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Intraventricular Haemorrhage and Hydrocephalus Caused by Intraventricular Parasitic Granuloma Suggesting Cerebral Sparganosis

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Summary

A 67-year-old female farmer presented with acute loss of consciousness. Computed tomograms showed a calcified mass in the cavum septi pellucidi with intraventricular haemorrhage and obstructive hydrocephalus. The patient became fully conscious after urgent external ventriculostomy and subsequently underwent craniotomy for the excision of the mass. Pathological examination of the mass demonstrated a granuloma surrounding a degenerating larva compatible with Spirometra mansonoides whose mimicking an intraventricular tumour has not been reported before.

Keywords: Cerebral sparganosis; granuloma; intraventricular haemorrhage; spirometra mansoni.

Introduction

Cases of cerebral sparganosis have been more common in Asia[3–10, 12, 15–18, 20] than elsewhere[2, 11, 13, 14]. Pathologically the disease is a granuloma induced by the plerocercoid larva of Spirometra mansonoides, a cestode tapeworm whose primary hosts are dogs and cats rather than humans.

Epilepsy was present in 12 of the 18 cases in a previous review; other symptoms included headache and focal neurological signs[9]. Patients most commonly had the lesion located in the convexity of the hemispheres and all made a good recovery after surgical removal of the lesion. Here we report a patient with a bleeding granuloma in the lateral ventricles, a previously unreported site for this parasite.

Case Report

A 67-year-old female farmer developed vomiting and unconsciousness while taking a bath. Two months prior to this admission the patient had an episode of right hemiparesis which resolved completely in a few days. The patient had been otherwise healthy and had not applied raw meat to wounds. Within 5 hours of her ictus, the patient was admitted to Chang Gung Memorial Hospital where she responded to painful stimulus by uttering incomprehensible sounds and keeping the eyes closed. Despite a mild weakness of all limbs, the patient localized painful stimuli. There were no cranial nerve deficits. Urgent computed tomography (CT) scans revealed a calcified intraventricular mass with haemorrhage and hydrocephalus (Fig. 1). A complete blood count showed no eosinophilia.

The patient was treated with immediate bilateral external ventriculostomy. She became fully conscious in one week and then underwent a right craniotomy. Through a transcortical and transcallosal approach a grey mass was identified beneath the corpus callosum. In addition to a fresh haematoma in the cavum septi pellucidi and organized haematomas between the right frontal horn and the mass, multiple calcified linear substances were evident under the operating microscope. Surrounding the right anterior septal vein, the mass was adherent to the genu of the corpus callosum, the medial aspect of the head of the right caudate nucleus and the medial end of the right thalamostriate vein, the right foramen of Monro being intact. The septum pellucidi was fenestrated at the posterior margin of the mass in order to free the mass from the anterior half of the cavum septi pellucidi. While the patent left anterior septal vein was involved in the mass, the left thalamostriate vein and the left foramen of Monro were intact as well as the internal cerebral veins. The diagnosis of parasitic granuloma was made on the frozen section of the mass. The surgeon excised the right anterior septal vein and most of the mass, leaving the portion adherent to the patent left anterior septal vein. The portion within the left frontal horn was also left because of the limited exposure (Fig. 1). The tip of the external ventriculostomy catheter was placed at the right atrium at the end of the operation and was removed 8 days later.

Three weeks after the craniotomy the patient was discharged home in a fully conscious state and her family were informed of the possibility of re-bleeding of the residual granuloma in the future. The patient remained conscious and without focal weakness at the 6th month and 22nd month follow-up.

Pathological examination of the intraventricular mass demonstrated an integumentary structure surrounded by granulomatous and astroglial cells (Fig. 2).

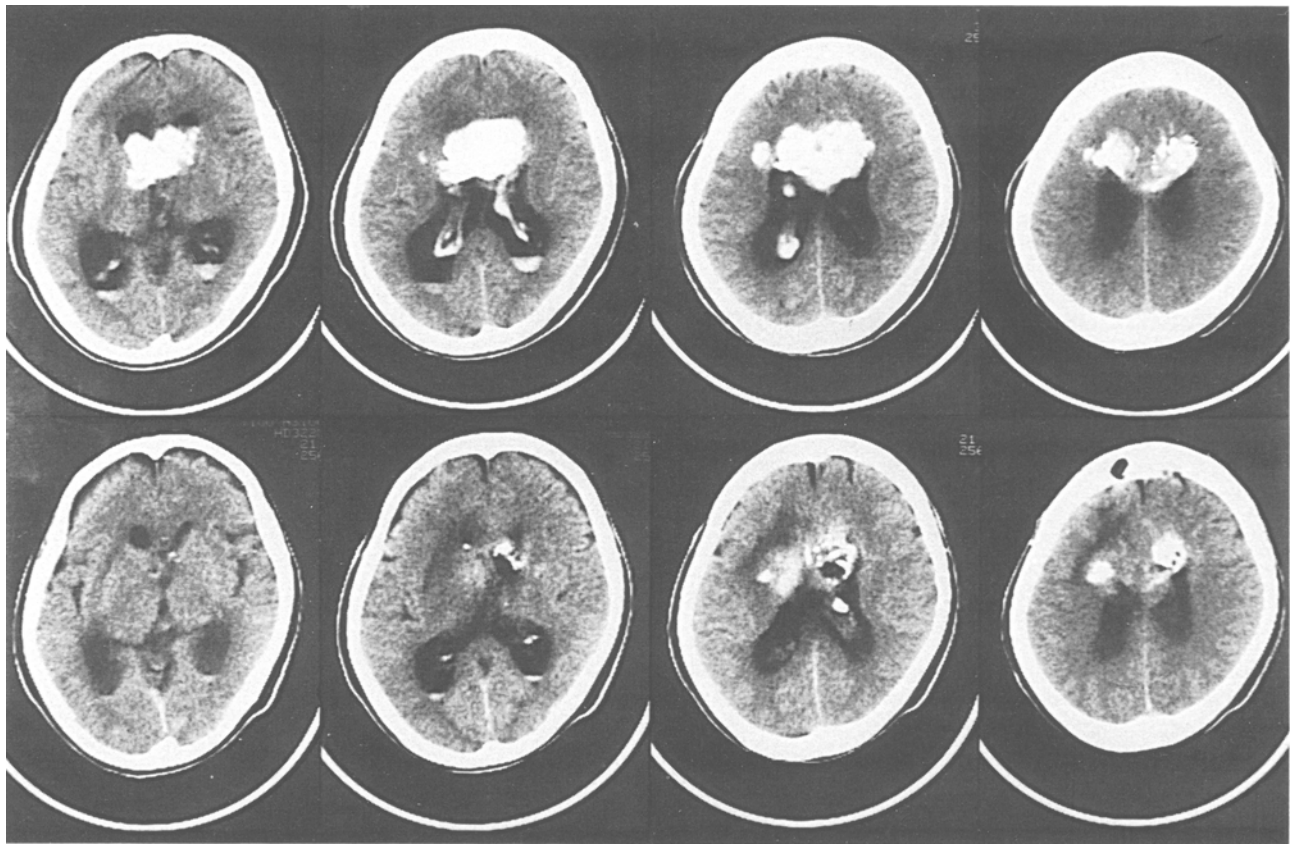
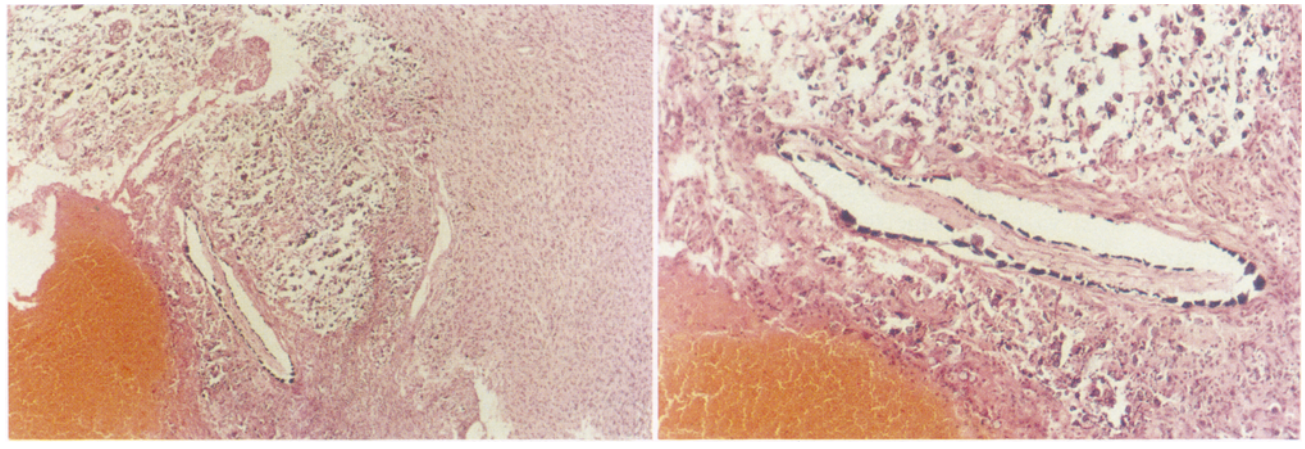


Fig. 1. Upper row: Pre-operative unenhanced CT showed the calcified mass, the intraventricular haemorrhage and the obstructive hydrocephalus with normal 3rd ventricle. Lower row: unenhanced CT 10 days after the operation showed the residual granuloma and the resolving hydrocephalus



a b

Fig. 2. (a) Characteristic of foreign body reaction, the mononucleated cells and the multinucleated giant cells surrounding the degenerating integument. The gliosis surrounding the granuloma may give rise to a false impression of a low-grade astrocytoma (× 122). (b) The longitudinal strips of smooth muscles and at least one oval corpuscle was visible in the body stroma (× 292). All the specimen were stained with haematoxylin and eosin

Discussion

Though rare, the diagnosis of cerebral sparganosis can be made with certainty when the larva is intact. This is not the case in our patient because the intraventricular granuloma was excised piecemeal and the cleft at the bothrium of the sparganum was not found in our surgical specimen[4, 6]. Accordingly, one cannot conclude that the structure in Fig. 2 was a pleroceroid

larva of Spirometra mansonoides. However the pathological features of an integumentary structure surrounded by granulomatous and astroglial cells were so consistent with those of the reported cases[2, 4, 5, 7, 9, 11, 13, 20] that a diagnosis of cerebral sparganosis was sustainable in this patient. The diagnostic problem in this case highlighted the possibility of misinterpreting the reactive gliosis as an astrocytoma (Fig. 2) had the parasitic granuloma escaped the notice of the pathologist, a point previously stressed in a report from our centre[18]. Indeed, cerebral sparganosis is often mistaken as brain tumour before operation except in the endemic areas where Enzyme-Linked Immuno-Sorbent Assay (ELISA) was employed[5].

The unique, previously unreported feature of this case was the parasitic invasion of the lateral ventricles. The peculiar location of the parasitoma gave rise to the special complications of bleeding and hydrocephalus. In contrast with the other cases in the literature, the large size of the parasitic granuloma in our patient probably reflects its location at a silent area of the brain. The operative finding of the old clots between the right frontal horn and the parasitic granuloma indicated that previous bleedings might have occurred without causing discernible symptoms. Had the fresh haematoma in the cavum septi pellucidi not compressed the foramina of Monro, the patient might not have presented with acute hydrocephalus. As the mass extended into both lateral ventricles, the surgeon found it difficult to excise the mass in the left frontal horn via the right frontal transcortico-transcallosal approach. Limited by the pericallosal arteries, an interhemispheric approach might not provide enough exposure to the frontal horns. With the benefits of knowing the histological diagnosis of a parasitic granuloma during the operation, the surgeon was able to decrease the risks by subtotal excision of the mass. The finding of chronic thrombosis allowed excision of the involved anterior septal vein with impunity.

Faeces of the primary host are shedded into fresh water and the eggs of the adult tapeworm are ingested by water fleas (Cyclops, first intermediate host) in which the eggs develop into procercoid larvae. When the Cyclops are eaten by mice, frogs, snakes and birds (second intermediate host) the procercoid larvae become plerocercoid larvae in the flesh of the second hosts. When the flesh of the second host together with the plerocercoid larvae are ingested by dogs and cats, the plerocercoid larvae mature into adult tapeworms and complete a life cycle[19]. Unable to mature when ingested by man, the larva penetrates the intestinal wall, leaves the peritoneum, migrates in the subcutaneous tissue and passes through one of the foramina in the skull base to reach the brain. As they do in the subcutaneous tissue of the body, the larvae tend to migrate to a lower resistance area of the brain, e.g. subcortical white mater and the subarachnoid space [12, 14, 16]. The deep seated location of the larva in this patient appeared an exception to the notion that the larvae have a predilection for low resistance tissue. The corpus callosum, composed largely of longitudinally oriented axons rather than densely packed neurones, is sufficiently similar to the subcortical white mater to account for the location of the parasite in our patient.

Niclosamides are effective antihelminthic agents while the tapeworm is still within the intestine of the primary host. Once the tapeworm extends beyond the intestine of the man, there is no evidence to support the therapeutic value of Praziquantal and Albendazole which have been effective agents for neurocysticercosis caused by Taenia folium, a tapeworm which primarily affects men[1]. It seems unnecessary to give these drugs to a patient in whom the larva is dead or surgically removed.

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