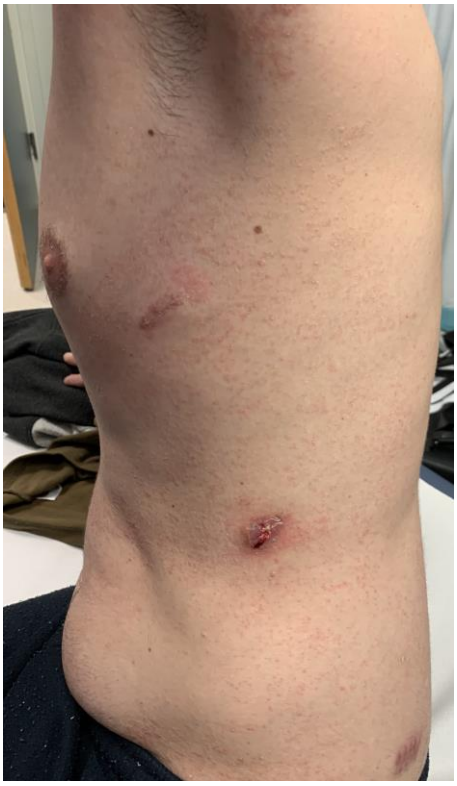
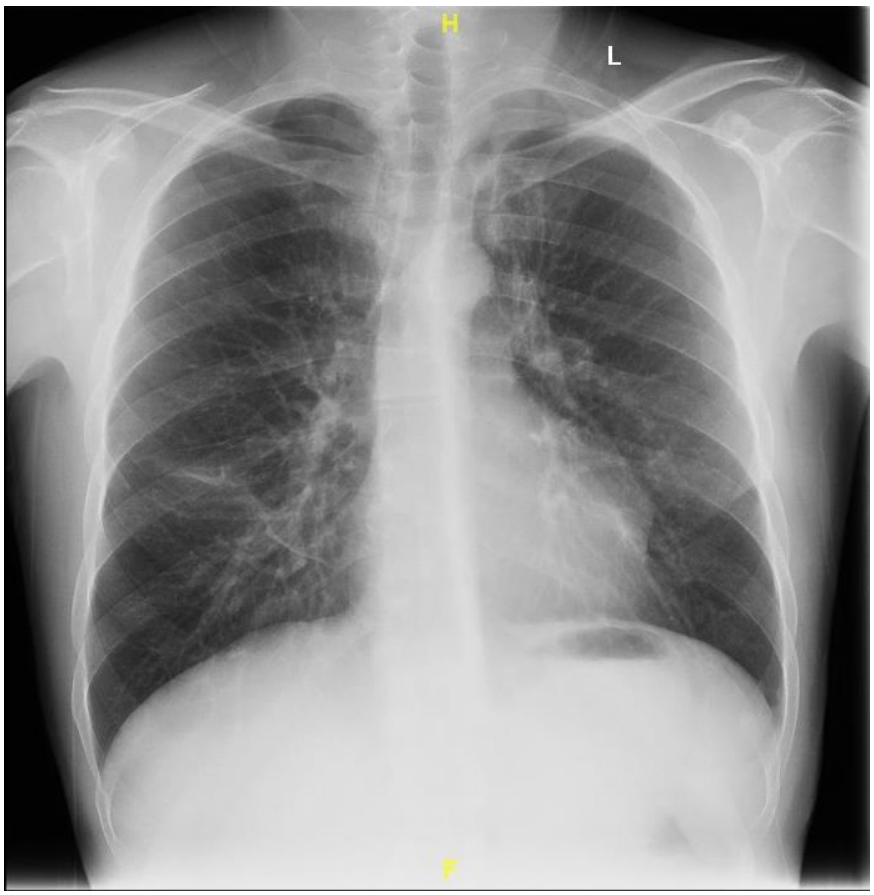


Figure 3 Chest X-ray 2 weeks post-surgery, showing full re-expansion of the lung.



Discussion

Giant bullae are classified in many ways, such as; Reid[8], Wakabayashi[9], and DeVries & Wolfe's[10] practical classification into 4 groups. However, we favour the classification by Klingman which categorizes giant bullae according to the quality of the underlying lung.[11] Type 1 is where the giant bulla is surrounded by normal lung, type 2 by emphysematous lung and multiple other bullae and type 3 is that of vanishing lung syndrome (VLS).

VLS is the most extreme form of giant bullous disease where the bullae, with their low compliance, continue to be ventilated preferentially and the remaining lung involutes into nothing but a hilar bud. No lobes are discernible and unfortunately the condition is not amenable to surgical cure at this stage.

The indications for surgery in giant bullae include dyspnoeic patients with high residual volume and hyperinflation present, and the emergence of complications. Surgery is also indicated as a prophylactic measure in bullae occupying more than 50% of a hemithorax, and also if a longstanding bulla is occupying one third of the hemithorax, since this prolonged compression renders the remaining lung less likely to expand.

The commonest complications of giant bullae include infection, haemorrhage, and pneumothorax.[12-13] A rare complication that has been reported with giant bullous disease is superior vena cava obstruction, which unless treated early could lead to death.[14] Ischaemic stroke caused by air emboli from a ruptured giant bulla has been reported to occur although this is a very rare complication.[15] Interestingly, giant bullae are an emerging risk factor or even aetiological factor for lung cancer. Multiple cases of lung cancer associated with giant bulla and with differing histology have been reported.[16-17] More research about this association is needed, but this could potentially give another valid reason for preventative surgical treatment of giant bullae, especially in older patients, to improve the prognosis.

The case described here was a Klingman type 2 with the complication of recurrent pneumothorax. He had no other potential causes besides cigarette and cannabis smoking.

Marijuana abuse is highly linked with giant bullous disease of the lung. It is suggested that marijuana is more likely to cause giant emphysematous bullae when compared to tobacco smoking, which in contrast has a higher chance of causing emphysematous changes with smaller bullae.[18-20] This could potentially be due to the higher inspiratory pressures and prolonged breath-holding that are associated with marijuana smoking, accompanied by direct toxic damage to the lung parenchyma.[21] Studies indicate that bullous disease secondary to marijuana abuse tends to occur in a younger cohort of patients, and tends to be asymmetrical – as is the case in our patient.[22-23] It also seems to have a specific predilection for the lung apices.[18, 24]

Giant bullae affect gaseous exchange in a variety of ways. First of all, a ventilation-perfusion mismatch is created, since the destruction of alveolar walls creates a bulla which has minimal blood supply. Apart from that, the giant bulla compresses nearby parenchyma which has a better blood supply, therefore limiting ventilation in these airspaces.[4] Secondly, the destruction of the air space walls decreases the elastance hence increasing the compliance of the lung in that

hemithorax. This leads to air entering preferentially into the bulla on ventilation, down the pathway of least resistance resulting in a decrease in the ventilatory capacity of the lung, since gaseous exchange is minimal across the fibrous membrane of the giant bulla.

A study performed in 2005 by Palla et al showed that elective surgery is the treatment of choice for bullectomy. For the cases studied, the mortality rate in the first post-operative year was 7.3%, while the late mortality rate at 5 years post-op was 4.9%.[25] In our case, VATS-bullectomy was utilised, which has been proven to be a safe and effective treatment modality for giant bullae, with long-term symptomatic improvement.[26-27] In fact, VATS bullectomy results in a shorter hospital stay, quicker recovery, less post-operative pain and most importantly better ventilation with reduced chest infection rates, when compared to thoracotomy, and is therefore preferred.[6] Debate continues in the thoracic surgical community on the use of cellulose mesh and fibrin glue to reinforce the suture line[28] or use of BioGlue™ (Cryolife, Kennesaw, Georgia, USA) to reduce staple line dehiscence.[29] The technique described here relies on the presence of healthy lung to staple through at the margin of the bulla. If doubt exists on the strength of tissue one can lay open the bulla and staple the base from within, with the walls of the bulla acting as their own buttress.

Conclusion

There is a misconception that cannabis is less harmful to the lungs than cigarettes. This is not the case as we can see from numerous young people with very severe bullous lung disease. Cigarette smokers do not present so young and with such large bullae. Perhaps the fact that cigarettes are smoked through a filter for large particles plays a part in this difference.

Patients with giant bullae can undergo a novel minimally invasive operation, which carries less risk and a shorter recovery time than the conventional open operation. However, the long-term prognosis of the patient depends on their ability to stop smoking.

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