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Meningeal Actinomyces Mimicking Meningioma, Case Report

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ABSTRACT

Background: Human actinomycosis, a chronic, granulomatous infectious disease, has been recognized for a long time. Actinomyces sp. Is a commensal, filamentous, gram-positive, acid-fast-negative bacteria that can cause pyogranulomatous inflammation in animals and humans. In Central Nervous System (CNS) affection is a rare presentation disease and is usually due to extension from infected lesions or seeding from distant sites. Histologically, is characterized by a severe pyogranulomatous meningoencephalitis with intralesional filamentous bacteria that were also visible on cytology of the Cerebral Spinal Fluid (CSF).

Case Presentation: We present a rare case report of meningeal actinomyces mimicking to invasive meningioma in a 58-year-old man, a healthy farmer with no significant history, who began one month earlier with decreased visual acuity in the left eye, disorientation was found, and after with decreased MSD strength, progressive stabbing headache, and bilateral amaurosis. The MRI shows a left occipital dural lesion with a diagnosis of aggressive infiltrating meningioma. He underwent resection of the lesion that was characterized by a yellowish white granular nodule, the meninges show diffuse micronodules. Histologically, a chronic granulomatous lesion with actinomyces is identified. The patient is discharged with antibiotics improving his condition. Infection at other sites was ruled out.

Discussion: We present a case of a patient with rapidly progressive symptoms and a lesion suggestive of a meningioma.

Keywords: Meningioma; Cerebral pseudotumor mimicking meningioma; Cerebral actinomyces

BACKGROUND

Actinomyces species are gram-positive, facultative anaerobic bacilli, it an uncommon infectious disease caused predominantly by *Actinomyces Israelii*. Inside the tissue, these bacteria form masses consisting of aggregates of branching, filamentous bacilli.[1] Actinomycosis usually involves the cervicofacial, thoracic, abdominal, and pelvic region, dissemination is uncommon.[1] The cervicofacial area is the most common site of involvement, and affection and the affection of the CNS is rare, presenting a mortality up to 28%.[2] Its manifestations could be similar to infectious conditions from other etiologies, It is well established that actinomycosis is an endogenous infection and cause actinomycosis in immunodeficiency patients.[3] Moreover, they should know that actinomycosis may mimic the process of malignancy at various anatomical locations as well as abscesses.[4] Osteomyelitis and bacterial meningoencephalitis by Actinomyces are rare and usually occurs at the mandibular level, causing by this bacterial specie, especially after traumatic brain injury.[5, 6] Non-neutrophilic pleocytosis has been found on CSF analysis.[3] A varied range of Actinomyces species are being increasingly associated with infections at many body sites.[6,7] The presence of "sulfur granules" at the infection sites is a typical histopathological change, which often contains abscesses with yellowish sulfur-like granules.[6] Thus, histopathological examination of infected tissue is usually a added sensitive technique compared to bacterial culture, possibly revealing typical yellowish sulfur granules containing filamentous Gram-positive bacilli and inflammatory cells.[6]

The aim of this paper was reported a rare case of meningeal actinomyces that mimicking a meningioma.

CASE PRESENTATION

58-year-old man, farmer, with a history of high blood pressure for 3 years, treated, controlled, asymptomatic. Onset with visual disturbances, decreased visual acuity of the left eye until reaching bilateral amaurosis, one month of evolution, later presents non-dense pyramidal syndrome, with progressive stabbing headache and physical examination shows bilateral papilledema Frinsen grade II. MRI Imaging studies were performed and showed a 4cm dural occipital lobe lesion that was diagnosed as a meningioma (Figure 1a and 1b), and subsequently underwent complete surgical resection of the lesion (Figure 1c, 1d and 1e). The angio-MRI showed hypervascularity lesion (Figure 1f). The gross up, the lesion was nodular, measuring 4x4x4cm. Whitish with a soft myxoid appearance. Histologically, meninges and bone with dense mixed inflammatory infiltrate with granulomatous formation and giant multinucleated cells were identified. Lesions resembling grains of sulfur are observed (Figure 2a and 2b), which were PAS Positive (Figure 2c), also observing a large number of foamy macrophages and loaded with a positive PAS content (Figure 2d). were noticed. Sulfur granules seen with Grocott positive stain (Figure 2e) were also observed in macrophages (Figure 2f). Numerous blood vessels with concentric fibrosis and endothelial hyperplasia are observed (Figure 2g) also vessels with central concentric fibrous reaction were demonstrated with Masson's stain (Figure 2h). Based on these findings, the diagnosis of meningeal actinomyces was made.

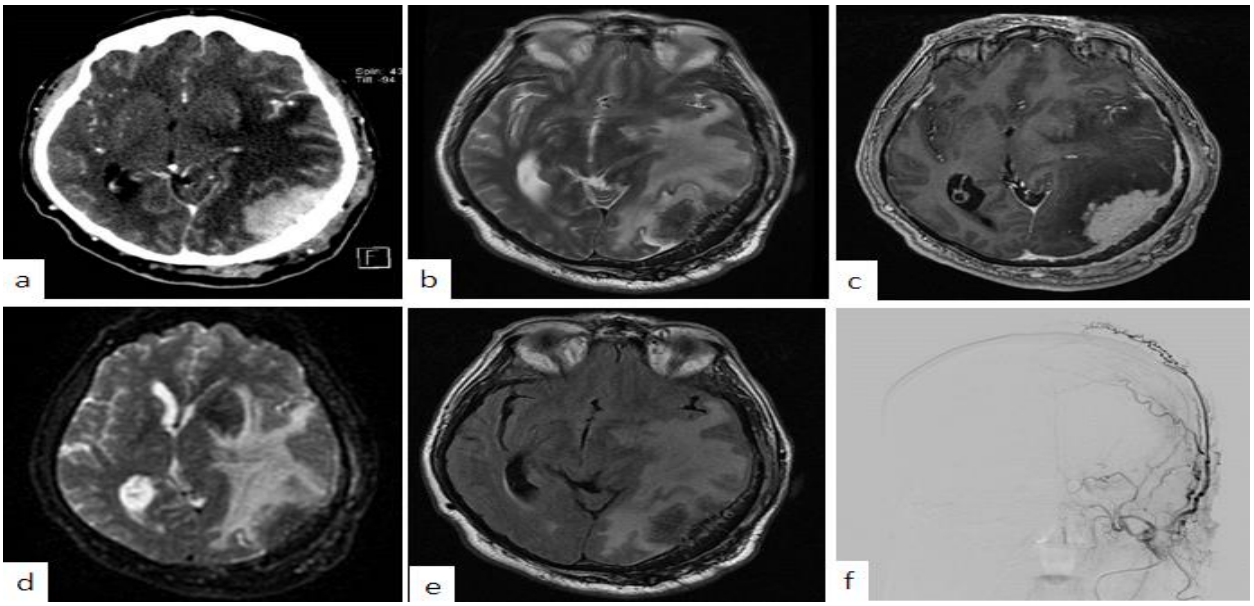

Figure 1: Radiological images. (a) Brain CT showing an hyperdense mass within the left parietal lobe cortex and subcortical white matter with homogeneous contrast enhancement. There is vasogenic edema with left-to-right midline shift. (b)The MRI showed a T2 coronal imaging that showed a temporoparietal lesion. (c) FSPGR+C and (d) FSPGR and (e) DWI weighted MRI showing a mass within the left parietal lobe with homogeneous contrast enhancement and pachymeningeal involvement. Diffusion-weighted MRI shows vasogenic edema that involves the frontal, parietal and occipital lobes and (f) angio-TAC showed a hypervascularity area in occipital lobe.

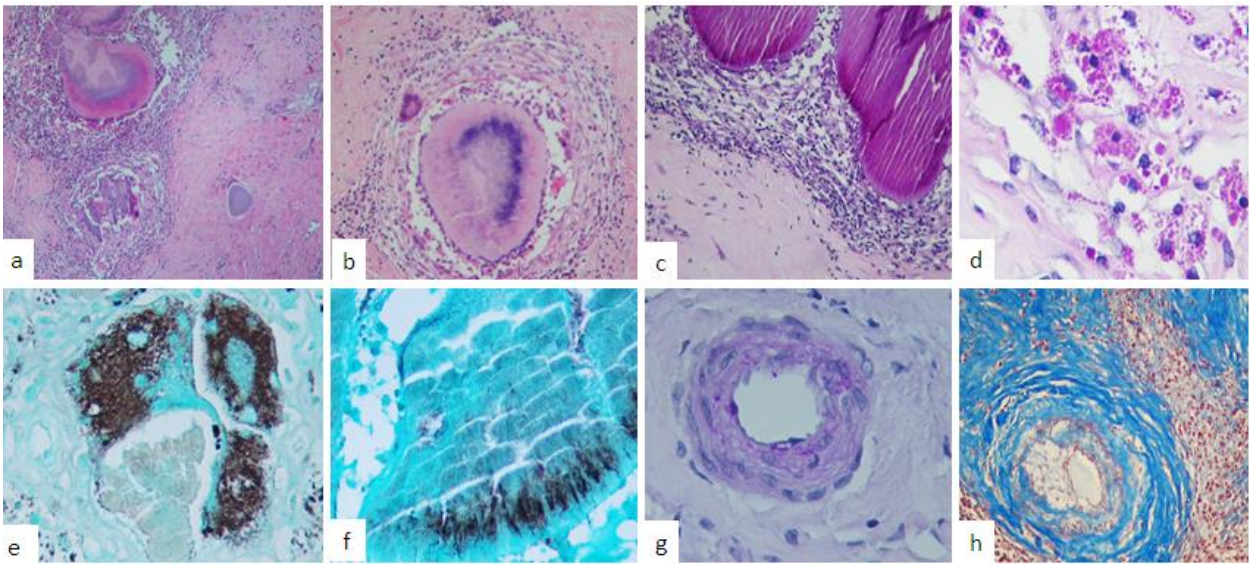

Figure 2: Histological photos. (a) shows a panoramic view of the lesion, observed Cluster of actinomyces with grains of sulfur in standard stain, surrounded by inflammatory cells (H&E x10), (b) observed a close-up of a sulfur granule with inflammatory reaction and multinuclear giant cells (H&Ex400), (c) with PAS staining observing intensely positive these lesions delimited by inflammatory cells, (d) abundant macrophages with PAS positive material (x400) were also observed. (e) and (f) showed the expression of Grocott positive staining in the sulfur

granules that show crescents radial filaments (x400) and (g) PAS stain showed thickened vessels and concentric aspect and in (h) observed an area of fibrosis with hyperplastic vessels with concentric distribution that are intensely blue with the Masson stain (x400).

DISCUSSION

Actinomycosis is caused by filamentous Gram-positive anaerobic bacteria that is a normal commensal but causes suppurative and granulomatous inflammation,[3] after disruption of anatomical barriers an uncommon chronic suppurative infection that rarely affects the CNS.[5-7] Actinomycotic brain abscess is a rare condition with uncertain clinical features, but it is more common than meningeal involvement, which forms granuloma,[7] that causes mass effect, it is solid and radiologically resembles meningioma.4 As in our case .

Here we report the case of a 58-year-old non-immune-competent man with an actinomycotic meningeal tumor who presented with sensory aphasia and mild right hemiparesis. She had no febrile episode or headache. Moreover, he did not have any periodontal or otorhino-laryngological neither dermatological disease, nor the results of laboratory tests were normal. A computed tomography scan showed an irregular, low-density area in the left parietal lobe. MRI showed low-signal intensity in a T1 weighted image, high-signal intensity in a T2 weighted image, and mixed intensity on a diffusion weighted image. Thallium-201 chloride scintigraphy showed definite accumulation of thallium in the lesion and the patient's condition gradually deteriorated. Ten days after gadolinium administration, a T1 weighted image showed a multi- lobulated irregular mass in the left parietal lobe. The patient subsequently underwent craniotomy and evacuation of meningeal tumor.

Kommedal et al.[6] showed that 4 to 10 species were found in specimens from 12 spontaneous brain abscesses with an involvement of A. meyeri. Ravindra N, et al.[5] reported a series of 17 histologically confirmed cases of CNS actinomycosis and review clinical, imaging, and histopathologic features mean aged at presentation was 31.4 years, with male predilection (3.25:1). Mean duration of symptoms was 2.95 months. Systemic symptoms were noted in 5 patients, although no systemic focus was detectable. Pachymeningitis was most common type (9; 52.94%), and chronic abscess was identified in 7 cases. History of previous surgery for osteomyelitis was forthcoming in 3 cases.[5] All patients underwent surgical excision/aspiration of the lesions. Histologically,[5] lesions revealed characteristic suppurative granulomatous response with giant cells and actinomycotic colonies were detected within necrotic centers. Cultures failed to grow Actinomyces in all. Follow-up data were available in 9 patients, and all had good outcomes at the median follow-up period of 32 months after antibiotic treatment for mean period of 8.4weeks. Histopathology remains the cornerstone for diagnosis of actinomycosis as on culture confirmation is very rare in Ravindra N, et. Serie.[5]

Vargas PJ et al,[8] reported SLE patients diagnosed and confined for a CNS infection, total of 23 SLE patients (22 females). Nineteen cases (82.6%) were meningitis, and four (17.4%) were diagnoses of brain abscess and just one with *Actinomyces spp*.

Neurological involvement includes a meningeal, granulomatous and also pseudo tumoral form. Intracerebral actinomycosis abscess have been described as a common form. Loftier J, et al.[7] reported a case with no history of immunodepression that was admitted with unexplained fever, inappropriate behavior, and spatial and temporal disorientation. Brain imaging showed multilocation cerebral abscesses. Sterotaxica biopsy permits diagnosis of actinomycosis.[6] We reported a rare case of a solid lesion that displaced the brain and suggested a meningioma-type tumor. Histologically, we observed the formation of granulomas with giant cells and also an unusual angiogenic response in actinomycetes, which were large vessels with concentric fibrous reaction. The etiopathogenesis of the changes and the clinical-morphological correlations of this very rare infectious disease of the SNC has not been specifically described in this infection.[8]

Vascular changes as hyperplasia of muscular layer or endarteritis reaction are not common findings described in association to actinomyces. This case is interesting due to the granulomatous reaction associated with vascular lesions rarely described in this disease, and which could be a contribution and suggests the intense inflammatory response that leads to fibrosis and vascular damage.

Neuropathological examination revealed both in the cortex and white matter the presence of numerous disseminates bodies staining intensively with H&E, PAS- and Gram-positive stain and in the ZN negative method. Their structure, compact or fine-powdered corresponded to actinomyces colonies. They either showed no reaction or were surrounded by a wall of inflammatory cells such as: neutrophilic leucocytes, lymphocytes, plasmatic cells and histiocytes. The micro abscesses were not encapsulated.[8] It is an invasive bacterial disease and usually characterized by the formation of granulomatous tissue, necrosis and major reactive fibrosis, draining sinuses, abscesses, which is an indolent progressing granulomatous disease. This case was classified to a granulomatous form of brain actinomycosis.

Persistent neutrophilic meningitis is a poorly described variant of chronic meningitis characterized by the persistence of neutrophils in the CSF over extended periods of time (greater than 1 week) in association with ongoing signs of meningeal inflammation and negative CSF cultures for bacteria and other pathogens. Etiologies of this syndrome are both infectious and noninfectious.[7] Among infectious causes, bacteria such as Nocardia and Actinomyces and systemic mycoses such as Aspergillus and the zygomycetes are the predominant pathogens.[1] Although the incidence of persistent neutrophilic meningitis is difficult to ascertain, a review of available literature on CNS infections suggests that this entity is not rare.[8,9]

CONCUSSION

We present a rare case of actinomyces in a solid form that suggested meningioma in a patient without immunosuppression with granuloma formation and with secondary endarteritis and vascular changes with a clinical

evolution of one month. The important difference between actinomycosis and nocardiosis has been always discussed.

ABBREVIATIONS

CD: Cluster of Differentiation; CT: Computed Tomography; DFSP: Dermatofibrosarcoma Protuberans. H&E, PAS- and Gram-positive stain and in the Ziehl-Nielsen

DATA SHARING STATEMENT

The data utilized and/or analyzed during this case report are available from the corresponding author on reasonable request.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

No institutional endorsement was required to publish the case details. The parents of the child gave informed written consent to participate in the study of their child's condition.

CONSENT FOR PUBLICATION

The parents of the child gave informed written consent for this case to be published in a peer-reviewed journal.

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AUTHOR CONTRIBUTIONS

All authors made a crucial contribution to the work reported, whether that's within the beginning, study project, execution, acquisition of data, analysis, and interpretation, or in all these areas; took part in drafting, revising, or critically studying the article; gave final approval of the version to be published; have concurred on the journal to which the article has been submitted; and agree to be answerable for all angles of the work.

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DISCLOSURE

The authors announce that they have no competing interests.

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