Late complication after tropic storm accident: subcutaneous and intracranial actinomycetoma

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ABSTRACT

We report a 53-year-old farmer who developed subcutaneous and cerebral masses 24 years after penetrating trauma during a tropic storm. Computed tomography scans, magnetic resonance imaging and histopathology disclosed actinomycetoma, a disease that rarely develops after trauma and is only occasionally seen with intracranial manifestation. Clinically, the cutaneous manifestation resembled acne keloidalis nuchae or dissecting folliculitis of the scalp. He was treated by neurosurgery and antibiosis.

Key words: Actinomycetoma • Cerebral infection • Nocardiosis

INTRODUCTION

Mycetoma is a clinical syndrome of localised, indolent, swollen lesions with sinuses involving cutaneous and subcutaneous structures. It results from traumatic implementation of microorganisms of the soil. Actinomycetomas can be differentiated into two major categories according to the organisms responsible: (a) actinomycetomas caused by *Actinomyces* spp., that is bacteria, and (b) eumycetomas caused by fungi (1,2).

Actinomyces spp. are stainable by haematoxylin–eosin, where the periphery of actinomycetomas stains red (Höppli–Splendore reaction). They also react with Grocott's stain with a deep blue periphery and black elements in the centre. A network of fibrous material can be identified with higher magnification microscopy (1).

Actinomycosis of central nervous system (CNS) by is a rare manifestation. Types of lesions include brain abscess (2/3 of cases), meningitis or meningoencephalitis, actinomycetoma, subdural empyema and epidural abscess (3–5). In most of the cases, CNS infection develops from distant sites like the lungs or contiguous foci (ears and sinuses). Penetrating head injury is another less common cause (5). Here, we report a case of CNS actinomycosis after a tropical storm accident.

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CASE REPORT

History and clinical findings

A 53-year-old farmer was seen by one of us (SN) in March 2007 in a neurosurgical hospital. This man said that he had suffered a penetrating injury to the occipital scalp in 1983 during

Key Points

- actinomycetomas can be differentiated into two major categories according to the organisms responsible: (a) actinomycetomas caused by Actinomyces spp.,that is bacteria, and (b) eumycetomas caused by fungi
- here, we report a case of CNS actinomycosis after a tropical storm accident
- a 53-year-old farmer was seen by one of us (SN) in March 2007 in a neurosurgical hospital
- he had suffered a penetrating injury to the occipital scalp in 1983 during a tropical storm that had hit the town of Vadodara, Gujarat, India

Key Points

- the neurosurgeon referred the case to one of us (SV) who gave a clinical diagnosis of acne keloidalis nuchae, assuming at that point in time that we may be dealing with two separate pathologies, that is extracranial and intracranial, which need not be interrelated
- no microorganisms were found from the skin tissue

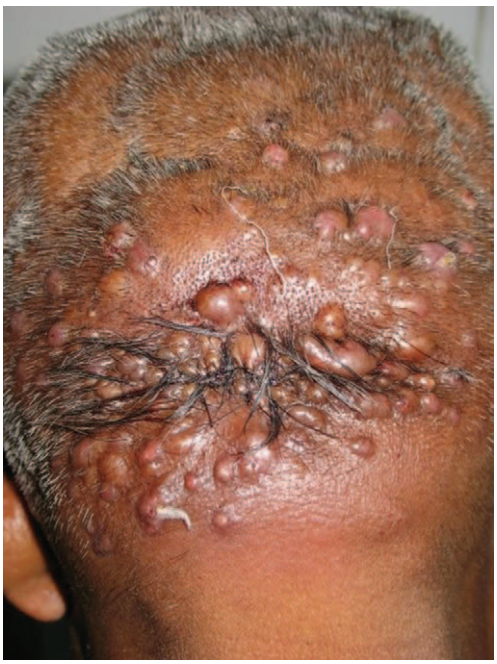

Figure 1. Clinical presentation of the patient suggested acne keloidalis nuchae.

a tropical storm that had hit the town of Vadodara, Gujarat, India. He was treated by a local surgeon who sutured the wound and gave him a course of antibiotics, following which his recovery was uneventful. About 9 years later, he started developing swellings on the scalp, which appeared spontaneously, were painful, exuded pus and healed on their own (Figure 1). He also noticed significant thickening of the scalp. At some point during these episodes, he sought the help of general practitioners who treated him intermittently.

The patient developed severe headache since the beginning of 2007, which prompted him to seek the opinion of a neurophysician. He also had dimness of the vision, which he ignored until the headache was finally investigated. The neurologist documented severe papilloedema. A computed tomography (CT) scan was carried out that showed an intracranial mass, which is described in detail in the following discussion. There was no localising sign like hemiparesis or hemiplegia or speech abnormality.

CT scan (plain and contrast enhanced) showed a hyperdense, intracranial but extracerebral lesion in the left occipitoparietal region with involvement of the posterior sagittal sinus and extending to the right occipital region with a dural base lesion of 28 × 26 mm size on the left side of falx. There was evidence of a surrounding oedema in the left occipito-temporoparietal lobe and focal, ill-defined enhancement of the splenium of callosum on the left side. An effaced left lateral ventricle with a shift to the right side was noted. There was evidence for defects in the left occipital bone and the parietal bone with sclerosis. A large epicranial enhancing soft tissue mass was found in the left occipitoparietal region, and a small epicranial abscess was seen frontally.

The neurosurgeon referred the case to one of us (SV) who gave a clinical diagnosis of acne keloidalis nuchae, assuming at that point in time that we may be dealing with two separate pathologies, that is extracranial and intracranial, which need not be interrelated.

During this time, a magnetic resonance imaging (MRI) was carried out that showed some significant findings, and the clinical photographs with all the imaging and histopathology reports were sent to Germany to one of us (TK) who documented multiple interconnecting channels between outside the skull and within, concluding the assumption that there indeed was a related pathology.

Sagittal MRI of the head (T1 weighted, after i.v. contrast administration) showed an inhomogeneous enhancing mass of the scalp most impressive in the occipital region containing a circumscribed abscess in the frontal midline, size 40·2 × 30·0 × 26·1 mm (Figure 2). There were communications between the scalp formation and the intracranial epidural space by multiple transosseus fistulas. An epidural infiltration with mass effect and associated thrombosis of the sigmoid and straight sinus can be seen. There was a remarkable thickening of the local dura. Intracerebral, extra-axial heterogeneous enhancing mass was found in the left occipital region with perifocal oedema and compression posterior body of the left ventricle.

Histopathology of a deep skin biopsy shows a granulomatous and foreign body granulomatous inflammation surrounding hair shafts lying free in the dermis. A moderately dense superficial and deep lymphocytoplasmic infiltrate is present around the vessels. The overlying epidermis shows an irregular psoriasiform and pseudocarcinomatous acanthosis. Granulomas consist of lymphocytes, epithelioid cells, histiocytes and foreign body giant cells, some of which engulf free hair shafts. A few foci of suppuration are also present. Therefore, the first diagnosis was acne keloidalis.

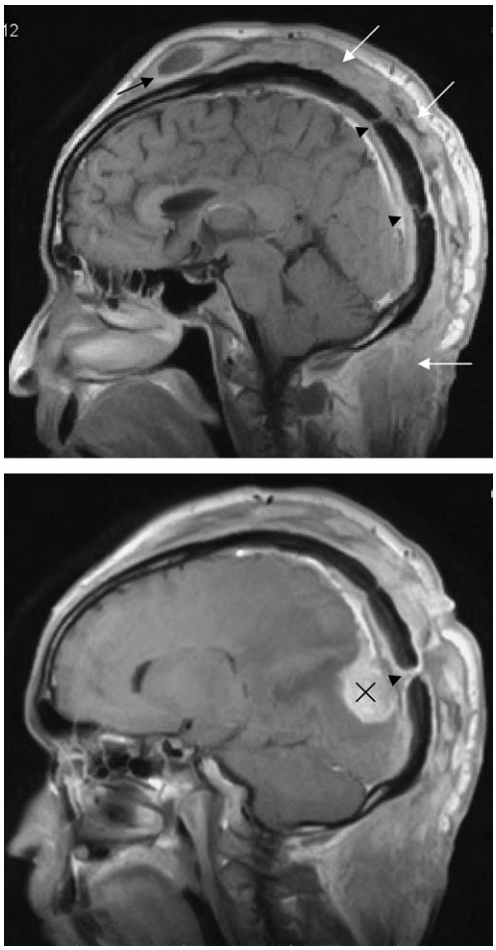

Figure 2. Sagittal magnetic resonance imaging of the head (T1 weighted, after i.v. contrast administration). Inhomogeneous enhancing mass of the scalp most impressive in the occipital region (white arrows) containing a circumscribed abscess in the frontal midline (black arrow). Communication between the scalp formation and the intracranial epidural space by means of multiple transosseus fistulas (arrowheads). Epidural infiltration with mass effect and associated thrombosis of the sigmoid and straight sinus. Thickening of the local dura. Intracerebral, extra-axial heterogeneous enhancing mass (cross) in the left occipital region with perifocal oedema and compression posterior body of the left ventricle.

No microorganisms were found from the skin tissue. Some tissue samples of the dura were sent for histopathology that showed the typical sulphur granule (in German 'Druse') with a dense neutrophilic surrounding infiltrate. Sections showed gram-positive and branched microorganisms that suggested to be *Actinomyces* spp. (Figure 3). In contrast to *Nocardia* spp., the organisms were acid-fast negative. Blood samples screened for intravascular spread of fungi were found to be negative in culture. Culture of organism from the skin tissue was unable to grow any organism.

A clinical diagnosis of actinomycetoma was made jointly.

Treatment and outcome

During the discussions between the dermatologists, it was decided that the neurosurgeon goes ahead and tries to excise the lesion by craniotomy in an attempt to reduce pressure symptoms of headache and dimness of vision. Much to the surprise of the neurosurgeon, the craniotomy instruments broke during surgery, implying uncharacteristic thickness and hardness of the scalp and bone, findings that were later corroborated by imaging studies. The scalp was also unusually vascular, and there was difficulty closing the wound. The tumour was found to be lying on either side of the sagittal sinus at the junction of the sagittal and transverse sinus. An attempt was made to remove as much of the tumour as possible. This resulted in a mild alleviation of symptoms reported by the patient.

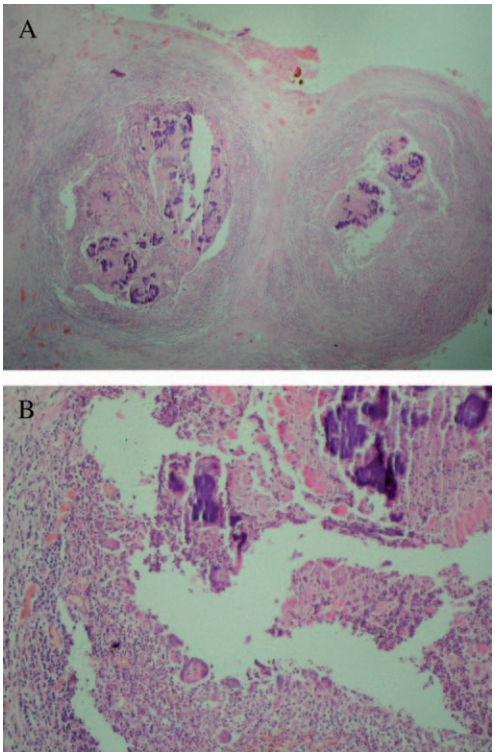

Figure 3. Histopathology of a tissue sample showing actinomycetoma formation surrounded by a heavy inflammatory infiltrate dominated by neutrophils (haematoxylin–eosin stain). (A) Overview (×20) and (B) detail (×250).

Key Points

- it was decided that the neurosurgeon goes ahead and tries to excise the lesion by craniotomy in an attempt to reduce pressure symptoms of headache and dimness of vision
- much to the surprise of the neurosurgeon, the craniotomy instruments broke during surgery, implying uncharacteristic thickness and hardness of the scalp and bone, findings that were later corroborated by imaging studies
- an attempt was made to remove as much of the tumour as possible. This resulted in a mild alleviation of symptoms reported by the patient

Key Points

- the MRI scan shows a severe involvement of CNS structures including an intracerebral sinus vein thrombosis and large brain abscess
- the patient had not sought treatment until very recently, but he was ina remarkably better clinical condition than suspected from the brain images
- the clue to diagnosis was the combination of a history of the trauma and the histopathology
- microbial and mycological cultures were used to substantiate the diagnostic accuracy
- regular treatment of actinomycosis of the CNS includes a combination of brain surgery and antibiotic treatment as performed in the present case
- intravenous antibiosis is recommended at least for thoracic actinomycosis for 2–6 weeks followed by 6–12 months of oral antibiotic therapy
- our patient was unable to follow our recommendations of i.v. antibiosis; oral therapy is ongoin
- another recently available therapeutic option for such CNS infections is linezolid, an oxazolidone with bacteriostatic activity against virtually all gram-positive bacteria

The patient was advised to take injectable penicillin but could not come from his home to the hospital on a regular basis so an option of cotrimoxazole and trimethoprim 800 + 160 mg was exercised. The patient has been tolerating treatment well. Headaches are significantly reduced and the vision is less blurred according to the patient. However, the little pus-exuding nodules continued to grow on the scalp, albeit at less frequent duration. Clavulanate-potentiated amoxicillin 675 mg twice a day has been instituted for the past 2 months without any further neurological improvement. The discharging nodules have been reduced in number and frequency.

DISCUSSION

Penetrating head injury is a very rare cause of actinomycosis of the CNS (5). If correctly diagnosed and properly treated, the disease might have a favourable prognosis in contrast to other CNS infections (6,7). In contrast, infections by *Nocardia* spp. are often induced by direct traumatic inoculation (8,9), inhalation, surgery (10) and eating contaminated food after a periodontal abscess (11).

In India, nocardiosis is an uncommon infection. In a recent study including 12 cases from India, 75% of patients were males and 11 patients had an underlying disease. Only three patients had CNS nocardiosis (12). *Nocardia* spp. infection was a differential diagnosis in our case. Because the organisms were acid-fast negative, *Actinomyces* spp.-induced mycetoma was suggested (1). Among the *Nocardia* spp., *Nocardia dossonvillei* also is acid-fast negative but further identification was impossible (1).

In the present case, the long delay from primary trauma, that is hurricane accident, to conclusive diagnosis and treatment is remarkable. The first noticeable clinical sign was the formation of mycetomas of cutaneous tissue resembling folliculitis keloidalis nuchae. The differential diagnosis was a dissecting folliculitis or deep scalp mycosis.

We assume that bone involvement secondarily led to brain affection. The MRI scan shows a severe involvement of CNS structures including an intracerebral sinus vein thrombosis and large brain abscess. Despite the patient had not sought treatment until very recently, he was in a remarkably better clinical condition than suspected from the brain images.

The clue to diagnosis was the combination of a history of the trauma and the histopathology. Microbial and mycological cultures were used to substantiate the diagnostic accuracy. Recently, tsunami survivors were seen with atypical skin infections (13,14). Hurricane victims might be in a comparable situation with multiple traumas, exposing the organism to various microbes of the soil (15).

Regular treatment of actinomycosis of the CNS includes a combination of brain surgery and antibiotic treatment as performed in the present case. Intravenous antibiosis is recommended at least for thoracic actinomycosis for 2–6 weeks followed by 6–12 months of oral antibiotic therapy. Penicillin, ampicillin, amoxicillin–clavulanate, doxycycline, moxifloxacin and clindamycin are used for thoracic and oral diseases (16,17). Our patient was unable to follow our recommendations of i.v. antibiosis. Oral therapy is ongoing.

Regular treatment of nocardiosis consists of trimethoprim–sulphamethoxazole, imipenem, dapsone and/or amikain (18–20). Patients who fail to respond to the standard regimens may benefit from amoxicillin–clavulanate. In a recent study from Mexico, 15 of 21 cases showed a complete clinical and microbial cure, 2 improved and only 4 failed (9). Choosing trimethoprim–sulphamethoxazole followed by amoxicillin–clavulanate, the major families of the order *Actinomycetaceae* were covered.

Another recently available therapeutic option for such CNS infections is linezolid, an oxazolidone with bacteriostatic activity against virtually all gram-positive bacteria. It shows a good liquor penetration and few side-effects (21).

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