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Case Report

Syringomyelia presenting as a delayed complication of treatment for nocardia brain abscess

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Objective: Syringomyelia is defined as a dilatation of the central canal of the spinal cord which often leads to neurologic impairment. Syringomyelia has not previously been reported as a late complication for the treatment of brain abscess. In this report, we review a case involving this unusual association.
Clinical presentation: A 25 year-old woman sustained a nocardia brain abscess initially presenting as a pulmonary infection. Treatment led to the development of multiloculated hydrocephalus and syringomyelia.
Intervention: Treatment included placement of multiple ventriculoperitoneal shunts and a syringo-pleural shunt. This resulted in stabilization of neurologic symptoms.
Conclusion: The possibility of developing syringomyelia should be considered in any case involving post-infectious hydrocephalus.
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Keywords: syringomyelia; brain abscess; nocardia

Introduction

Syringomyelia refers to the development of longitudinal cavities within the spinal cord.[1] The cavities can arise from the central canal of the spinal cord. In this situation the dilatation is sometimes refered to as hydromyelia; however, in some instances the lining of the central canal (ependyma) is incompetent and extension of the cavity occurs into areas away from the central canal. As the cavity distends it exerts pressure on the spinal cord parenchyma causing damage which is manifested by neurologic deficits in a given patient.[2,3]

The most common cause of syringomyelia is Arnold Chiari 1 malformation and its associated hindbrain abnormalities.[1] However, a number of other etiologies have been described. These include tumors, post-traumatic and post-infections. Syringomyelia associated with infections usually result from meningitis with subsequent scarring of either the subarachnoid space or the region of the foramen magnum. In this report, we describe a case of syringomyelia due to nocardia brain abscess. We found no other reports in the literature describing the development of syringomyelia after treatment for a brain abscess.[4–6]

Case report

A 25 year-old female was admitted to an outside hospital with a chief complaint of cough and shortness of breath. Her medical history was significant for ulcerative colitis for which she was taking prednisone and immuran. Sputum cultures were positive for nocardia asteroides. She was then started on intravenous trinethroprim-sulfamethoxazoles (TMP-SMX). Because of complaints of headache and lethargy, a computerized tomography (CT) scan with contrast was obtained which demonstrated a ring enhancing lesion in the occipital lobe consistent with a brain abscess (Figure 1). Ventricular enlargement was also seen on the initial CT scan (Figure 1). She then underwent a craniotomy for culture and drainage of the abscess.

Cultures of the brain abscess also grew nocardia asteroides. The patient became comatose 2 weeks after the surgery. Subsequent CT scanning showed interval development of frank hydrocephalus indicating worsening of the ventricular enlargement seen on initial CT scan. Diffuse ependymal enhancement was also seen on CT suggesting the presence of ventriculitis

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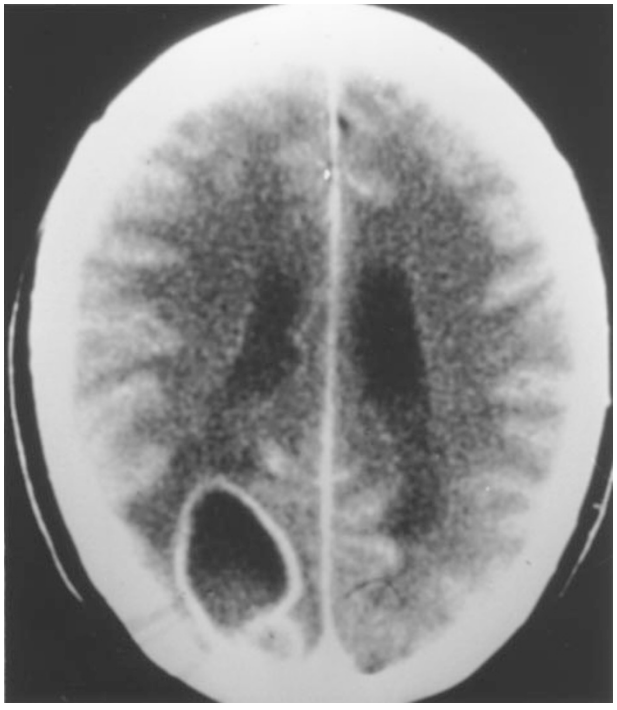

Figure 1 Axial CT scan of the brain with contrast showing enhancing mass in the occipital lobe (brain abscess). Ventricular enlargement is also seen

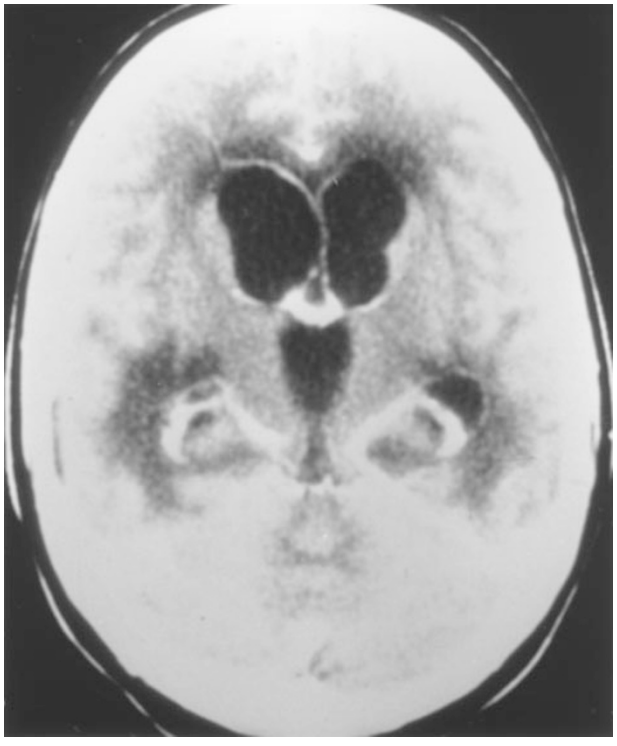

Figure 2 Axial CT scan of the brain with contrast showing worsening of initial ventricular enlargement (hydrocephalus). Frank enhancement of the ependyma is also seen suggesting ventriculitis

(Figure 2). There was no evidence of recurrence of the abscess on follow up CT scan. She was then transferred to the neurosurgical service at Temple University Hospital. On admission, the patient was afebrile. She was comatose with weak flexion to noxious stimuli in all extremities. Pupillary light reflexes were intact. Her gag reflex was depressed. Laboratory values revealed a white blood cell count of 12.0×10 d/L. Admission chest X-ray revealed no evidence of pneumonia.

Hospital course

Shortly after admission, a right frontal ventriculostomy was placed for relief of hydrocephalus. It was noted on follow up scans that there was inadequate drainage of the ventricles due to loculation and compartmentalization of the ventricular system (Figure 3). This required placement of multiple ventricular catheters for drainage of the ventricles. An attempt to recommunicate the ventricles by intraoperative fenestration was not thought possible because the cavities formed were not large enough to allow such an approach. Multiple cultures taken from cerebrospinal fluid remained negative for nocardia asteroides or other organisms. The CSF protein of the obtained fluid was normal (11 mg/dl). Multiple ventriculostomies were then converted to internal shunts. Follow up CT scans showed adequate drainage of the ventricular system (Figure 4).

After a 3-month hospitalization, the patient gradually improved neurologically, such that she was arousable and able to follow commands in all extremities. At the time of transfer to a rehabilitation service (5 months after admission to our service), she still exhibited profound neurologic deficits characterized by paresis of all extremities, dysarthric speech, left facial weakness and a depressed gag reflex.

She was maintained on oral TMP-SMX for a total of 1 year after the initial diagnosis of nocardia.

Subsequent course

Over the ensuing 9 months after transfer to the rehabilitation service, the patient continued to improve neurologically with improved strength. She was wheel-chair bound, but had functional strength in her upper extremities. Cognitive function including speech, memory and ability to perform activities of daily living also improved. However, during the period of recovery she was readmitted to the neurosurgery service on seven occasions for revision of her ventriculoperitoneal shunts due to malfunction.

Three years after the initial diagnosis of nocardia abscess, the patient complained of neck pain, upper extremity pain and motor loss of the upper extremities. Cranial cervical magnetic resonance imaging (MRI) showed dilatation of the fourth ventricle consistent with malfunction of the patient's previously placed fourth ventricular shunt. In addition, a holocord

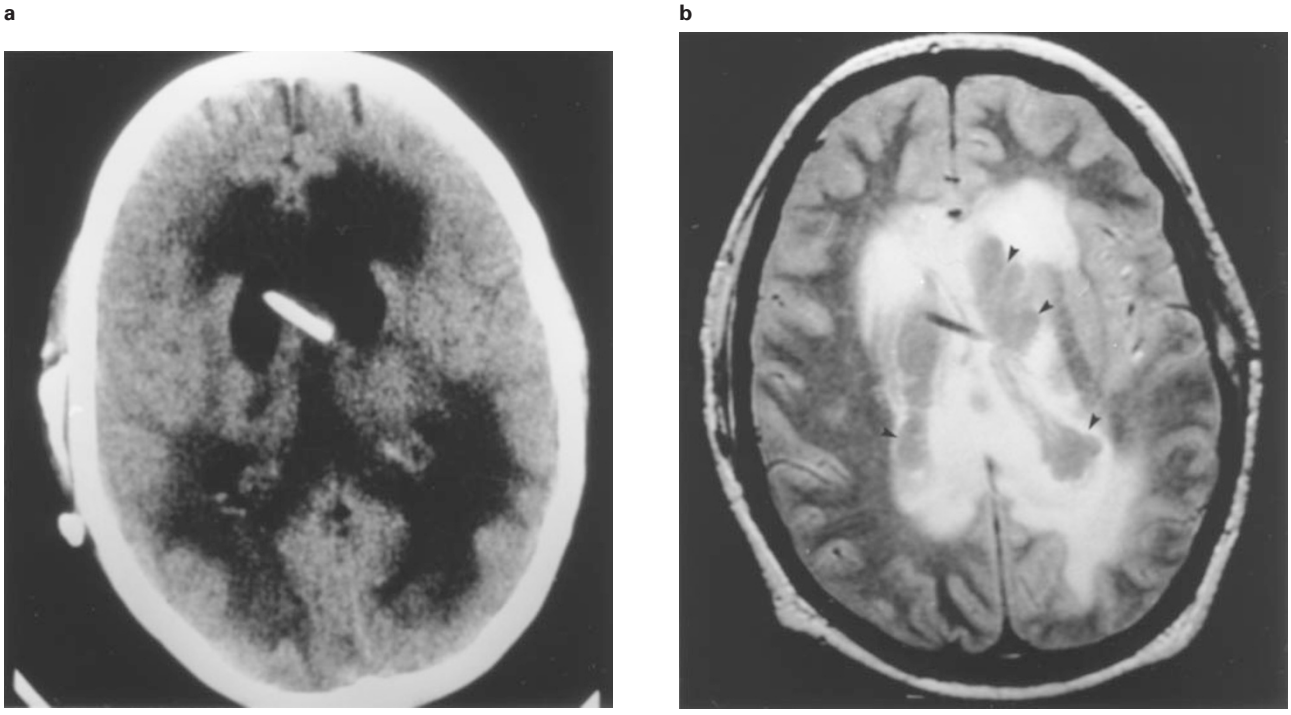

Figure 3 (**a**) Axial CT scan of the brain after placement of ventriculostomy showing lack of decompression of ventricles. (**b**) Axial MRI of the brain (T-1 weighted image) showing loculations in ventricular system. See arrows pointing to loculations

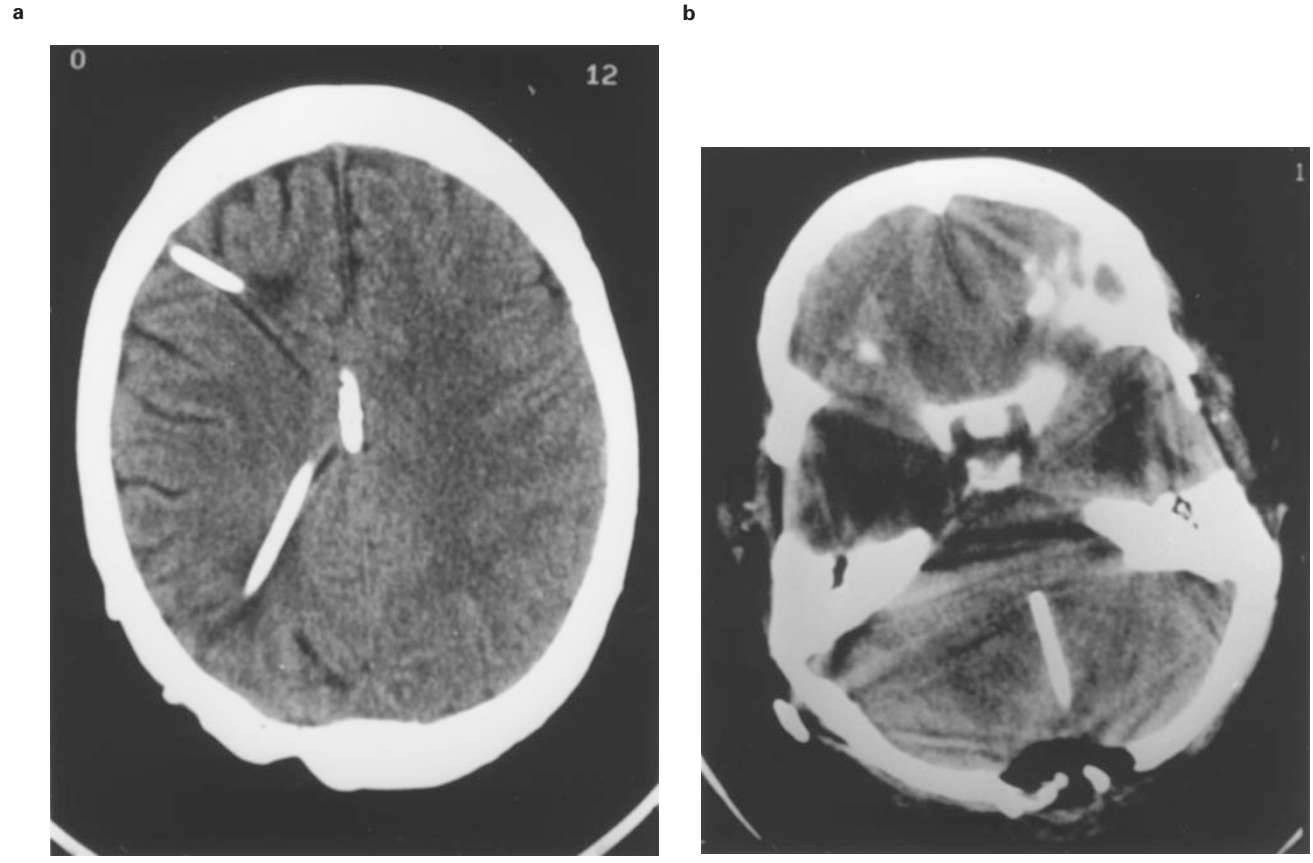

Figure 4 (**a**) Axial CT scan of the brain showing adequate decompression of ventricular system by a right frontal, right parietal and interhemispheric shunts. (**b**) Axial CT scan showing a ventricular catheter in fourth ventricle

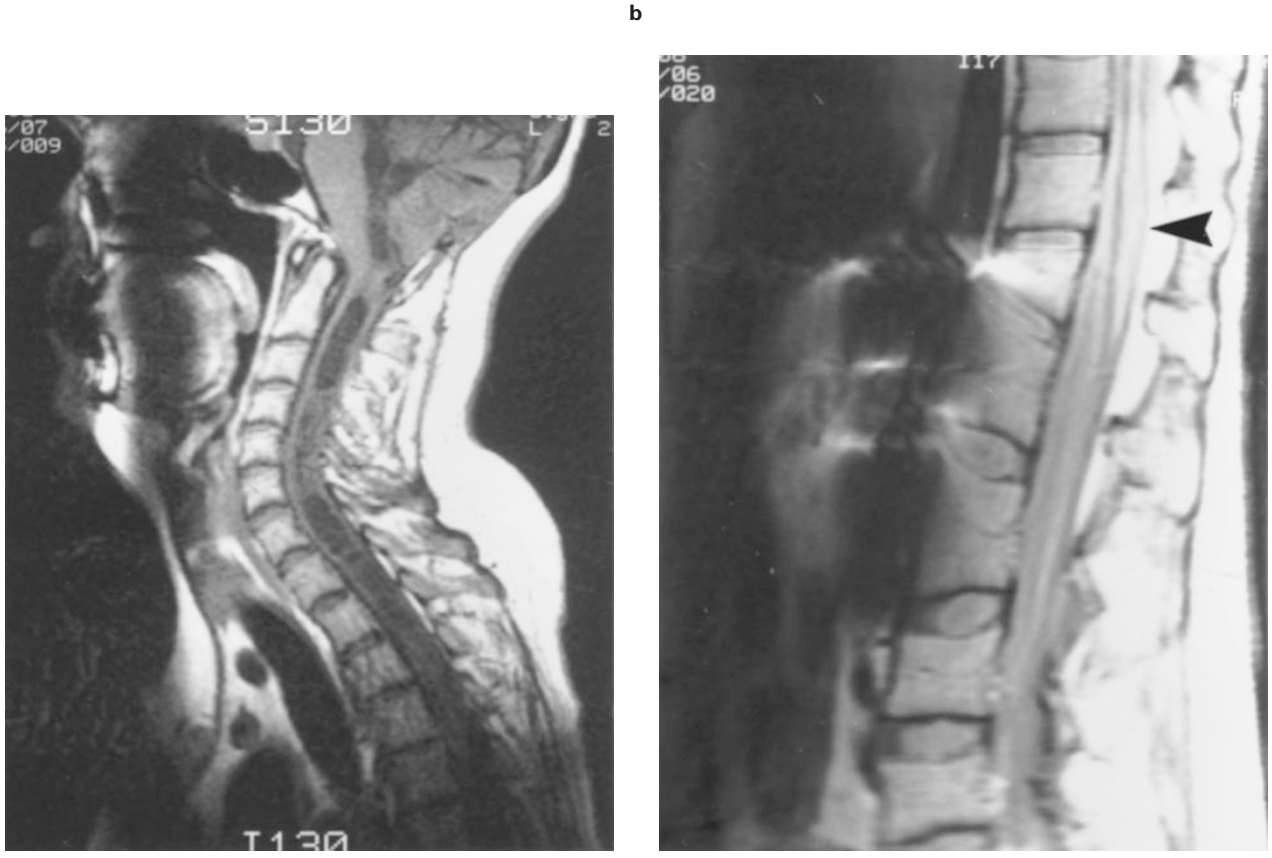

Figure 5 (**a**) Sagittal MRI (T-1 weighted image) showing cervicothoracic syrinx. (**b**) Sagittal MRI (T-2 weighted image) showing extension of syrinx through the entire length of spinal cord (holocord). See arrow pointing to syrinx cavity

syrinx was noted extending from C2 to T12 (Figure 5). The patient underwent revision of the fourth ventricular shunt and placement of a zero pressure syringo-pleural shunt in the mid-cervical region. The patient's pain syndrome and upper extremity strength returned to baseline after surgery. However, she subsequently expired due to complications of ulcerative colitis 4 years after her last surgery.

Discussion

Nocardia is a gram positive partially acid fast aerobic actinomycete.[7] In the environment, it is found in soil and decaying vegetables. It is primarily a pulmonary pathogen which presents most commonly as pneumonia.[8–11] When nocardia gives rise to a brain abscess, the infection usually originates in the lungs.[4] However, there are cases reported which arise originally in the brain. Nocardia is a rare cause of brain abscess comprising 1% to 2% of cases in most series.[12–14]

As a pathogen, it is commonly associated with immunocompromised patients.[15,16] Nocardia brain abscesses are usually multiloculated and thick walled.[17,18] Nocardia carries a significantly higher rate of mortality than brain abscesses due to other pathogens. Mortality ranges from 24% to 50%.[4]

Syringomyelia occurring as a delayed manifestation of treatment for a brain abscess has not previously been reported.[2,4,5,19,20] In this case, the pathophysiology of syringomyelia could be due to either of two causes. The first cause could be continued inflammatory scarring of the subarachnoid space and central canal of the spinal cord as occurred in the ventricular system. However, we did not observe any enhancement of the leptomeninges in relation to the basal cisterns or craniocervical junction on MRI with contrast. The presence of enhancement in these regions would serve as evidence for inflammatory scarring of the subarachnoid space. The other etiology could be chronic fourth ventricular outflow obstruction due to a combination of loculation of the fourth ventricle and shunt malfunction. These two factors would lead to transmission of fluid into the central canal of the spinal cord.

In this report on an unusual complication of brain abscess, the occurrence of multiloculated hydrocephalus in this patient may be related to the organism type (nocardia asteroides) which typically presents as a multiloculated brain abscess.[7] Ventriculitis in this case led to a tremendous post-infectious inflammatory response causing multiloculated hydrocephalus and ultimately syringomyelia. Treatment led to stabiliza-

tion of neurologic symptoms. The possible development of syringomyelia should be considered in all patients with multiloculated hydrocephalus.

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