ASPERGILLUS INFECTION OF THE NECK WITH AN EXTRADURAL COMPONENT: AN UNUSUAL PRESENTATION

M B K Lin, S G Chee

ABSTRACT

A 66-year-old Chinese male, diabetic and hypertensive for more than 10 years, had excision of right cerebellar abscess in 1985. Histology then was suggestive of Aspergillus colonies. He presented to the ENT Department 7 years later, in September 1992, with complaints of a left-sided neck lump and fullness for 3 months which was shown on histology and culture to be Aspergillosis. CT scan findings revealed extradural involvement with erosions of parts of the cervical vertebrae. The patient, however, did not show any signs of spinal cord involvement. This case illustrates the long indolent period which Aspergillus infection can take. It also highlights the fact that it can mimic the radiological features of a highly malignant lesion. Follow-up CT scans of the patient revealed resolution of the lesion with itraconazole therapy.

Keywords: *Aspergillus, extradural involvement, itraconazole therapy.*

SINGAPORE MED J 1995; Vol 36: 678-681

INTRODUCTION

Aspergillus epidural abscess either from contiguous or haematogenous spread has been described previously by a few authors[1-5]. In all of these cases, there were signs and symptoms of cord compression.

We describe a case who presented with a clinically and radiologically invasive neck tumour but diagnosed on both histology and culture to be Aspergillosis. In this case, erosions of the cervical spine and an extradural component was seen on CT scan but the patient had no signs of cord compression.

CASE REPORT

Our case report centres on NAC, a 66-year-old Chinese male. He had a significant 10-year history of diabetes mellitus with diabetic nephropathy and was currently on insulin. He also had mild hypertension but was not on any medical treatment. There was a significant past history of cerebellar abscesses when he presented with giddiness and vomiting in May 1985. He was then noted to have otitis externa of the right side. A CT scan subsequently revealed 2 cerebellar abscesses on the right side which were excised. Histology was suggestive of Aspergillosis but no definitive growth was found when specimens were cultured. The source of the abscesses was then thought to be from the right otitis externa, hence a right mastoidectomy was performed one month later (June 1985). Again, specimens were not conclusive of Aspergillosis. He was followed up and admitted again subsequently in July 1986 for a problem of bladder calculi and had a vesicolithotomy done. In May 1987, he was admitted for treatment of urinary tract infection. A follow-up CT scan of the head in September 1987 revealed no residual abscess. He presented again to the ENT Outpatient Clinic in September 1992 with complaints of a left neck mass for 3 months which was associated with pain on and off.

Physical examination revealed fullness of the neck on the left side, just lateral to the old craniotomy scar. Postnasal space examination showed a left pharyngeal swelling. The left tympanic membrane was normal while the right was noted to have been repaired. Haematological investigation was unremarkable. The Hb was 11.7 g/dl, total white was 10,800/μl. No granulocytopenia was noted. Urea 128 mg/dl, sodium 146 mmol/l, potassium 4.9 mmol/l, chloride 108 mmol/l, creatinine 2.9 μmol/l and sugar 180 mg/dl. Anti EBV-VCA IgA and anti EBV-Ea/Ig were not significant. Postnasal space biopsy done at the outpatient was negative for malignancy. Subsequent CT scan of the neck done on 17 Septembeer 1992 showed distortion of the nasopharynx with elevation of the left wall due to a soft tissue mass. Extradural component of this mass was seen in the C2-3 thecal space, especially on the left antero-lateral aspect. Impression was that of a tumour. (Figs 1, 2 and 3)

The patient was subsequently admitted for biopsy under general anaesthesia. Excision biopsy of the left neck mass

Fig 1 - Axial CT scan of the neck at the level of C3 vertebra done on 17 September 1992. There is a ill-defined mass in the left paraspinal region with extension into the left parapharyngeal space. Distortion of the nasopharynx is also seen.

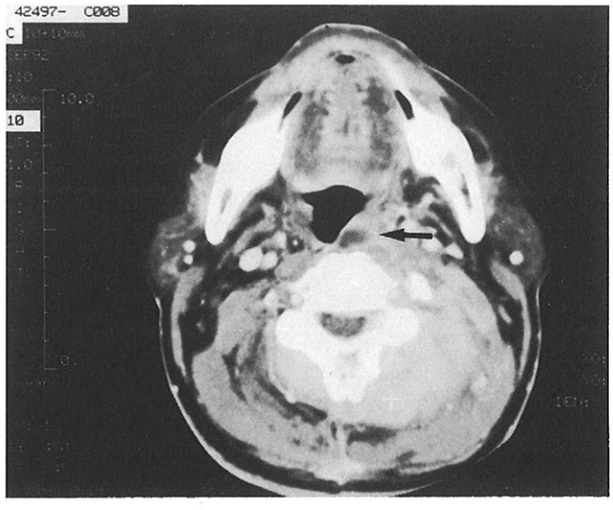

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Fig 2 - Scan taken 10mm inferior to that of Fig 1 showing the extent of the lesion.

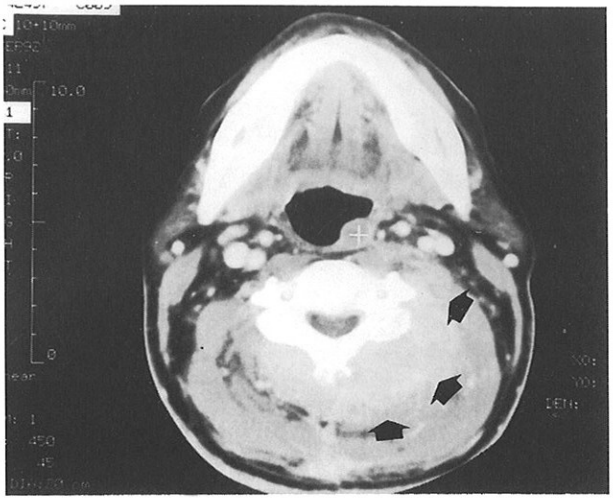


Fig 3 - A magnified view of the C2 vertebra showing the extradural component of the lesion.

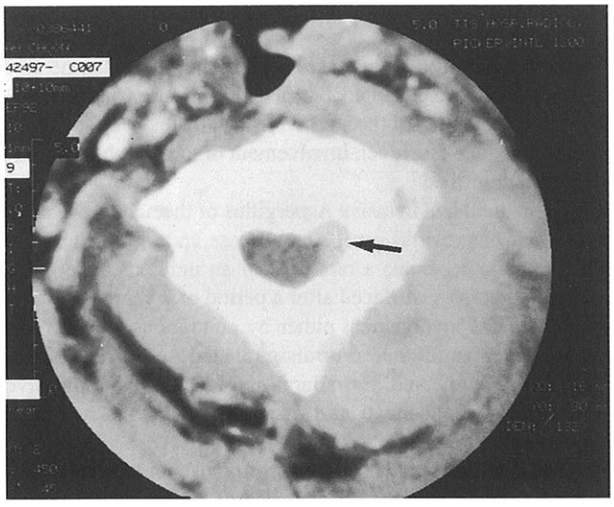
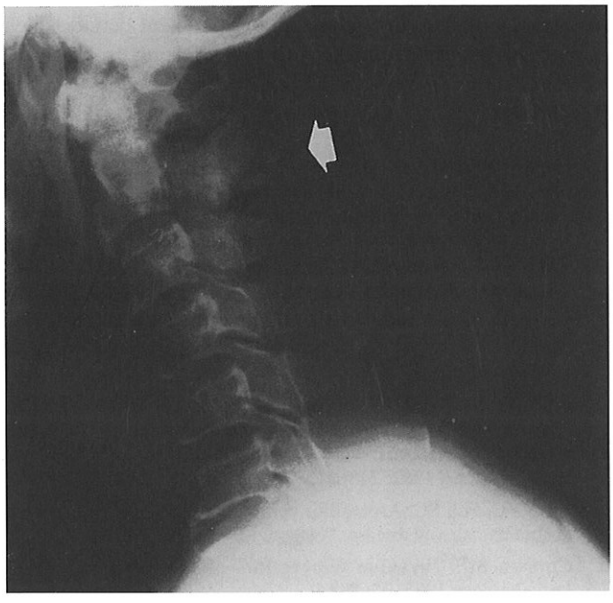


Fig 4 - Lateral cervical spine X-ray showing erosions of the spinous process of C2.



revealed an indurated left paraspinal muscle, which on frozen section, revealed fungal colonies. Tumour-like tissues on frozen section revealed chronic inflammation. Pharyngeal biopsy at the same setting showed a 1cm x 2cm moderately tense swelling behind the posterior pillar of the tonsil. On deep biopsy, purulent material was discharged with a reduction in mass.

Subsequent histology from the left paraspinous muscle showed granulomatous tissue with inflammation and fibrosis. Within the cavities were clumps of fungal organisms consistent with Aspergillosis. Subsequent cultures from the wound grew *Aspergillus sp*. The patient was then referred to the Medical Unit and was started on itraconazole. A cervical spine X-ray revealed erosions of the spinous process of C2. (Fig 4). A follow-up CT scan of the neck on 28 October 1992 showed asymmetry of the nasopharynx with the left being more bulky than the right. The muscles of the neck were noted to be ill-defined and swollen in the posterior part of the neck especially on the left side. An oval-shaped hypodense area adjacent to the spaces of C2 and C3 which contained locules of air was thought to be an abscess cavity or the result of a recent operative (Fig 5) procedure.

Erosions were also seen in the posterior elements of C2, C3 and the left lamina of C3. The left half of C1 and posterior arch appeared slightly moth-eaten. Spread of inflammatory process into the spinal canal at C2-3 with filling up of the left lateral recess of C2 as well as an extradural mass in the spinal cord was noted (Fig 6). The patient was referred to the neurosurgeons who decided against any surgical intervention as the fungal mass had already encrusted the upper cervical spinal cord. Any clearance would cause high morbidity and even death. Hence it was decided to refrain from surgery unless any neurological signs appeared.

The patient was subsequently discharged in November 1992 with itraconazole 200 mg bd. The dose of itraconazole was subsequently reduced to 100 mg bd and continued for a total of 12 months from the time of commencement. On the last follow-up on 11 November 1992, he was noted to be well. A follow-up scan of the neck 8 months later on 7 July 1993 showed resolution of the inflammatory process with areas of scarring (Fig 7 and 8).

DISCUSSION

Aspergillus infections are classically described in the following sites:

1. Lungs - mainly due to *Aspergillus fumigatus* and classified into 3 major types: mycetoma, invasive (acute pneumonia) and allergic bronchopulmonary Aspergillosis.
2. Sinuses - chronic sinusitis
3. Otomycosis - growth of Aspergillus on cerumen and detritus within the external auditory canal.
4. Eye - chronic Aspergillus keratitis due to trauma to the cornea
   - endopthalmitis from trauma or surgery.
5. Intracardiac or intravascular prosthesis infection.
6. Invasive Aspergillus of the head and neck.

Aspergillus is ubiquitous in the environment and is frequently present in decaying vegetation. It also occurs as a commensal in 10% of random sputum samples. The portals of entry are usually via inhalation, skin (especially old surgical scars), the alimentary canal, and the paranasal sinuses.

Invasive Aspergillus of the head and neck is a rare condition[6] whereby the fungus becomes an opportunistic pathogen in certain hosts. Certain predisposing factors that have been associated with this aggressive disease in previous reports are as follows:

1. Immunosuppression, especially granulocytopaenia[5] in 90% of the cases. This maybe due to the presence of an underlying haematologic malignancy[5], tuberculosis, treatment with broad spectrum antibiotics, long-term steroids, antineoplastics, radiotherapy or collagen vascular disease.
2. Underlying debilitating disease, such as malignancies and

Fig 5 - Axial CT scan of the neck at the level of C3 done on 28 October 1992. (post biopsy)

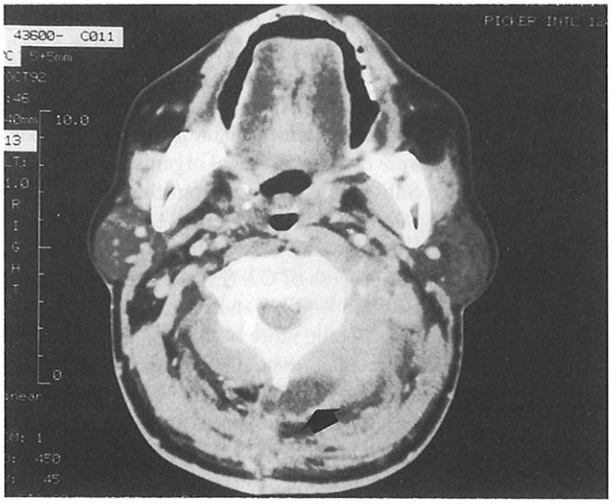


Fig 6 - Magnified view of the scan of the neck done on 28 October 1992.

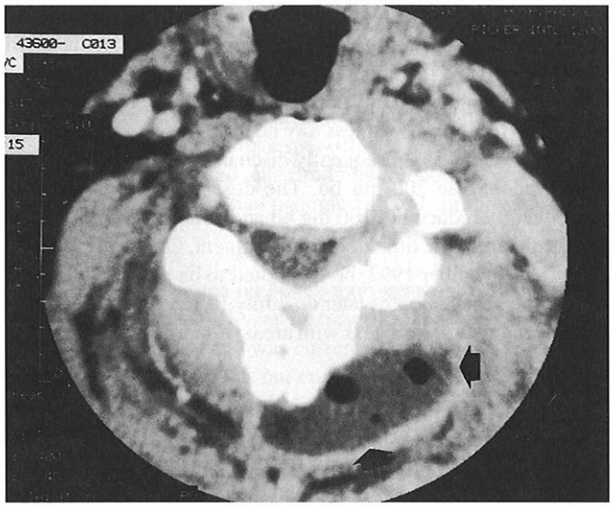


Fig 7 - Axial CT scan at the level of C3 done on 7 July 1993, ten months later. There are areas of scarring seen in the left paraspinal region. The main bulk of the lesion has largely resolved.

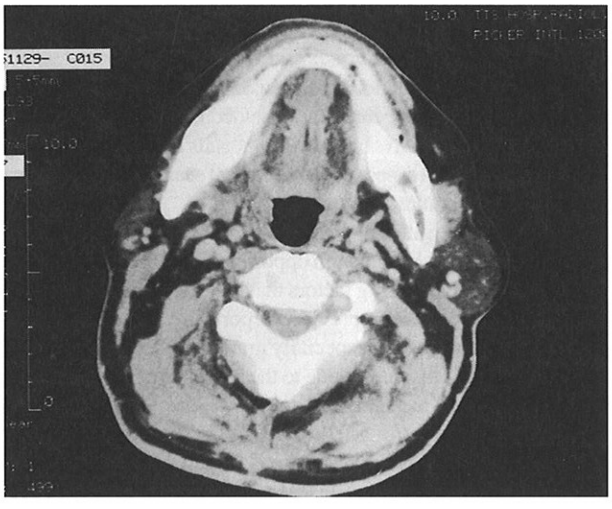
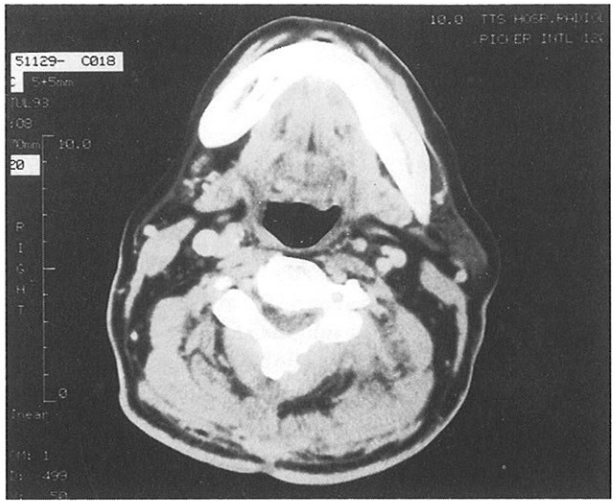


Fig 8 - Scan 15 mm inferior to the above scan showing scarring in the left paraspinal region.



diabetes.

3. Breakdown of local barriers in old operative sites are an important source of the infection.

In the majority of the cases reported[6-8] the outcome was invariably fatal despite intensive medical and surgical treatment. The affected regions of the neck were usually the deep spaces[6,8] as in our patient. However, involvement of the lymph nodes has also been described[7].

Our patient had invasive Aspergillus of the neck[1] probably as a result of previous cerebellar abscess and otitis externa in 1985. This underscores a rare case of an indolent Aspergillus infection that has resurfaced after a period of 7 years.

Extradural involvement either by contiguous spread from pulmonary aspergilloma[1-5] or disseminated Aspergillosis have been well documented. Extradural involvement from invasive Aspergillus of the head and neck, however has not been documented previously.

It is generally agreed that antifungal chemotherapy and surgical debridement are the mainstay of treatment in invasive Aspergillus. Another important factor that should not be neglected is the recovery of immunologic competence[6].

This unusual case highlights the problems of diagnosis when both the clinical and radiological findings are suggestive of a malignancy. It also highlights the long indolent period which Aspergillus infection can take before manifesting itself.

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