Trigeminal Neuralgia Caused by Cysticercosis of the Cavum Meckelii

Report of Two Cases

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The differential diagnosis between primary and secondary trigeminal neuralgia often has considerable practical importance because of its therapeutic implications. However, in cases in which there is a known etiological agent, a secondary neuralgia may simulate clinically a primary form of this condition. Such event occurred in our 2 cases, and in itself constitutes one of the reasons for this report. In the cases under discussion, prosopalgiæ with the characteristics of a primary trigeminal neuralgia were caused by cysticerci located in the cavum meckelii. To our knowledge the medical literature does not record any previous report on cysticercosis of the cavum meckelii. A bibliographical survey was done in the current indices, and several original articles on neurocysticercosis were reviewed, covering a total of 1,190 cases (clinical observations, clinicopathological or pathological reports only), and no references were found on paroxysmal trigeminal neuralgia not associated with other focal neurological manifestations.

Case Summaries

Case 1. Reg. HC 567430. I.M.F., a 31-year-old white female, had suffered periodic vomiting, headache, and temporary loss of vision for 10 years. Paroxysmal pain in the territory of the 3rd branch of the left trigeminal nerve had been present for 7 months prior to admission. She had had infestation by tapeworms since infancy and frequently had ingested undercooked pork.

Examination. Physical and neurological findings were normal. Cerebrospinal fluid (cisternal): initial pressure was 45 cm. of water; final pressure was 11 cm. (10 cc. of fluid withdrawn). The fluid contained 125 cells/c.mm. (lymphocytes 71 per cent, monocytes 18 per cent, eosinophils 8 per cent); total proteins were 30 mg. per cent; chlorides 700 mg. per cent; sugar 53 mg. per cent; Pandy test and Takata-Ara colloidal reaction were positive; Wassermann test was negative and complement fixation test for cysticercosis was positive. Ocular fundi showed papilledema in the right eye and a blurred nasal border in the left eye. Roentgenograms of the skull disclosed no intracranial calcifications or signs of hypertension. Electroencephalogram showed alteration of the alpha rhythm in the occipital areas, with 4 to 7 c./sec. waves. Peripheral blood contained 11 per cent eosinophils. Complement fixation test for cysticercosis in the blood serum was positive.

Course. The patient was treated with sulfa drugs, Prednisone, vitamins B1 and B6, hydantoin and analgesics. The clinical diagnosis was of a hypertensive form of neurocysticercosis, and of primary trigeminal neuralgia. There was no improvement of the neuralgia, and the therapy was reevaluated.

Operation. The cavum meckelii was explored by the technique of Frazier and Spiller, and a cyst was found to be adherent to the Gasserian ganglion and roots (Fig. 1). The cyst was removed together with the sensory roots (Fig. 2).

Pathological Diagnosis. Cysticercus cellulosae (Fig. 3). Course. Postoperatively there was total disappearance of the neuralgia, and analgesia in the territory of distribution of the left trigeminal nerve.

Five months later, the symptomatology of intracranial hypertension became more manifest, and a ventriculogram demonstrated a ventriculocisternal block. Surgical opening of the lamina terminalis then was performed, and the patient was discharged without any further developments. Two and a half years after the first admission the patient was reexamined and was well.

Case 2. Reg. HC 619296. M.M., a 38-year-old white male, had lightning pains in the territory of the 2nd

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Fig. 1. Case 1. Schematic representation of the cyst at operation, embedded in the Gasserian ganglion.

Fig. 2. Case 1. Operative specimen, including the cyst and its bed, formed by the posterior roots of the trigeminal nerve.
nerve involvement in cysticercosis, referred to the existence of trigeminal signs or symptoms in 12 cases, but an analysis of these cases reveals a constant association with other neurologic manifestations.

Briceño et al., in a review of the autopsy findings in 97 cases of neurocysticercosis, did not refer to localization of the parasite in the cavum meckelii, despite having 22 cases of localization of parasites in the middle cranial fossa.

Arriagada et al., in a careful study of cranial-nerve involvement in cysticercosis, did not refer to clinical forms comparable to ours. Arriagada et al. reported 2 cases of irritative signs of the trigeminal nerve (pain or dysesthesia), appearing late in the course of the disease, one of which was a case of intracranial hypertension.

Involvement of the trigeminal nerve may occur in cases of cysticercosis of the posterior fossa, particularly in those with a syndrome of the cerebellopontine angle, a rather rare event.

In our review of the literature, we were unable to find any reference to paroxysmal trigeminal neuralgia without other focal neurologic signs. In our 2 cases the etiology and topography of the lesion were documented surgically. In Case 1 the cysticercosis was manifested by intracranial hypertension, and the only focal sign was the trigeminal neuralgia; the parasitic etiology for this neuralgia was suspected prior to surgery. In Case 2 a pre-operative diagnosis of primary trigeminal neuralgia was made.

Summary

The authors report 2 cases of trigeminal neuralgia, secondary to cysticercosis of the cavum meckelii. In 1 of the cases, despite the primary features of the clinical manifestations, an etiology of cysticercosis was suspected prior to operation because of the presence of other signs of neurocysticercosis. In the other case, cysticercosis of the cavum meckelii was a purely surgical finding. Both patients manifested postoperative clinical improvement. In Case 1, analgesia in the territory of the

Fig. 3. Case 1. Photomicrograph showing the walls of the Cysticercus cellulosae (×9).

branch of the right trigeminal nerve for 3 years, with progressive increase in frequency. Past history was noncontributory.

Examination. Physical and neurological findings were normal on admission. Results of laboratory tests also were noncontributory. A diagnosis of primary trigeminal neuralgia was made.

Course. The treatment consisted of vitamins B1 and B12, associated with hydantoinates. No clinical improvement was observed.

Operation. Surgical exploration of the right cavum meckelii, by the technique of Brazier and Spiller, was performed. A posterior rhizotomy had been planned, but a cystic structure was found upon the roots (Fig. 4). This structure was removed entirely, and the roots were left intact (Fig. 5).

Histological Report. The tissue removed demonstrated a necrotic Cysticercus and inflammatory reaction (Fig. 6).

Course. The neuralgia disappeared completely after the operation, and only a small area of hypalgesia persisted in the field of the 2nd branch of the right trigeminal nerve 12 days after operation. In view of the surgical findings, further laboratory and roentgen-ray examinations were made, failing completely to demonstrate evidence of active cysticercosis. The surgically removed cyst consisted most likely of a solitary manifestation of the disease.

Comments

Guccione stated that Wollemberg and Bittorf had included, among the irritative signs of the cranial nerves caused by cysticercosis, pain in the territory of the trigeminal nerve and episodes of trismus; and Biondi et al. had described, as signs of deficit, hypesthesia in the trigeminal field and reduction in the intensity of the corneopalpebral reflex. Ginsburg and Askanazy, referred to by Forui in his review of 356 reported cases, had mentioned compression of the trigeminal nerve at its emergence from the cranium. Brinck considered involvement of the trigeminal nerve very rare, and never had observed persistent deficit in the functions of this nerve; the neuralgic manifestations were considered as exceptionally rare.

Asenjo and Roeca, in an article on cranial-