CASE REPORT
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Spontaneous Cerebrospinal Fluid Rhinorrhea with Pneumocephalus: An Unusual Manifestation of Nasal Tuberculosis

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An unusual case of spontaneous cerebrospinal fluid (CSF) rhinorrhea with a pneumocephalus is described in a middle-aged woman who presented with a watery nasal discharge for 1 week and headache, vomiting, and fever for 1 day. The neurological examination revealed meningeal signs and bilateral papilledema. The CSF picture suggested pyogenic meningitis, and computed tomography of the brain revealed pneumocephalus. Diagnostic nasal endoscopy showed outpouching of the dura from the left olfactory cleft with a CSF leak and granular nasal mucosa. The defect was repaired surgically, and a biopsy of that area revealed granulomatous changes suggestive of tuberculosis. The patient recovered completely with standard four-drug antitubercular therapy. To our knowledge spontaneous CSF rhinorrhea with pneumocephalus occurring secondary to nasal tuberculosis has not been previously reported.

Keywords: Cerebrospinal fluid rhinorrhea; Pneumocephalus; Tuberculosis

INTRODUCTION

Cerebrospinal fluid (CSF) rhinorrhea can result from head trauma, intracranial surgery, or destructive lesions, and pneumocephalus is a consequence of connection between the intra- and extracranial spaces. Granulomatous diseases like tuberculosis and histoplasmosis causing CSF rhinorrhea and pneumocephalus are very unusual. In countries such as India where tuberculosis is endemic, it is important to consider tuberculosis in the differential diagnosis of all granulomatous lesions. Nasal tuberculosis is a rare manifestation of *Mycobacterium tuberculosis* infection. Diagnosis requires a high index of suspicion and treatment is anti-tubercular therapy.

CASE REPORT

A 51-year-old woman presented with a 7-day history of watery nasal discharge and headache, vomiting, and fever for 1 day. There was no history of head injury. Her pulse was 80/min, and blood pressure was 140/90 mmHg. She was conscious and oriented at presentation. She had signs of meningeal irritation and bilateral papilledema. Her laboratory parameters showed hemoglobin 13 g/dL, white blood cell (WBC) count 13,500/mm^3 (neutrophils 88%, lymphocytes 12%), erythrocyte sedimentation rate (ESR) 90 mm/hr, and glucose 141 mg/dL (7.83 mmol/L). The Mantoux test was strongly positive (> 14 mm). Renal function tests, electrolytes, urinalysis, and a chest X-ray

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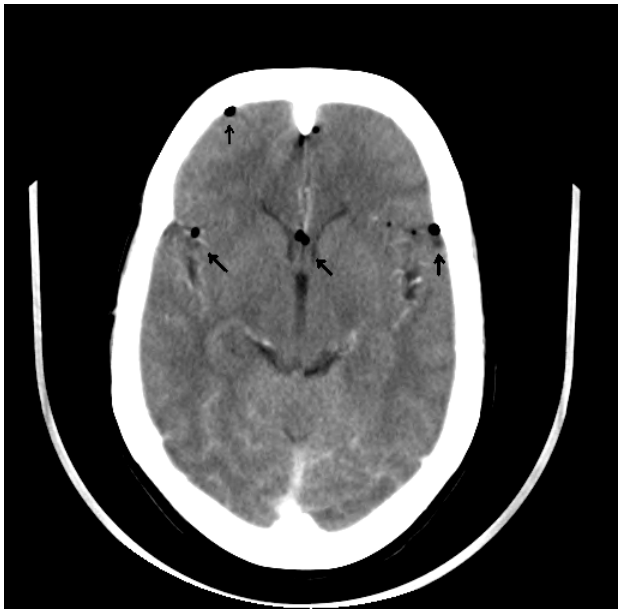

Figure 1. Axial brain computed tomography showing meningeal enhancement and air in the subarachnoid space suggestive of pneumocephalus.

were normal. The vasculitis workup was normal (negative antineutrophil cytoplasmic autoantibody [ANCA] and antinuclear antibody). Contrast-enhanced computed tomography (CT) of the brain revealed meningeal enhancement with pneumocephalus (Fig. 1). CT of the osteomeatal complex did not show any fractures, bony defects, or destructive lesions. A diagnostic lumbar puncture was performed; the CSF analysis revealed glucose 10 mg/dL (0.56 mmol/L), protein 139 mg/dL, and WBC count 550/mm^3 (neutrophils 92%, lymphocytes 8%). Gram's stain showed Gram-positive cocci in pairs, and the culture grew pneumococci. Ceftriaxone was started intravenously. Over the next 3 days, her clinical improvement was dramatic; her neck stiffness resolved within 1 week. Diagnostic nasal endoscopy revealed outpouching of the dura from the left olfactory cleft, with a CSF leak and granular nasal mucosa. A necrotic bony area was noted around the olfactory cleft. After 1 month, she underwent endoscopic repair of the defect. Outpouching of dura from the left olfactory cleft was noted intra-operatively, and the CSF leak was confirmed on hyperventilation. The area was curetted, cauterized, and packed with (Shree Balaji Surgical Pvt. Ltd., Mumbai, India), Gelfoam (General Surgical Co. Pvt. Ltd., Chennai, India), abdominal fat, and bismuth iodoform paraffin paste packs. Postoperatively, there was no CSF leak, which was confirmed at repeat endoscopy. Histopathologically, the intra-operative biopsy revealed granulomatous changes suggestive of tuberculosis (Fig. 2). She was started on standard four-drug antituberculosis therapy (ethambutol, isoniazid, rifampicin, and pyrazinamide). The specimen was inadequate for cultures.

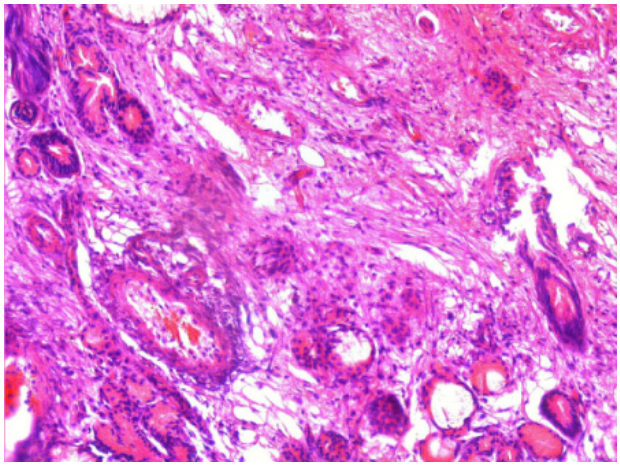

Figure 2. Photomicrograph showings nasal epithelium with dense stromal infiltration with plasma cells and lymphocytes, forming granulomatous lesions (H&E, × 100).

She recovered completely with 6 months of therapy. There were no further episodes of CSF rhinorrhea, and the ESR had normalized at the 1-year follow-up.

DISCUSSION

CSF rhinorrhea commonly follows head trauma, intracranial surgery, or destructive lesions [1]. There is disruption of the arachnoid and dura, coupled with an osseous defect and a CSF pressure gradient that is either continuously or intermittently greater than the healing tensile strength of the disrupted tissue. This causes separation of the dural fibers and CSF leakage. Most fistulas heal with conservative management and without complications, but pneumocephalus, meningitis, and hydrocephalus are potential fatal complications [1,2].

Pneumocephalus is an accumulation of intracranial air that occurs when there is a connection between the intra- and extracranial spaces. Common causes of pneumocephalus include trauma, surgery, and neoplasm; cases occasionally occur secondary to nasogastric tube insertion, bag-valve mask ventilation, nasotracheal intubation, lumbar puncture, otitis media, post-radiation necrosis, and CSF infection with gas-producing or unidentified organisms [2,3]. There are also reports of cases following meningitis, pneumosinus dilatans, and nose blowing [3-5].

Granulomatous diseases like tuberculosis and histoplasmosis causing CSF rhinorrhea and pneumocephalus have not been reported.

Nasal tuberculosis is a rare manifestation of *M. tuberculosis* infection. It is most common in women older than 20 years [6]. Clinically, it presents as a nasopharyngeal mass or granulomas, often with concomitant enlarged lymph nodes; other presentations include nasal obstruction, epistaxis, and fever. In our case, bony erosion due to tuberculosis led to outpouching of the dura, a CSF leak, and subsequent bacterial meningitis.

In countries such as India where tuberculosis is endemic, it is important to consider tuberculosis in the differential diagnosis of all granulomatous lesions. The diagnosis is based on histopathological studies. Treatment with standard anti-tuberculous chemotherapy is satisfactory [6,7].

The diagnosis of tuberculosis in our case was based on the history, high ESR, positive Mantoux test, granulomatous nasal changes, histopathological picture, and clinical response to anti-tuberculosis therapy. The differential diagnosis in our case included Wegener's granulomatous; however, this was unlikely in our case because the ANCA was negative, the condition improved without any immunosuppressive therapy, and the patient was asymptomatic at the 1-year follow-up.

Repair of the defect causing the CSF leak is generally indicated if the leakage persists or if symptomatic pneumocephalus supervenes. This can be achieved either from below with transsphenoidal surgery or from above via a craniotomy [8,9]. Materials such as muscle, fascia, pericranium, or Tisseel glue may be used to patch the defect. The treatment is simple, and the prognosis is excellent [9].

The presented case shows one of the diverse manifestations of tuberculosis, a common disease in countries such as India, where tuberculosis is endemic. A high index of suspicion is required to make the diagnosis.

Conflict of interest

No potential conflict of interest relevant to this article is reported.

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