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Case Report

"Double Fistula": Bronchopleural and Pleurosubcutaneous- A Rare Complication of Closed Pleural Biopsy

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Abstract

Closed pleural biopsy is a vital diagnostic tool and currently recommended in cases of undiagnosed exudative pleural effusion. It is a simple procedure without any complications and seldom associated with complications like pneumothorax, intra pleural haemorrhage and secondary infections. An unreported complication till date is described here in which a double fistula consisting of bronchopleural and pleurosubcutaneous fistulas developed in a patient following needle biopsy of parietal pleura. Being an extremely rare complication, it should not serve as a contraindication for pleural biopsy in cases where it is indicated.

Keywords: Bronchopleural; Pleurosubcutaneous; Complication; Pleural biopsy

Introduction

Closed-needle biopsy of pleura is commonly being used since its introduction several decades ago, to arrive at aetiological diagnosis in cases of undiagnosed exudative pleural effusion with non diagnostic cytology and adenosine deaminase levels. Pleural biopsies are of utmost value especially in the suspicion of granulomatous and malignant diseases of the pleura [1]. Presently flexible thoracoscopy using local anaesthesia is a preferred technique to obtain pleural tissue but needs the sophisticated instruments and expertise with associated risks of greater invasiveness. Alternately percutaneous pleural biopsy under image guidance is recommended but blind procedure is almost equally safe and effective. This is of particular importance in a developing country like India where the facilities of thoracoscopy and imaging guided cutting needle biopsies are not easily available. In such circumstances needle biopsies are performed by closed technique without image guidance using reverse bevel needle, such as Abram's or Cope needle in which a small piece of the parietal pleura is obtained for histopathological or microbiologic evaluation [2].

Percutaneous pleural biopsy is a simple procedure with minimal complications. Commonly reported complications include site pain, vasovagal reaction, transient fever, secondary infection, site seeding of cancer cells, site hematoma, hemothorax, pneumothorax and accidental breaking of biopsy needle [1,3]. The pleural biopsy needle can also be mistakenly inserted into the liver, spleen, or kidney during the procedure causing trauma to these organs [4].

A unusual case report having pneumothorax, pneumomediastinum and subcutaneous emphysema following closed percutaneous pleural biopsy has been described [5], but to the best of our knowledge, development of persistent double fistula in the form of Bronchopleural (BPF) and Pleurosubcutaneous (PSF) fistulas following needle biopsy of parietal pleura, presenting later on as swelling with coughing, has not been reported previously in known English literature. We are presenting one such case of broncho-pleuro-subcutaneous fistula caused by closed pleural biopsy.

Case Report

A 45 years old male presented to outpatient department with complaints of pain and swelling on the postero-lateral chest wall on the right side and an unusual crackling or bubbly sensation surrounding this swelling for two months. This swelling and unusual crackling sensation appears only while coughing or straining at stool. There was no history of trauma but the patient underwent closed pleural biopsy for right sided pleural effusion six months back and was also complaining of dull aching chest pain and breathlessness since then. Physical examination revealed a linear scar mark of 1 cm on the right infrascapular area, suggestive of pleural biopsy mark. On

Figure 1: Scar mark and swelling appeared with Valsalva manoeuvre on the postero-lateral chest wall on the right side.

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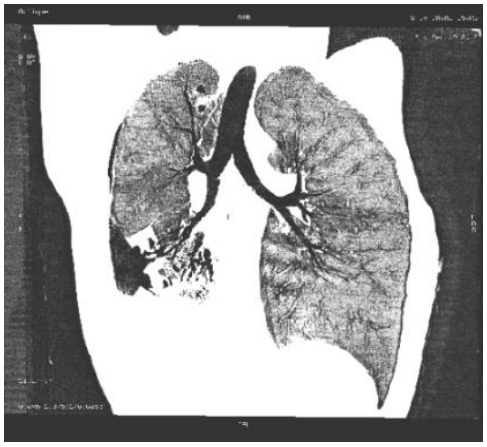

Figure 2: Coronal reformatted MinIP image in lung window is showing brocho-pleural fistula associated with middle lobe bronchus.

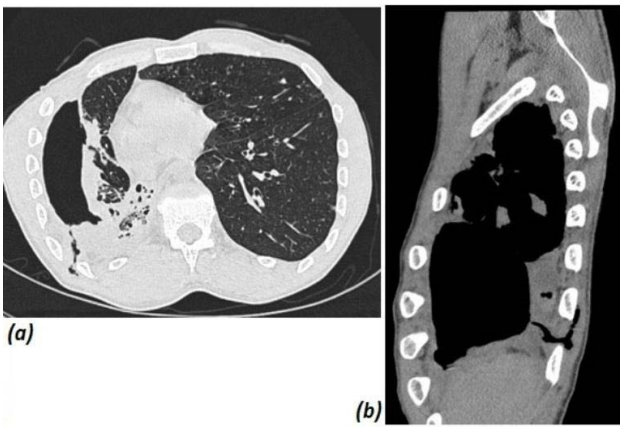

Figure 3: (a) Curved replanar axial image in lung window and (b) parasagittal reformatted image in mediastinal window showing radiolucent tract extending from right sided pneumothorax to subcutaneous plane.

coughing and on performing Valsalva maneuver (imitating strain) an ill defined swelling of 3 × 3 cm size appeared at right infrascapular area overlying the eighth and ninth inter-costal spaces; which was smooth, non-tender, soft, fluctuant and increased in size with forceful coughing. The signs of inflammation, such as redness and increased local temperature, were absent. Localized palpable crepitations were also found over and around swelling, so a suspicion of double fistula consisting of bronchopleural and pleurosubcutaneous was made (Figure 1). All routine blood investigations were normal. Chest X- ray PA view showed loculated chronic pneumothorax and fibrocalcified lesions on right side. High Resolution Computerized Tomography (HRCT) scans of the chest showed hydropneumothorax with significant pleural thickening, lung volume loss with patchy consolidatory changes, fibrobronchiectatic changes on right side. Coronal reformatted MinIP image showed brochopleural fistula associated with middle lobe bronchus (Figure 2) while curved replanar axial and parasagittal reformatted image showed radiolucent tract extending from right sided pneumothorax to subcutaneous plane (Figure 3a and 3b).

Patient advised for bronchoscopic glue insertion for closure of BPF, but he refused and managed conservatively with inter costal tube drainage for pneumothorax and to release pressure from BPF on pleurosubcutaneous tract to enhance healing. The tube was kept for 15 days leading to closure of both the fistulas following which it was removed. Patients improved symptomatically with resolution of major symptoms and discharged with anti TB medication.

Discussion

Apart from well known complications of pleural biopsy, many new complications are emerging like one in our patient. Our patient developed broncho-pleuro-subcutaneous fistula following percutaneous needle biopsy of pleura. Bronchopleural Fistula (BPF) is an unnatural communication between the bronchial tree and pleural space as evidenced by continued air leak post-pneumothorax. The most common cause of BPF is post pulmonary resection followed by other causes including pleuroparenchymal fibroelastosis, post chemo or radiotherapy, necrotic lung infection, persistent spontaneous pneumothorax, tuberculosis, iatrogenic and a complication of mechanical ventilation [6]. Diagnosis of BPF is established by clinical evaluation, Contrast Enhanced Computed Tomography (CECT) thorax and radio aerosol scanning (eg; xenon ventilation nuclear scintigraphy) [7]. Treatment of persistent BPF primarily includes bronchoscopic insertion of glues, coils, sealants and rarely surgery [6].

Pleurosubcutaneous Fistula (PSF) is a very rare pathological entity consisting communication between the pleura and subcutaneous tissue and only two cases following high dose radiotherapy for breast cancer has been reported in known literature. Diagnosis of PSF can be made by transcutaneous USG, color doppler, HRCT thorax and MRI, which shows the existence of a fistulous tract between the pleural cavity and the subcutaneous tissue [8].

Probably these fistulas in our patient were complications of closed pleural biopsy procedure as it is performed through percutaneous route after giving a small incision of approx 1cm and introducing Abrams' pleural biopsy needle through it. During this blind procedure, the visceral pleura and lung parenchyma incised inadvertently, leaving a small bronchopleural fistula that lead to development of pneumothorax [4]. Symptoms of this pneumothorax were probably masked due to concurrent pleural effusion. Further closure of skin incision by scar formation and impaired healing of pleuro cutaneous tract due to unknown factors lead to development of pleurosubcutaneous fistula that appeared later on while coughing or straining at stool.

Recently Aiyappan et al [9] described a case of 'double fistula' consisting different fistulas (broncho-pleuro-cutaneous fistula) due to a different etiology. They described a case of broncho-pleuro-cutaneous fistula which developed following tube thoracostomy.

However, the possibility of developing such fistulas following pleural biopsy should never serve as a contraindication to this routine procedure of high importance where facilities of thoracoscopy and image guidance are not available. All the chest physician and cardiothoracic surgeon should be aware of this rare complication, so that early diagnosis can be made and managed accordingly. Diagnosis can be suspected in patients where history of pleural biopsy or similar procedure like tube thoracostomy is there, and presenting with swelling appearing or increasing with coughing.

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