Comment

Localized mediastinal amyloid deposition is unusual [3, 4], whereas mediastinal lymphadenopathy caused by systemic amyloidosis has been reported frequently and requires histologic conformation because radiologic findings are typically nonspecific [1, 5–7]. Takamori and colleagues [3] reported a 33-year-old woman with mediastinal thymic amyloidosis (diameter, 8.5 cm), with cystic changes and calcification [8]. She had a history of rheumatoid arthritis. The resected specimen showed amyloid A protein–derived amyloid, which usually causes secondary systemic amyloidosis. Takeshita and colleagues [4] reported a 71-year-old man with enlargement of a paraaortic lymph node (diameter, 4 cm). Immunoglobulin λ–chain was identified, which confirmed a diagnosis of localized AL amyloidosis [4].

Amyloid deposition simulates both inflammatory and neoplastic conditions [2]. Amorphous or irregular calcifications are occasionally seen within amyloid deposits [2]. Amyloid-infiltrated nodes frequently show punctate [2, 5, 6] or dense foci of calcification [7] and are often associated with parenchymal lung disease or pleural effusion [6]. The differential diagnosis includes metastatic lymph nodes, lymphoma, mediastinal tumor, tuberculosis, sarcoidosis, Castleman disease, silicosis, and other granulomatous disease [5, 6]. Because computed tomography findings are nonspecific, definitive diagnosis requires pathologic confirmation by surgical excision or biopsy under mediastinoscopy or transbronchial needle aspiration [5]. Our patient had an enlarged, irregularly shaped nodule with peripheral punctate calcification, without intrathoracic involvement. Despite the fact that the radiologic findings were not typical of thymic neoplasm, surgical confirmation was decided upon because the gradual enlargement of the tumor and accompanying diplopia suggested myasthenia gravis.

Positive histology results for amyloid must be followed by immunohistochemical analysis to determine fibril type and need for treatment [1]. Classification of amyloidosis is based on the different subunit amyloid proteins, which identify organ involvement and disease manifestation. AL amyloidosis is the most heterogeneous form of the disease and can be systemic or localized. The amyloidogenic light chains in localized AL amyloidosis are produced by subtle cloning of lymphoplasmacytes, which are localized near amyloid deposits. Hence, local control is usually sufficient for localized amyloidosis [1]. In contrast, the fibrils in systemic AL amyloidosis are derived from circulating monoclonal light chains produced by clonal plasma cells, which can be benign or malignant [1]. Thus, differential diagnosis must consider whether AL amyloidosis is localized or systemic. In the present case, although bone marrow examination was not performed, there was no other organ dysfunction or symptoms, including monoclonal γ–globulinemia or Bence-Jones proteinuria. Localized AL amyloidosis of the thymus was diagnosed in our patient, and follow-up without treatment was chosen.

Our patient complained of transient diplopia, which immediately and completely resolved after thymectomy. The resected specimens had amyloid deposition in the thymic tissue, without thymoma or thymic hypertrophy. Ocular palsy is frequently observed and often difficult to diagnose. One study found that 25% of cases of ocular palsy are undiagnosed [8]. Castelluccia and colleagues reported that among 31 patients with isolated diplopia, 10 were undiagnosed, 10 had myasthenia gravis, and the remaining patients had encephalopathy with multiple infarcts, multiple sclerosis, infectious neuritis, mitochondrial myopathy, extracranial muscle myositis, and cerebral tumor [8]. Although there are a number of possible causes for the diplopia in our patient (including self-limiting infectious neuritis as an incidental complication), clinical follow-up for myasthenic symptoms is ongoing.

References


Bulky Mediastinal Aspergillosis Mimicking Cancer in an Immunocompetent Patient

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We describe the case of a previously healthy 42-year-old woman who presented with a chronic cough and...
occasional night sweats. Radiologic exploration showed a bulky mediastinal mass surrounding the aortic arch, associated with a left subclavicular lymph node and a cerebral round lesion, mimicking a disseminated lung cancer. Surgical left subclavicular and computed tomography–guided mediastinal biopsy specimens showed granulomatous patterns. Mycologic culture of both samples grew *Aspergillus flavus*. Resolution was obtained after 9 months of oral voriconazole therapy.


**Mediastinal masses are mainly due to mediastinal involvement of a lung cancer, lymphoma, or tuberculosis in endemic countries. Mediastinal masses resulting from invasive aspergillosis (IA) are extremely rare and occur mainly in severely immunosuppressed patients. We report a case of disseminated aspergillosis in an immunocompetent patient, resulting from *Aspergillus flavus* species, with bulky mediastinal involvement mimicking advanced lung cancer.**

In January 2012, a 42-year-old Algerian woman was referred to our thoracic unit (Institut Mutualiste Mon tsouris, Paris, France) for investigation of large mediastinal and left hilar masses. The patient had always lived in Algeria. She had never smoked and did not take any medications. Since February 2011, she had experienced a cough and low-grade fever. Computed tomography (CT) of the thorax showed large mediastinal and left hilar masses. Microbiologic cultures of her bronchial aspirates were negative for mycobacteria. Granulomatous patterns were found on the bronchial biopsy specimens. She was given antituberculous therapy for 6 months, but without improvement. Because her symptoms and the lesions as shown on CT did not resolve, she came to France for further investigation.

On admission, she presented with a cough and occasional night sweats, but no fever. Her body weight had remained stable during 2011. The results of physical examination were unremarkable except for a palpable 2.5-cm left subclavicular lymph node. Her white blood cell count, hemoglobin, C-reactive protein, liver enzyme, and serum immunoglobulin levels were within normal values. The results of serologic testing for human immunodeficiency virus were negative.

Computed tomography of the thorax showed a mediastinal mass surrounding the left pulmonary artery and the descending aorta, associated with compression of the left main bronchus (Fig 1). Cerebral CT scan revealed a 1-cm solitary round lesion in the right frontal lobe, which was enhanced by contrast perfusion and was surrounded by minimal edema (Fig 2). Bronchoscopy revealed progressive narrowing of the left main bronchus with enlargement of the left main division. Analysis of the bronchial biopsy specimens showed nonspecific inflammatory patterns. Cultures of bronchial aspirations were sterile. Surgical biopsy specimens from the left subclavicular node and the CT-guided transcutaneous mediastinal mass revealed nonnecrotizing granulomas, with epithelioid macrophagic giant cells. Extensive fibrosis was observed in the mediastinal sample. Grocott-Gomori methenamine silver staining revealed fungal hyphae (Fig 3). Mycology cultures grew the *Aspergillus flavus* complex. Antifungal susceptibility tests showed minimal inhibitory concentrations of 0.19, 0.064, 0.004, and 1.5 mg/mL for voriconazole, posaconazole, caspofungin, and amphotericin B, respectively.

Anti-*Aspergillus* antibodies were positive (69 UA/mL; n < 5 UA/mL; EIA, Bio-Rad; Marne la Coquette, France) within the serum, with no specific metabolic or somatic precipitins (immunoelectrophoresis [IEP], Bio-Rad).

In February 2012, voriconazole therapy (200 mg twice a day) was started, and the patient returned to Algeria. In June 2012, a CT scan showed that the mediastino hilar mass had decreased (Fig 2). Voriconazole was discontinued in December 2012. In November 2013, a CT scan showed resolution of the mediastinal mass but persistence of a slight thickening of the left main bronchus wall. By 11 months later, the patient was still asymptomatic.

**Comment**

Mediastinal masses due to IA are extremely rare, especially in nonimmunosuppressed patients. The presence of

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**Fig 1.** Contrast-enhanced computed tomographic (CT) scan of the thorax before treatment. (A) Mediastinal mass surrounding the left pulmonary artery and the descending aorta, associated with compression of the left main bronchus. (B) CT scan of the thorax 11 months after completion of treatment, showing resolution of the mediastinal mass but persistence of slight thickening of the left main bronchus wall.
subclavicular lymph nodes and a contrast-enhanced brain lesion first suggested disseminated lung cancer.

Aspergillus flavus is a ubiquitous saprophytic soil organism [1]. In developing countries, A. flavus is the most frequent Aspergillus species isolated from the environment and is associated with sinusitis and with skin and brain infections [1, 2]. Invasive aspergillosis occurs mainly in severely immunosuppressed patients. In rare cases, IA develops in patients with other pathologic conditions, such as bacterial or viral pneumonia, chronic obstructive pulmonary disease, sepsis, liver failure, alcoholism, or sinusitis, or after operation [3–5]. Our patient did not have any predisposing conditions.

Two cases of A. flavus infection have been published [6, 7]. Both were in young adults and involved the vena cava. The first patient died despite treatment with amphotericin B [6], and the second presented with other sites of septic emboli but recovered after treatment with voriconazole and caspofungin [7]. A fatal case of mediastinal IA caused by A. fumigatus, with extension to the pulmonary vasculature, has also been described in an immunocompetent patient [8]. In our patient, the primary site of infection was probably broncho-pulmonary, consequent on to aerosol exposure, followed by hematogenic dissemination to the brain and subclavicular lymph nodes.

The guidelines recommend voriconazole as the first-line therapy for IA, but the optimal duration of therapy for chronic infections is not known [9]. Treatment is usually given intravenously for 7 days before a switch to oral use, accompanied by careful therapeutic monitoring. However, because our patient was returning to Algeria, voriconazole was started at 200 mg twice daily by mouth, and serologic concentrations were not monitored. After 4 months of therapy, the cerebral lesion had disappeared, whereas the mediastinal mass took longer to disappear. After 9 months of treatment, we decided to stop voriconazole despite bronchial thickening on the CT scan. By 11 months later, the IA had not recurred, suggesting that treatment could be safely discontinued while minimal lesions were still visible on CT scans.
Successful Repair of Benign Left Atriogastric Fistula After Transhiatal Esophagectomy

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Benign gastrocardiac fistula is a rare complication following esophagectomy that can occur when the gastric tube is placed in the retrosternal space. We describe a benign fistula between the left atrium and the gastric conduit following transhiatal esophagectomy. The surgical technique that was used for successfully repair is also discussed.


Benign gastroatrial fistula is a rare complication following esophagectomy, with only about six cases reported in the literature [1–5]. It appears to be more common after retrosternal gastric tube reconstruction. Ischemia and gastric acid stasis in the dependent portion of the conduit are speculated to lead to the fistulization with the right ventricle that lies immediately posterior. In case of transhiatal esophagectomy, the conduit is in the posterior mediastinum and is less likely to fistulize to the heart. Nevertheless, the lesser curvature staple line lies directly posterior to the left atrium. The case here demonstrates that such fistulae can occur even after transhiatal esophagectomy, and prompt surgical repair is required; otherwise, they are fatal.

A 56-year old male patient underwent neoadjuvant chemoradiation followed by transhiatal esophagectomy 3.5 years ago for T3N1M0 distal adenocarcinoma. Of note, the lesser curvature staple line was oversewn using a 3-0 silk suture as part of the procedure. An esophagram on day 6 was normal. The patient was discharged on a full liquid diet and jejunal tube feeding for 4 weeks. Four-week follow-up was unremarkable, and his diet was advanced. Complete pathologic response was noted to the neoadjuvant chemoradiation, with no residual tumor in the specimen.

Surveillance CT at 1 year demonstrated four liver metastases. He received chemotherapy for 6 months. A repeated CT scan revealed regression of the liver lesions. He underwent radiofrequency ablation of the liver metastases. Chemotherapy was continued, and surveillance PET-CT scan 1 year later showed no uptake in the liver lesions and no other metastasis.

Three and a half years after the initial esophagectomy, the patient was admitted to the hospital with fever and streptococcal bacteremia. The next day, he developed massive hematemesis that prompted an urgent upper endoscopy. A large amount of blood was noted in the conduit along with an ulcer with adherent clot at the base. The ulcer was present along the lesser curvature staple line. It was recognized that the right gastric artery was ligated at the time of initial surgery and no major vascular pedicle should be present along the lesser curvature, raising the suspicion of a cardioaortic fistula as the cause of the bleeding. A CT scan confirmed the presence of left atriocutaneous fistula along the lesser curvature staple line (Fig 1).

The patient underwent a median sternotomy and repair of the fistula on cardiopulmonary bypass. After the sternotomy, a pericardial flap was created reflecting the anterior pericardium and the fat pad. Cardiopulmonary bypass was initiated, and cardiac arrest was achieved through antegrade cardioplegia. This was done with minimal manipulation to prevent exsanguination into the conduit or air embolism to the left atrium. The Sondergard’s groove was dissected, and left atrium was opened. A 6-mm ulcer was found in the posterior midline of the left atrium just inferior to the right inferior pulmonary vein (Fig 2A). The left atrium was thoroughly irrigated, and the ulcer was closed in two layers using 3-0 Prolene suture. Fairly generous bites of the endothelium and myocardium were taken to achieve good endothelial approximation. Subsequently, the left atrium was closed in standard fashion, and a left ventricular vent was left to facilitate closure of the gastric conduit ulcer with heart beating, but empty.

Aortic cross clamp was removed after placing a root vent, and the heart was retracted superiorly (Fig 2B), exposing the gastric conduit. The posterior pericardium

References