

The Dowling-Orlando technique in a giant primary cerebral hydatid cyst: a case report

Technika Dowlinga i Orlando w leczeniu olbrzymiej pierwotnej torbieli bąblowcowej mózgu: opis przypadku

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Abstract

Hydatid cyst disease is a parasitic illness that is rarely located in the brain. We present a case of a 26-year-old female who complained of headache, nausea, and vomiting. The diagnosis was intracranial hydatid cyst disease and was confirmed with radiological and serological tests. Neurological examination revealed papilloedema and left-sided pyramidal signs. She was operated on using the Dowling-Orlando technique. The cyst was removed without rupture, and therapy was completed with albendazole for a period of four weeks. In this article, we discuss the application of the Dowling-Orlando technique, microsurgery, the Valsalva manoeuvre and the positioning of the patient such that gravity could facilitate surgical removal of a giant cerebral hydatid cyst.

Key words: Dowling-Orlando technique, giant cerebral cyst, hydatid disease.

Introduction

Hydatid cyst is a widespread zoonotic disease which frequently involves the liver, lungs and, rarely, the brain. It is also a specific condition for which man-

Streszczenie

Bąblowica jest chorobą pasożytniczą, która rzadko umiejscawia się w mózgu. Przedstawiono przypadek dotyczący 26-letniej chorej, która skarżyła się na ból głowy, nudności i wymioty. Rozpoznanie bąblowicy śródczaszkowej potwierdzono w badaniach obrazowych i serologicznych. Pacjentka była operowana z wykorzystaniem techniki Dowlinga i Orlando, mikrocirurgii, manewru Valsalvy i ułożenia w taki sposób, aby grawitacja ułatwiła chirurgiczne usunięcie z mózgu olbrzymiej torbieli bąblowca.

Słowa kluczowe: technika Dowlinga i Orlando, olbrzymia torbiel mózgu, bąblowica.

datory notification of healthcare authorities is necessary. The most common agent is *Echinococcus granulosus*, which causes unilocular hydatid cyst formation [1,2]. A hydatid cyst consists of a hydatid sac (vesicle filled with fluid) with a thin, adventitial layer (neural tissue

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reaction) that may contain scolexes scattered in a limpid fluid [3].

Involvement of the central nervous system (CNS) can be observed in nearly 2-3% of hydatid cyst patients. Therefore, most CNS hydatid cyst cases are secondary conditions [4,5]. Cerebral hydatid cysts are generally located in the irrigation area of the middle cerebral artery [6]. Hydatid cysts may become life-threatening if ruptured during surgery after scolexes are scattered. The condition may be accompanied by re-plantation, recurrence, anaphylaxis and chemical meningitis [5,7]. The management of these cysts consists of surgical intervention and medical treatment. The Dowling-Orlando technique is currently the most popular technique used in surgical treatment of hydatid cysts. When the Dowling technique is combined with microsurgery, large cerebral hydatid cysts can be successfully removed without rupture [8,9].

Case report

A 26-year-old female patient was referred to our clinic with complaints of headache, nausea and vomiting that persisted for 4 weeks. Neurological examination of the patient showed bilateral papillae stasis and hemiparesis at the left upper and lower extremities (2/5). On the left side, the Babinski sign was present and deep tendon reflexes were brisk. Cranial computed tomography (CT) demonstrated a 7×8 cm mass located in the right parietal-occipital region. The lesion did not take up any contrast agent. There was effacement of the occipital horn of the ipsilateral ventricle with a midline

shift to the left due to the presence of a giant hydatid cyst (Fig. 1A). Upon magnetic resonance imaging (MRI), a cystic mass located in the right parietal-occipital region was present at a size of approximately $7 \times 8 \times 9$ cm with a regular border. The lesion was hypointense on T1-weighted images (Fig. 1B), and hyperintense on T2-weighted images (Fig. 1C). No primary focus was observed on thoracic CT, abdominal ultrasonography or MRI. The serological testing for *E. granulosus* was positive.

Operative technique

Under general anaesthesia, the patient was placed in the left lateral position. Wide, right fronto-parieto-occipital craniotomy was performed to remove the cyst.

A large opening in the dura matter suitable for the craniotomy was made. The surface of the cortex was highly hyperaemic, and the cortex was very fragile. Arachnoid dissection and sulcal incision were performed using a surgical microscope. The wall of the cyst was exposed and then dissected from surrounding tissues by careful microsurgical dissection. A soft catheter was inserted between the cyst and the cerebral tissue. Some warm hypertonic saline solution was provided through the catheter to increase hydrostatic pressure. A Valsalva manoeuvre was conducted with the help of the anaesthesia team. The head of the operating table was lowered so that gravity would have an effect. Slight pressure and slow movements were applied to the adjacent tissues of the brain to remove the cyst without rupturing it. After removal of the cyst, hypertonic saline

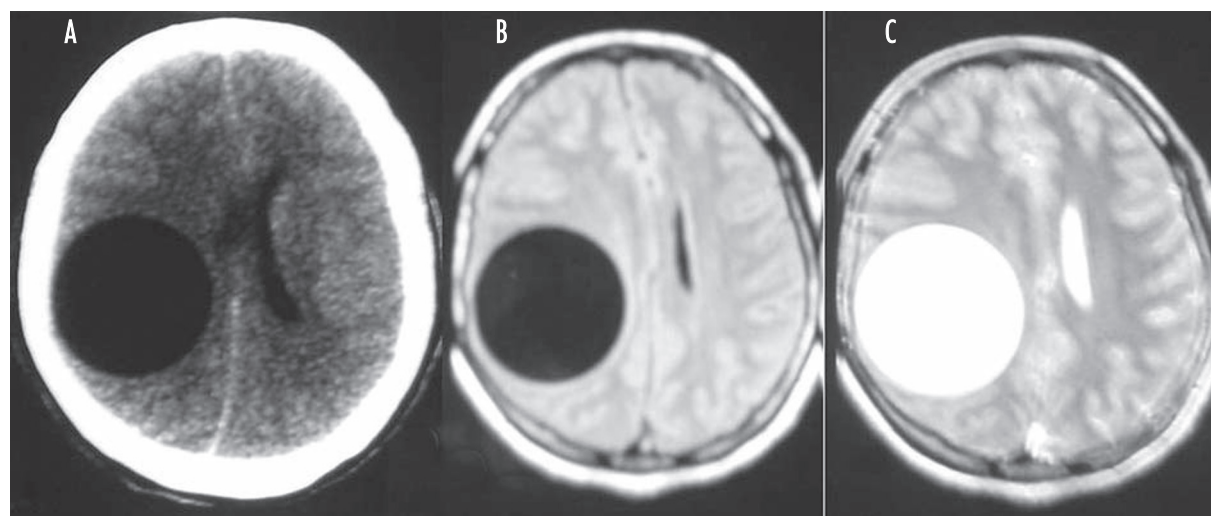


Fig. 1. Axial computed tomography (A) and magnetic resonance images (B, C) showing an intra-axially located lesion with the density of cerebrospinal fluid

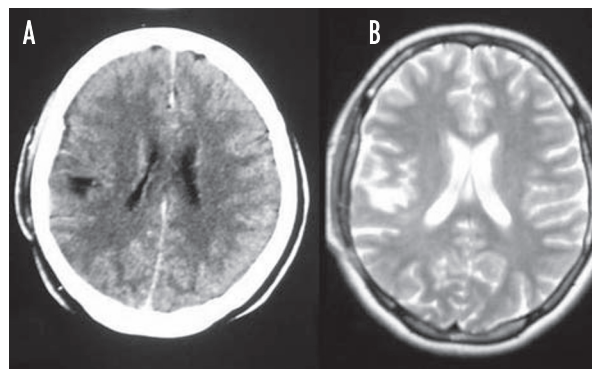


Fig. 2. Hypodense, irregular areas and hyperintense regions suggesting cystic malacic changes on postoperative computed tomography (A) and magnetic resonance imaging (B)

injection was applied to the new cavity. Then the dura mater was sutured, and the operation ended. No post-operative complication developed in the patient. A significant improvement was observed in hemiparesis (4/5). Postoperative CT and MRI did not reveal any residu-

al cystic tissue. Albendazole was administered as post-operative prophylactic therapy for 4 weeks.

Discussion

Cerebral hydatid cysts may not show any symptoms until they reach a considerably large size. The most effective method for treating these cysts is to remove the cyst by surgical intervention. The goal is to remove the cyst without rupturing it [1,3,10].

If the cyst ruptures during surgery, it may lead to serious conditions that impact morbidity and mortality. Various authors have indicated a rupture ratio during surgical intervention of 0.3–53%. Recurrence is inevitable if a hydatid cyst ruptures [6,11]. El-Shamam *et al.* observed that recurrence occurred in all ruptured cysts [11]. The Dowling-Orlando technique can be safely used in the surgical removal of giant hydatid cysts [8,9,12,13]. Unfortunately, some difficulties may be encountered. The surgeon must be very careful as the

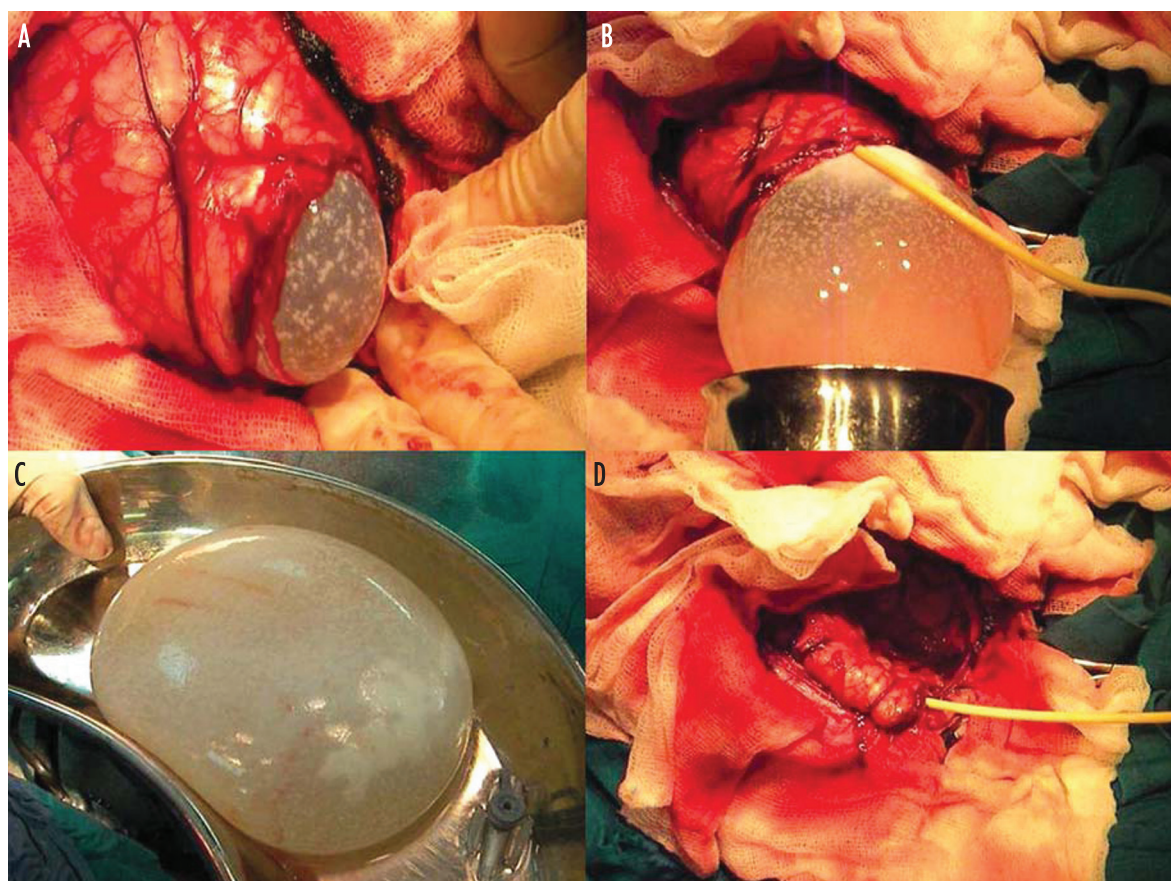


Fig. 3. The cyst protruding from the brain (A); the rubber catheter inserted between the cyst and the brain (B); the cyst after removal (C); the residual cavity after removal of the cyst (D)

cortex and the dura matter may not exist between the cyst walls and cranial vault after craniotomy. Many different microsurgical techniques and micro-instruments are employed during dissection [14,15]. The technique is considered as effective but also has considerable limitations. It could be difficult to apply this technique in patients whose cysts are in high-risk locations or where no instrument can be used. Additionally, pre-operative albendazole therapy may effectively reduce the tension of an intracranial hydatid cyst [16]. Nevertheless, the best operative approach may vary according to the size and location of the cyst, as well as its relationship with other neural tissues.

If the cyst is not under the cortex, it may be impossible to gain access to it without entering some neural tissues. The rupture rate of hydatid cysts is higher in patients in whom the cyst is located deep in the parenchyma. Retractions that are performed to reach a cyst located in deep tissues of the brain may cause temporary or permanent neurological deficits in some patients. The Dowling-Orlando technique can be used safely even in cases with large cerebral hydatid cysts that may be near to the cortex. The technique could be improved with microsurgery and by lowering the head of the operating table.

In conclusion, the Dowling-Orlando technique is the gold standard for the surgical treatment of cerebral hydatid cyst disease. It can also be effectively applied to the treatment of giant cerebral hydatid cyst disease without rupturing the cyst if it is combined with microsurgical techniques, the use of gravity and the Valsalva manoeuvre.

Disclosure

Authors report no conflict of interest.

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