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Clinical Records

Pneumocystis carinii infection in bilateral aural polyps in a human immunodeficiency virus-positive patient

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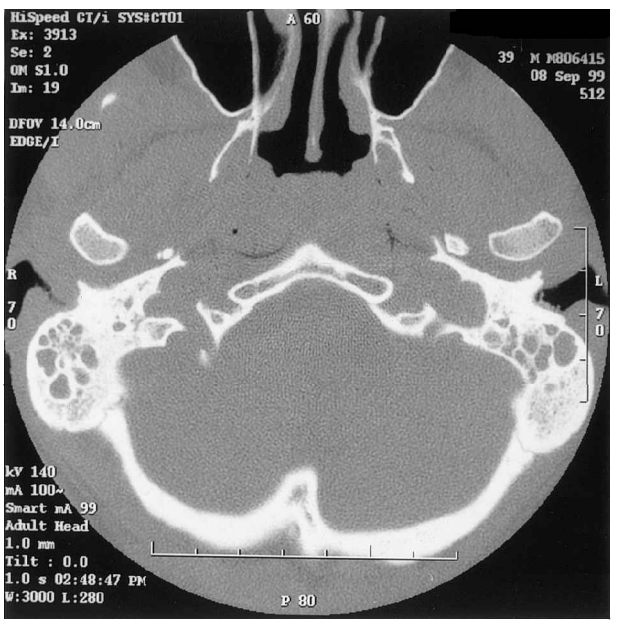
Abstract
Pneumocystis carinii is an opportunistic infection found in patients with impaired immunity. Under favourable conditions the parasite can spread via the blood stream or lymphatic vessels and cause extrapulmonary dissemination. We report a case of *P carinii* infection presenting as bilateral aural polyps, otitis media and mastoiditis in human immunodeficiency (HIV)-positive patient with no history of prior or concomitant *P carinii* infection.

Key words: Pneumocystis carinii; Ear, Middle; HIV

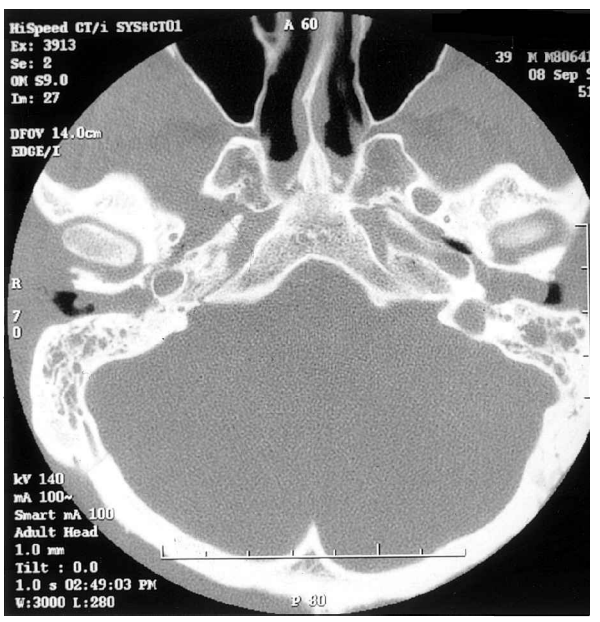
Case report

A 39-year-old bisexual HIV-positive male patient presented to us with two-week history of bilateral bloody otorrhoea and hearing loss. There was no history of prior ear infection nor hearing loss. Since initial diagnosis of HIV in 1991, he had not suffered any illness nor had he received any prophylactic treatment. He was not on any medication and was otherwise well and asymptomatic without cough or breathing difficulties.

On auroscopic examination, the external auditory canal was completely filled on both sides with fleshy reddish brown polyps with a purulent thick discharge obscuring the view of the tympanic membrane. He had no lymphadenopathy and auscultation of his chest was clear.



(a)



(b)

FIG. 1(a) and 1(b)
CT scan and mastoids showing soft tissue mass filling both the external auditory canals and opacity in both the middle ear and mastoids.

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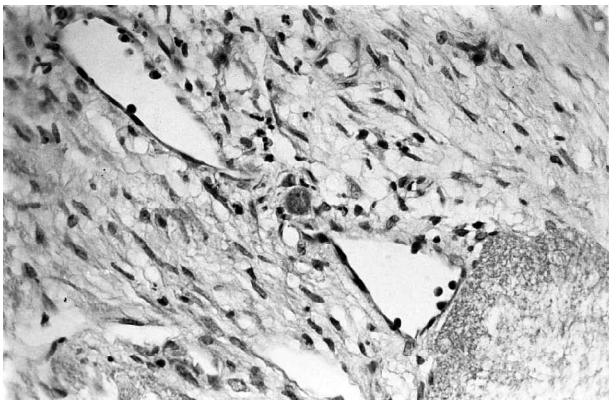
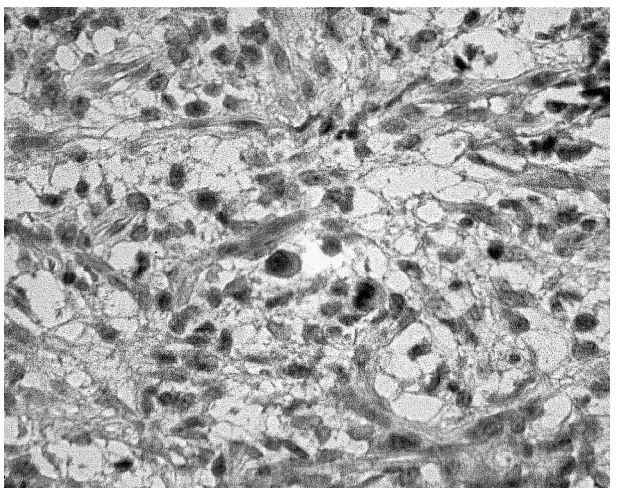


(a)



(b)

FIG. 2(a) and (b)

(a) Histological section showing foamy granular material and an enlarged endothelial cell with a large inclusion suggestive of cytomegalovirus infection (H&E; ×40). (b) High power view showing an enlarged endothelial cell (H&E; ×100)

A computerized tomography (CT) scan of his mastoids showed a soft tissue mass filling the external auditory meatus and opacity in the mastoid antrum and air cells (Figure 1(a) and 1(b)). An audiogram showed a bilateral mild conductive hearing loss and chest X-ray did not show any evidence of infection. Laboratory study showed a lymphocyte count of 179 cells/microlitre.

The patient underwent examination of both the ears under a general anaesthetic and was found to have fleshy aural polyps protruding through the tympanic membrane from the middle ear. The aural polyps were excised for histological examination. On haemotoxylin and eosin (H and E) staining, the polyps were composed of squamous epithelium with underlying foamy eosinophilic material. There was a paucity of inflammatory cells. A number of endothelial cell nuclei were enlarged and eosinophilic, with appearances suggestive of cytomegalovirus (CMV) infection (Figure 2(a)) for low power and Figure 2(b) for high power). Silver staining showed numerous cup and disc shaped cysts throughout the foamy granular sub-epithelial tissue, which is characteristic of *P carinii* infection (Figure 3).

The patient responded dramatically to twice daily, three weeks of oral cotrimoxazole (400 mgms of trimethoprim and 90 mgms of sulfamethoxazole), which totally cured his mastoiditis, the aural polyps completely disappeared and the tympanic perforation healed with minimal scarring.

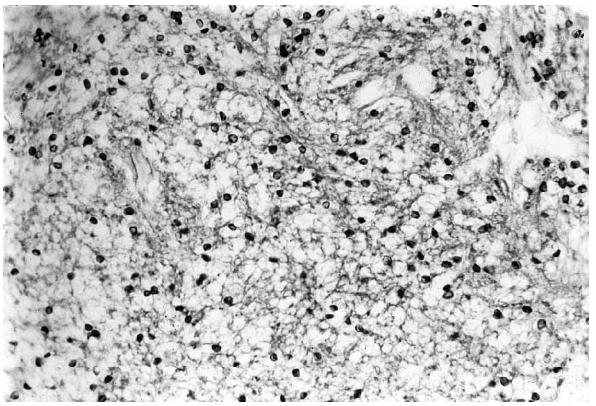


FIG. 3

Demonstrates numerous cup and disc shaped cysts throughout the foamy granular subepithelial tissue characteristic of *Pneumocystis carinii* organisms (Silver stain; ×40)

The patient has been followed up for two years, the tympanic membrane remains intact with a healed perforation with no evidence of infection and the patient has a normal audiogram.

Discussion

Unusual cutaneous lesions associated with acquired immunodeficiency syndrome (AIDS) include viral, fungal and parasitic infections. The taxonomy of *Pneumocystis* has been controversial. It was once considered to be a protozoan, but is now thought to be more closely related to fungi.[1–3] *P carinii* is thought to exist normally within the lungs of humans and other mammals and causes opportunistic infection in the compromised host, impaired cellular immunity being the major host factor that predisposes to infection. Extra-pulmonary manifestations of *P carinii* are rare, however, they are becoming recognized more frequently, with the increasing use of aerosolized pentamidine as prophylaxis for *P carinii* pneumonia (PCP). *P carinii* has been identified in immunosuppressed patients in lymph nodes, peripheral blood, myocardium, stomach, small intestine, bone marrow, adrenal gland, thyroid gland, and retina.[4,5] The first reported case of extra-pulmonary *P carinii* was first published in 1960.[6] Anderson reported *P. carinii* in the sinusoids of hilar lymph nodes of a Japanese man who had died of PCP. It was not until 1982 that extra-pulmonary *P carinii* infection was reported in a man with AIDS.[7]

Middle ear and mastoid air cells are normally sterile. Infection at this anatomical site could develop by several routes. The area could become infected by haematogenous spread of infection,[5] by extension of infection through lymphatic channels, by retrograde spread through the eustachian tube or from the external auditory canal through the tympanic membrane.[8] Our case is unusual as there was no on-going pulmonary infection with *P carinii*, nor was there any evidence of pre-existing ear disease. Infection of the middle ear with *P carinii* via the eustachian tube seems to be the most likely explanation. The embryological derivation of the epithelium of the tympanic membrane is analogous to that of the alveolus of the lungs; and it has been proposed that middle ear and mastoid air cells may be as congenial for the development of *P carinii* infection as are the lungs.

CMV is a beta herpesvirus[9] and has a worldwide distribution. The virus is not spread by casual contact but requires repeated or prolonged intimate exposure for

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transmission.[10] In late adolescents and young adults, CMV is often transmitted sexually, asymptomatic viral carriage in semen and cervical secretion is common. Once infected an individual carries the virus for life, the virus getting activated when there is a compromise of T-lymphocyte mediated immunity as occurs in AIDS,[3] leading to several clinical illnesses such as colitis, pneumonitis, hepatitis, chorioretinitis, renal tubulitis, meningoencephalitis, oesophagitis and adrenalitis.[11] CMV may itself contribute to further T-lymphocyte hyporesponsiveness, which often precedes superinfection with other opportunistic pathogens such as *P carinii*.[12]

To our knowledge this is the seventh case of bilateral aural pneumocystosis in the absence of pulmonary disease, in English literature. All the reported cases have been in men with the average age of presentation being 33 years. None of the records mention the extension of the disease from temporal bone until recently Patel *et al.* reported a case of otic pneumocystosis in a unilateral aural polyp, in a young HIV-positive man, extending into the middle cranial fossa[13] with extensive bony erosion.

All the reported cases of aural polyps with pneumocystosis have responded well to a combination of intravenous and oral antiprotozoal agents such as trimethoprim, sulfamethoxazole and dapsone and none required mastoid exploration.[5,8,13,14] Our patient responded very well to a 21-day treatment of oral trimethoprim and sulfamethaxazole combination and he has been followed up for two years; the tympanic membrane looks completely healed with normal hearing.

This case has been presented to illustrate how an apparently routine ENT problem can mask a far more serious diagnosis such as HIV, of which medical staff may be completely unaware.

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