Short Communication

An Intracranial Tuberculoma with Discharging Scalp Sinus

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SUMMARY: Our report highlights a case of intracranial tuberculoma in an elderly woman with chronic discharging sinus. A 77-year-old woman had a mass lesion extending to the scalp through a chronic discharging sinus for 2 years with an intermittent, non-radiating, dull, low-grade headache. Based on our survey of central nervous system tuberculosis, this case is a rare event of documented intracranial tuberculoma with concomitant discharge of the scalp sinus similar to the periodic geyser eruptions in developing countries.

Central nervous system (CNS) tuberculosis (TB) is a rare and severe form of TB due to *Mycobacterium tuberculosis* (MT) (1). It can manifest as meningitis, abscess, tuberculoma, and calvarial osteomyelitis. Intracranial tuberculomas constitute 1% of all CNS TB cases (2). Tuberculoma with a concomitant discharge of the scalp sinus is a rare presentation.

This 77-year-old woman presented with right frontal intermittent, non-radiating, dull, low-grade headache that had been bothering her for years. She also noticed a right frontal scalp discharging sinus. After each discharge, her symptoms decreased and completely subsided in 2–3 days; without any treatment. The symptomatology was periodical, recurring once every 20–30 days. She became drowsy one day before her admission to our intensive care unit. On admission, she had left hemiplegia, whereas the remaining neurologic and systemic examinations were unremarkable. Her past medical and surgical histories were also unremarkable, with no evidence of pulmonary TB. Chest computed tomography (CT) and magnetic resonance imaging (MRI) (Fig. 1) were performed the following days. Chest CT did not reveal fibrosis, collapse, or cavitation.

Preoperative findings: The right frontal scalp showed a 2 × 2 mm² thick-walled sinus with profuse cheesy discharge. On elevating the frontal flap, a tract was seen extending from the sinus into the frontal bone. The bone was thick and looked unhealthy. A small defect of 2 × 2

mm² was also observed. Right frontal craniotomy was performed. The underlying dura showed a similar defect with thickening and adhesion to the frontal cortex. It was full and bulging with inflammatory infiltrate. The tract was then followed into the mass lesion. The central part of the mass lesion was soft, suckable, cheesy white, thin in consistency, and vascular. The lesion had a well-defined, thick wall and a clear interface, separating it from the surrounding healthy brain. It was meticulously dissected to avoid opening it into the ventricle. After a complete resection without sucking the gliosis layer around the lesion, the brain was lax and pulsatile. The unhealthy dura was excised and closed using a pericranial graft from a healthy portion. The involved bone was nibbled, and the remaining healthy bone was replaced. The sinus tract was excised, and the skin was closed in the usual fashion. The lesion was sent for acid fast bacilli (AFB) staining and a histopathological examination (Fig. 2).

The postoperative course was uneventful. Based on the histopathology report, the patient was started on anti-TB therapy. At follow-up, performed 18 months later, she was able to pursue activities of daily living independently. CT showed no further recurrence.

CNS-TB is uncommon, accounting for only 10% of all cases of tuberculosis (3). Most tuberculosis infections are caused by *Mycobacterium tuberculosis*. The bacilli reach the CNS through the hematogenous route secondary to disease in other parts of the body. The original description by Rich suggested a two-stage development of CNS tuberculosis. First, the tuberculous lesions (rich in focus) develop in the brain during the stage of bacteremia. Second, the rupture or growth of one or more of these lesions causes the development of CNS tuberculosis.

The rare presentation of intracranial tuberculomas constitutes 1% of all CNS TB and is usually seen in young patients and adults with uncontrolled diabetes

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Fig. 1. (Color online) Preoperative CT. A: Bone windows showing bony destruction with a small defect. B: Obvious bony defect and the sinus tract. C: Brain windows showing bony destruction with a defect. D: Follow up scan after 2 years, showing no recurrence. Preoperative MRI E T1WI showing a right frontal solitary thick-walled hypointense lesion. F: T2WI showing a hyperintense lesion with significant perilesional edema and midline shift. G: T1WI with gadolinium contrast showing heterogeneous wall enhancement. H: DWI showing the tumor lesion.

Fig. 2. (Color online) A: Histopathology (low power) of tuberculoma specimen showing central caseous necrosis (white arrowheads) surrounded by Langerhans's giant cells with a peripheral mantle of lymphocytes. B: Acid Fast Bacilli (AFB) staining of cheesy material showing pink acid-fast bacteria (black arrowheads).

and immunosuppression (4–8). In 15%–33% of cases, tuberculomas can be multiple (9). Tuberculomas are firm, avascular, and spherical, with sizes varying between 2 and 10 cm in diameter. They are well-circumscribed and compress the brain tissue, which shows edema and gliosis. The inside of the masses contains necrotic areas of caseation in which tubercle bacilli may be found.

Intracranial tuberculoma with intracranial communication from the scalp and with a chronic discharging sinus is a rare event in developing countries (10). The first case of scalp tuberculosis with bony involvement was reported in 1842 by Reid from Germany (11). This case is presented here because the patient's presentation is similar to periodic geyser eruptions. We think that such an interesting clinical phenomenon has not been reported before. How can we explain its genesis? Usually, in CNS-TB, one can show a discharging sinus with calvarial osteomyelitis,

but a discharging sinus associated with an intracranial tuberculoma is rare. We presume that the association could be due to the long disease period (2 years), lack of treatment, and the patient's (old) age. The symptomatology can be explained as follows (Fig. 3): The increase in the volume of the caseous material in the tuberculoma resulted in its enlargement and stretched its capsular wall. Then, it broke the wall of the tuberculoma, and the tubercle bacilli spilled into the overlying tissues. When the pressure was high enough, these bacilli, in turn, seeded the dura and caused local bone destruction. Over time, the granulation tissue tried to find its way to the scalp tissue through the least resistant track. As the contents were discharged through the sinus, the intra-tuberculoma pressure decreased, and her symptoms were alleviated. The contents of the tuberculoma would then be refilled with caseous material. This cycle repeated itself, causing waxing and waning of her symptoms. Although the mechanism

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Fig. 3. (Color online) Mechanism of recurring cycles of waxing waning symptomatology, with the operative sketch.

is known to be immunological, the specific stimulus needed for the rupture and growth of these lesions is still not completely understood. Cytokine tumor necrosis factor- α (TNF- α), which is critical in the neuropathogenesis of *M. tuberculosis* and alteration of blood-brain barrier (BBB) permeability (8), is probably involved in this process. As the disease progresses (chronically), an increase in the size of the tuberculoma and the perilesional edema leads to high intracranial pressure (ICP) and compresses the corticospinal tracts on the ipsilateral side. The rise of ICP results in headache, nausea, vomiting, drowsiness, and altered mentation, while compression of the ipsilateral corticospinal tracts results in contralateral hemiparesis (12). These grave symptoms finally prompted the patient's relatives to seek medical attention.

Since neurological symptoms can masquerade as a malignant glioma metastasis, a high level of suspicion is required to make a diagnosis. A positive Mantoux test or an elevated erythrocyte sedimentation rate is suggestive but not the diagnosis of tuberculosis. Unfortunately, such cases are found in less than 10% of patients. A history of TB/contact with TB also gives us clues to the diagnosis. CT of the brain shows an irregular, thick-walled, iso to hyperdense, heterogeneously enhancing lesion (13). They may be solitary or multiple and have a preference to occur in the frontal and parietal lobes. The radiographic appearance of tuberculomas is thought to depend on whether the lesion is noncaseating, caseating with a solid center, or caseating with a liquid center. The so-called "target sign," a central nidus of calcification surrounded by a ring of enhancement,

was once thought to be a diagnostic feature of a tuberculoma but is now known to be nonspecific. Brain MRI shows T2 shortening (hypointensity, due to the thick-walled mycobacteria, which contain mycolic acid and glycoarabinomannan) with conglomeration of multiple lesions and heterogeneous enhancement (14,15). Definitive diagnosis can only be made by histopathology, showing caseous granuloma with Langerhans' giant cells and a peripheral mantle of lymphocytes.

The mainstay of therapy is surgical excision when there is an impending herniation and antitubercular therapy. ESR and serial CT scans were used to evaluate recurrence.

Intracranial tuberculoma can have unusual presentations, and this case is a testament to it. A very high index of suspicion is needed, even in developing countries, to prevent delayed diagnosis and treatment of intracranial tuberculoma. CT and MRI are essential to make a differential diagnosis; however, histopathology is a *sine qua non* for definitive diagnosis. Surgical resection followed by antitubercular therapy is the cornerstone for achieving a complete cure.

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Conflict of interest None to declare.

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