A case report of endobronchial semi-invasive aspergillosis#

Figen KADAKAL[1], M. Atilla ÜYSAL[1], Mehmet Akif ÖZGÜL[1], Senem ELİBOL[1], Nur ÜRER[2], Atilla GÜRSES[1], Veysel YILMAZ[1]

[1] Yedikule Göğüs Hastalıkları ve Göğüs Cerrahisi Eğitim ve Araştırma Hastanesi,
[2] Yedikule Göğüs Hastalıkları ve Göğüs Cerrahisi Eğitim ve Araştırma Hastanesi, Patoloji Bölümü, İstanbul.

ÖZET

Bir endobronşiyal semi-invaziv aspergillozis olgusu

Pulmoner aspergillozis üç ana başlık altında sınıflandırılabilir. Bunlar; invaziv pulmoner aspergillozis, allerjik bronkopulmoner aspergillozis ve aspergillomadır. Bazen hastalığın birden fazla formu aynı anda bulunabilir. Akciğer dokusunda lokal invazyon oluşturan semi-invaziv aspergillozis aspergillomadan farklıdır. Biz akciğer grafisinde soliter pulmoner nodül olan 42 yaşında sağlıklı bir bayan hastada endobronşiyal semi-invaziv aspergillozis tanısı koyduk ve literatür bilgisi ışığında sunmayı amaçladık.

Anahtar Kelimeler: *Semi-invaziv aspergillozis, endobronşiyal, aspergilloma.*

[#] 17. Göğüs Hastalıkları Asya Pasifik Kongresi (29 Ağustos-1 Eylül 2003, İstanbul)'nde poster olarak sunulmuştur.

SUMMARY

A case report of endobronchial semi-invasive aspergillosis#

Kadakal F, Uysal MA, Ozgul MA, Elibol S, Urer N, Gürses A, Yilmaz V

Yedikule Training and Research Hospital for Chest Disease and Thoracic Surgery, Istanbul, Turkey.

Pulmonary aspergillosis may be classified under three main categories. These are invasive pulmonary aspergillosis, allergic bronchopulmonary aspergillosis and aspergilloma. Sometimes more than one form of the disease may be present at the same time. Semi-invasive aspergillosis is different from aspergilloma in that there is local invasion of the lung tissue. We have observed a previously healthy 42 year old female with a solitary pulmonary nodule on her radiograms. A diagnosis of endobronchial semi-invasive aspergillosis was established in this patient. We aimed to present this case report with a review of the literature.

Key Words: *Semi- invasive aspergillosis, endobronchial, aspergilloma.*

[#] It was presented as a poster presentation in 17th Asia Pacific Congress on Diseases of the Chest, August 29-September 1, 2003 Istanbul - TURKEY

Yazışma Adresi (Address for Correspondence):

Dr. M. Atilla ÜYSAL, Yedikule Göğüs Hastalıkları ve Göğüs Cerrahisi Eğitim ve Araştırma Hastanesi, İSTANBUL - TURKEY
e-mail: dratilla@yahoo.com, dratilla@netone.com.tr

The pathological definition of airway invasive aspergillosis is the presence of aspergillus organisms deep to the basement membrane of the broncial tree. This form of aspergillosis may either be present as acute airway invasive aspergillosis or chronic airway invasive aspergillosis [also known as Semi-Invasive Aspergillosis (SIA) or chronic necrotising aspergillosis] (1,2). SIA was first described in two reports in 1981 and 1982 (3,4). Tamaki and Kim presented endobronchial aspergilloma in 1980 and 2000, respectively (5,6). We have observed a previously healthy 42 year old female complaining of blood tinged sputum with a solitary pulmonary nodule on her radiograms. A diagnosis of endobronchial SIA was established in this patient. We aimed to present this case report with a review of the literature.

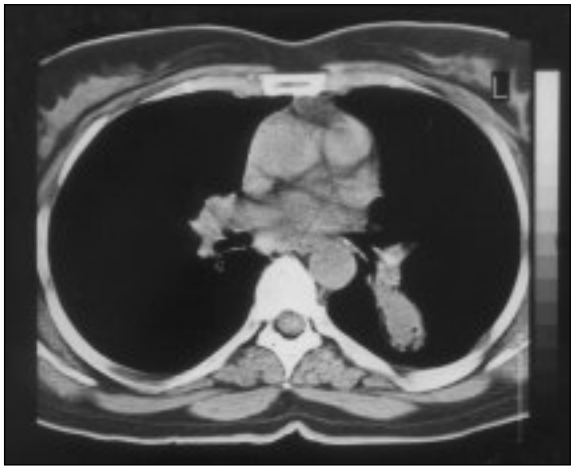
CASE REPORT

A 42 year-old female had been hospitalized in our clinic. She had been suffering from cough and intermittent blood-tingled sputum for six years. There was no history of other illnesses. Her physical findings and routine laboratory examinations were normal. Sputum cultures for bacteria and fungi were negative. Tuberculine skin test was negative. The chest X-ray showed an irregularly defined nodular lesion that was 2 x 3 cm on left perihilar area surrounded by satellite nodules (Picture 1).

A chest computed tomography (CT) scan showed a nodule that was relatively well-defined measuring 2 x 3 x 2 cm. Bullous changes were observed at the superior segment of the left lower lobe around the lesion (Picture 2,3).

Fiberoptic bronchoscopy showed mucosal irregularity and hyperemia at the subsegment of the superior segment of the left lower lobe. Bronchoscopic forceps biopsy were taken from the lesion. On microscopic examination of the biopsy specimens granuloma formation and aspergillus hyphea were observed. Massive hemoptysis developed two days following bronchoscopy and the patient consequently underwent emergency thoracotomy and left lower lobectomy.

Picture 1. The chest X-ray showed an irregularly defined nodular lesion that was 2 x 3 cm on left perihilar area surrounded by satellite nodules.

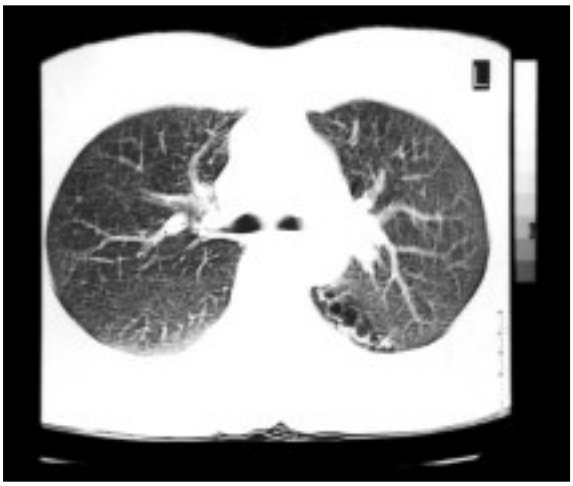


Picture 2. Bullous changes were observed at the superior segment of the left lower lobe around the lesion.

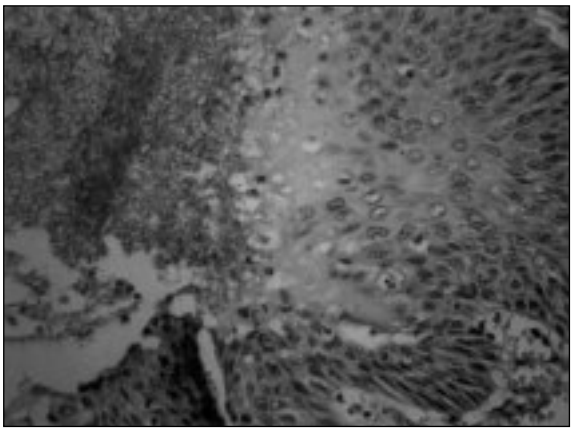
On gross pathologic examination the lesion involved the bronchial wall and was located in the subpleural area. Microscopic examination revealed spores of fungi and hyphae invading the squamous metaplasic cells on the inner wall of the cavity. In addition, there were fungus plugs in the bronchial lumen associated with ulcerative changes on the bronchial mucosa. The histopathologic findings supported the diagnosis of SIA in the lung (Picture 4).

DISCUSSION

Invasive aspergillosis is often used in the context of aggresive disease in immuno compromised



Picture 3. Bullous changes were observed at the superior segment of the left lower lobe around the lesion.



Picture 4. The histopathologic findings supported the diagnosis of seminvasive aspergillosis in the lung.

patients (1). The hyphae invade the adjacent lung parenchyma starting from the bronchial wall and subsequently the accompanying arterioles.

The pathological definition of airway invasive aspergillosis is the presence of aspergillus organisms deep to basement membrane of the bronchial tree which may develop acutely or chronically (also known as SIA or chronic necrotising aspergillosis) (1,2).

SIA is usually seen in middle-aged and elderly patients and it has also been described in patients with mild immunosuppresion. Our case was not immunosupressed and had no atopy. The CT scan showed a nodule surrounded by bullous changes at the superior segment of the left lower lobe. The original reports suggested that the growing mycetoma produced its own bronchiectatic cavity (2). In our case, we do not know whether bullous changes are the result of the mycetoma or are traces of previous small cavities.

The diagnosis of SIA is confirmed by histologic demonstration of tissue invasion by the fungus and the growth of aspergillus species on cultures. While the yield of transbronchial biopsy specimens or percutaneous aspirates is relatively poor, thoracoscopic or open lung biopsy are rarely performed in these patients (4). The diagnosis in our case was confirmed by histologic demonstration of hyphae in the biopsy specimen taken from bronchial biopsy.

Surgical resection is generally reserved for patients who are not tolerating antifungal therapy and patients with residual localised but active disease despite adequate antifungal therapy. Kim and coworkers has reported a case in whom SIA was treated by endobronchial forceps resection (6). Our patient had undergone emergency lung resection due to massive hemoptysis.

Pathologic findings have been infrequently described. There is a combinaton of necrosis, fibrosis and granulomatous inflammation with coagulative necrosis and vascular invasion by fungal hyphea. A less common manifestation is the presence of a large ectatic airway containing abundant fungal hyphea resembling a fungus ball. In contrast to the latter, focal necrosis of the airway wall is associated with invasive hyphea (7). Recently, a SIA in an immunocompotent host was presented by Ciledag et al. In this case, diagnosis was established by transthoracic fine-needle aspiration. Specimen showed tissue invasion by fungal hyphae and spores. Gram staining and culture of bronchial bronchoalveoler lavage yielded fungal hyphae and *Aspergillus fumigatus* respectively (8). Pathologic examination of the surgical specimen in our patient showed fungus spores and hyphea invading the squamous metaplasic cells in the inner wall of the cavity and bronchiolitis around the cavity leading to a diagnosis of endobronchial SIA.

It could be speculated that there was a previously localized cavity in the superior segment of the lower lobe. An aspergilloma colonized in the form of fungus ball within this cavity and after a while the microorganisms further spreaded to the bronchial wall resulting in SIA. Another scenario would be that fungus hyphea might produce its own bronchiectatic cavity (3).

In conclusion, SIA may be seen as a pulmonary nodule on the roentgenogram and it may be seen endobronchially. Endoscopic evaluation and biopsy are in valuables in the work up of patients with hemoptysis or lung nodules. Semi-invasive endobronchial aspergillosis should also be considered in the differential diagnosis of such cases including immuncompotent patients.

REFERENCES

1. *Buckingham SJ, Hansell DM. Aspergillus in the lung: Diverse and coincident forms. Eur Radiology 2003; 13: 1786-800.*
2. *Soubani AO, Chandrasekar PH. The clinical spectrum of pulmonary aspergillosis. Chest 2002; 121: 1988-99.*
3. *Gefter WB, Weingrad TR, Epstein DM, et al. "Semi-invasive" pulmonary aspergillosis: A new look at the spectrum of Aspergillus infections of the lung. Radiology 1981; 140: 313-21.*
4. *Binder RE, Faling LJ, Pugatch RD, et al. Chronic necrotising pulmonary aspergillosis: A discrete clinical entity. Medicine 1982; 61: 109-24.*
5. *Tamaki S, Danbara T, Natori H, et al. A resected case of endobronchial aspergilloma due to Aspergillus restrictus. Jpn J Thorac Dis 1980; 18: 464-9.*
6. *Kim JS, Rhee Y, Kang SM, et al. A case of endobronchial aspergilloma. Yonsei Medical Journal 2000; 41: 422-5.*
7. *Fraser RS, Müller NL, Colman N, Pare P. Fungi and actinomyces. In: Fraser RG, Pare AP (eds). Diagnosis of Disease of the Chest. Philadelphia: WB Saunders Company, 1999: 875-958.*
8. *Ciledag A, Celik G, Kumbasar Ozdemir O, et al. Semi-invasive aspergillosis in an immunocompetent host. Tüberküloz ve Toraks 2002; 50: 379-83.*