

INVASIVE ASPERGILLOSIS OF THE NECK AND UPPER MEDIASTINUM WITH INTRASPINAL EXTENSION: AN UNUSUAL PRESENTATION

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Aspergillosis refers to a variety of diseases caused by several species of *Aspergillus* organisms, which are abundant in the environment. The term "invasive aspergillosis" is generally used to imply a histopathologically demonstrated invasion of tissues by these spores,¹ and it represents an important cause of morbidity and mortality, particularly among immunocompromised patients. Our case illustrated invasive aspergillosis in a healthy patient following a traumatic wound, which presumably resulted in the direct implantation of the spores into the soft tissues. It also demonstrated the long asymptomatic period before clinical manifestation of the disease.

Case Report

A 34-year-old female patient was referred to our hospital with a mass on the left side of the neck, which had been gradually increasing in size over the previous eight months. The suspicion of malignancy was considered, with a high possibility of being a lymphoma. Two months prior to the presentation, she experienced left upper limb pain and weakness, as well as numbness in the left 4th and 5th fingers, mild dysphagia, and hoarseness of voice. On direct questioning, the patient gave a history of having had a penetrating injury at the left side of the neck by a palm tree branch 18 months previously.

Clinical examination showed a fixed mass occupying the left lower side of the neck, and left ptosis without palpable local lymphadenopathy or raised temperature. Neurological examinations showed left ptosis, nerve roots and/or brachial plexus lesion. Laryngopharyngoscopy showed paralysis of the left vocal cord. The remaining examination was normal and the laboratory findings were non-contributory. Chest x-ray showed a soft tissue opacity on the left side of the neck extending to the superior

mediastinum, displacing the trachea towards the right, and compressing the left lung apex. Ultrasound examination confirmed the plain film findings and revealed that the solid mass was encasing the neck vessels. CT scan showed a heterogeneous soft tissue mass with no calcification and only slight heterogeneous enhancement after intravenous contrast injection. The mass was seen to be arising mainly from the left scalenus muscle group, extending from almost the laryngeal level down to the superior mediastinum. The internal jugular vein was compressed with an intraluminal thrombus proximal to the mass (Figure 1). The common carotid artery was engulfed and displaced by the mass, as well as the trachea and esophagus. The thyroid, which was clearly differentiated from the mass, was displaced anteriorly.

Enlargement of the neural foramina at the C_{6/7} and C₇/T₁ levels on the left side was evident, with the mass extending through the foramina into the epidural space and displacing the cord towards the right (Figure 2). In the superior mediastinum, the mass also engulfed the supra-aortic vessels (Figure 3). Our initial diagnosis was a soft tissue malignancy, possibly lymphoma, neurogenic tumor, or malignant thymic mass. Teratoma was also considered.

An open surgical biopsy which was performed using a low cervical approach, showed a very indurated gritty mass, with a hard consistency. It was attached to the posterior structures of the neck and was not dissectible from the carotid sheath. The histopathology report indicated necrotising and caseating granulomata in a background of fibrosis and chronic inflammatory infiltrate, confirming its long duration. This picture was highly suggestive of fungal infection, which was confirmed by the positive PAS stain for fungal structures (Figure 4), most likely mucormycosis, and culture was advised. This showed a light mixed growth of *Aspergillus niger* and *Aspergillus fumigatus*. Screening for HIV and TB was negative. Immunoglobulin studies were normal without any sign of neutropenia.

As a result of the histological findings, an examination of the paranasal sinuses was done, as well as reviewing the chest x-ray and CT scan findings to determine whether there was a primary site for the neck infection. These were negative. A diagnosis of invasive aspergillosis was then made. Treatment was started with IV amphoterecin-B (total

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FIGURE 1. CT scan with IV contrast at C₇ level. Homogenous mass extending to the left neural foramina posteriorly, pushing the thyroid to the right side. Note the internal jugular vein thrombosis (arrow).

FIGURE 2. CT scan with IV contrast at T₁ level. Clear widening of the left neural foramina with epidural extension and cord displacement (one arrow). Note the engulfment of the left common carotid and vertebral arteries (two arrows).

FIGURE 3. CT scan with IV contrast at the upper mediastinal level. Extension of the mass to the upper mediastinum, engulfing the supra-aortic vessels (arrow) with tracheal displacement.

FIGURE 4. Microscopic view of PAS stain. Fungal hyphae identified (arrow) in multinucleated giant cells.

dose was 1.0 gm for the first 2 weeks of treatment, and was started with a test dose), followed by oral Intraconazole (400 mg daily for eight months). Nine months post-treatment, the patient showed a clear improvement in the weakness of her left upper limb, while radiologically there was almost complete regression of the soft tissue mass in the cervical and upper mediastinal areas, with no residual epidural extension.

Discussion

Invasive aspergillosis varies in severity and clinical course, depending upon the affected organ and the host. It is caused by many *Aspergilli* groups, however, the most common is the *Aspergillus fumigatus* group.² These groups live in soil, and derive nutrients from dead plants and animal matter.³

Invasive aspergillosis is a result of three factors¹: 1) suppression of immune response due to debilitating disease or therapy^{1,3}; 2) glucocorticoid therapy with resultant diminished inflammatory response and the disruption of the normal flora by anti-microbial agents⁵; and 3) local implantation of the fungus.^{1,4} The disease is well known in immunocompromised individuals,^{1,3} but has also been described in healthy individuals.⁶ Our patient proved to be healthy, from the normal hematological studies conducted, and she was not immunocompromised (normal humeral and T cell immunity without neutropenia). The vegetational

environment of the patient can be considered as a high-risk factor and a possible source of the infection following the traumatic wound.⁴

The usual pathogenesis of invasive aspergillosis is by dissemination from a primary site, such as the lungs,⁷ the paranasal sinuses, or by contiguous spread.⁸ In this particular patient, the x-ray and CT scan of the chest plus the paranasal sinuses radiographs were clear without a primary lesion. It was therefore postulated that the traumatic wound by a palm tree branch resulted in the implantation of a high concentration of the aspergillus spores into the soft tissue of the neck. This idea was supported by the histopathology report, which showed

granulomatous tissue fibrosis, indicating a long-standing process, which fits in with the history of the neck injury occurring 18 months previously. Our case showed a solid mass on the left side of the neck with mediastinal and epidural extension. The effects of the mass could explain all of the patient's symptoms, namely compression of the brachial plexus, the recurrent laryngeal nerve, the spinal cord, and the displacement of the esophagus.

In conclusion, this case highlights the following: 1) that there is usually a long dormant period before the clinical manifestation of aspergillosis; 2) that clinically and radiologically the disease may be suggestive of a malignant process; 3) that contamination of all traumatic wounds by aspergillus should be considered, particularly in healthy individuals.

References

1. Rinaldi MG Invasive aspergillosis Rev Infect Dis 1983;6:1061-77.
2. Bennet JE. *Aspergillus* species. In: Mandel GL, Bennet JE, Dolin R. Principles and Practice of Infectious Disease, 4th edition. New York: Churchill-Livingstone, 1995:2306-9.
3. Aspergillosis. In: Chandler M, Caplan B, Ajello C. Wolfe Medical Atlases, 1st edition. A Colour Atlas and Textbook of the Histology of Mycotic Disease. London: Wolfe Medical Publications, 1980:34-8.
4. Vainrub B, Macareno A, Mandel S. Wound zygomycosis (mucormycosis) in otherwise healthy adults Am J Med 1988;84:546.
5. Mashimoto H, Suyama N, Araki J, Asai S, Koga H, Kohno S, et al. A case of mediastinitis and bilateral pyothorax following acute epiglottitis with concurrent aspergillus infection (English abstract). Kansenshogaku Zasshi (Japan) 1992;66:648-52.
6. Pennington JE. Aspergillus lung disease Med Clin North Am 1980;64:475-90.
7. Bardana EJ Jr. The clinical spectrum of aspergillosis. Part II. Classification and description of saprophytic, allergic and invasive variants of human disease. Crit Rev Clin Lab Sci 1981;13:85-159.
8. Hendrix WC, Arruda LK, Platts-Mills TA, Haworth CS, Jabour R, Ward GW Jr. Aspergillus epidural and cord compression in patient with aspergilloma and empyema: survival and response to high-dose systemic amphotericin therapy. Am Rev Respir Dis 1992;145:1438-44.