Hydatid Cyst of the Third Ventricle, Managed Successfully using Dowling's Technique

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Abstract: We report a 16-year-old female with a hydatid cyst within the third ventricle who was treated by surgically removing the cyst using Dowling's technique. Via a cortical incision in the left superior parietal lobule, the surgical corridor was made towards the trigone of the left lateral ventricle and the cyst was removed intact by taking advantage of the gravity force and using hydrostatic expulsion. Afterwards, the cyst cavity was irrigated with 3% saline solution. Early postoperative period was uneventful, and her communication and motor abilities improved steadily by rehabilitation program. It can be concluded that with precise planning and using accurate technique, it may be possible to deliver even deep-seated hydatid cysts using Dowling's technique; however, this conclusion could not be generalized and each individual patients requires a tailored treatment strategy.

Keywords: Hydatid Cyst, Dowling's Technique, Third Ventricle.

INTRODUCTION

Cerebral hydatidosis is a serious condition and intracranial hydatid cysts comprise 2.3 to 3.4% of all intracranial space-occupying lesions [1]. The cysts are usually solitary, large, and confined to white matter [2]. While the definite treatment of cerebral hydatid cysts consists of surgical extirpation, these lesions impose a formidable challenge to the surgeon. Their fortressed location within brain tissue along with their high intra-lesional pressure makes their excision quite difficult. Besides, such lesions should be excised while they are physically intact and the walls of cyst are not breached. The surgical technique was originally introduced and developed by Dowling in 1929 [3] and later developed by others [4]. If a breakdown occurs in the cyst wall during its surgical removal, the clinical result will be disappointing since cyst rupture causes the daughter cysts and the scolices to get implanted in the surrounding brain tissue and their seeding *via* the cerebrospinal fluid (CSF) pathways to distant areas along the neuraxis would proceed.

Involvement of the ventricular system by hydatidosis is an extremely rare entity [5]. It is obvious that such cysts are more difficult to treat because of their deep location. The reasons are twofold: (1) access to the deep sites of brain is a more challenging surgical odyssey; (2) the surgical corridor must be as much large as to allow safe delivery of the cyst while as much small as possible to minimize the complications of destruction of normal brain tissue.

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We report a case of 16-year-old female with a hydatid cyst within the third ventricle. The cyst was removed intact *via* approach to the left atrium accessed through the left superior parietal lobule using Dowling's technique.

CASE REPORT

A 16 year-old female from a rural area of the Northwestern Iran was referred to our center with a 3-year history of seizures and progressive decline in the level of consciousness. She suffered from attacks of generalized tonic clonic seizures that had become intractable despite maximal medical treatment; she had gradually lost visual and hearing sensations.

On physical examination, the eyes were open but the patient was unable to communicate or obey motor commands. The best motor response was localizing painful stimulus, and she had no verbal or emotional expression. The brainstem and spinal reflexes were intact, and there was no lateralizing neurological sign. Bilateral optic atrophy was found in fundoscopy.

After developing hydrocephalus, the patient had undergone ventriculoperitoneal shunting 2 years ago to alleviate symptoms. However, the seizures remained uncontrolled and neurological deficits had progressed continuously.

On brain computed tomography (CT) and magnetic resonance imaging (MRI), a large spherical well defined cystic lesion was seen within the third ventricle with a slight extension to the left side. The signal intensirty of the cyst contents was similar to CSF. There was no edema in the surrounding brain tissue and the lesion did not show contrast enhancement.

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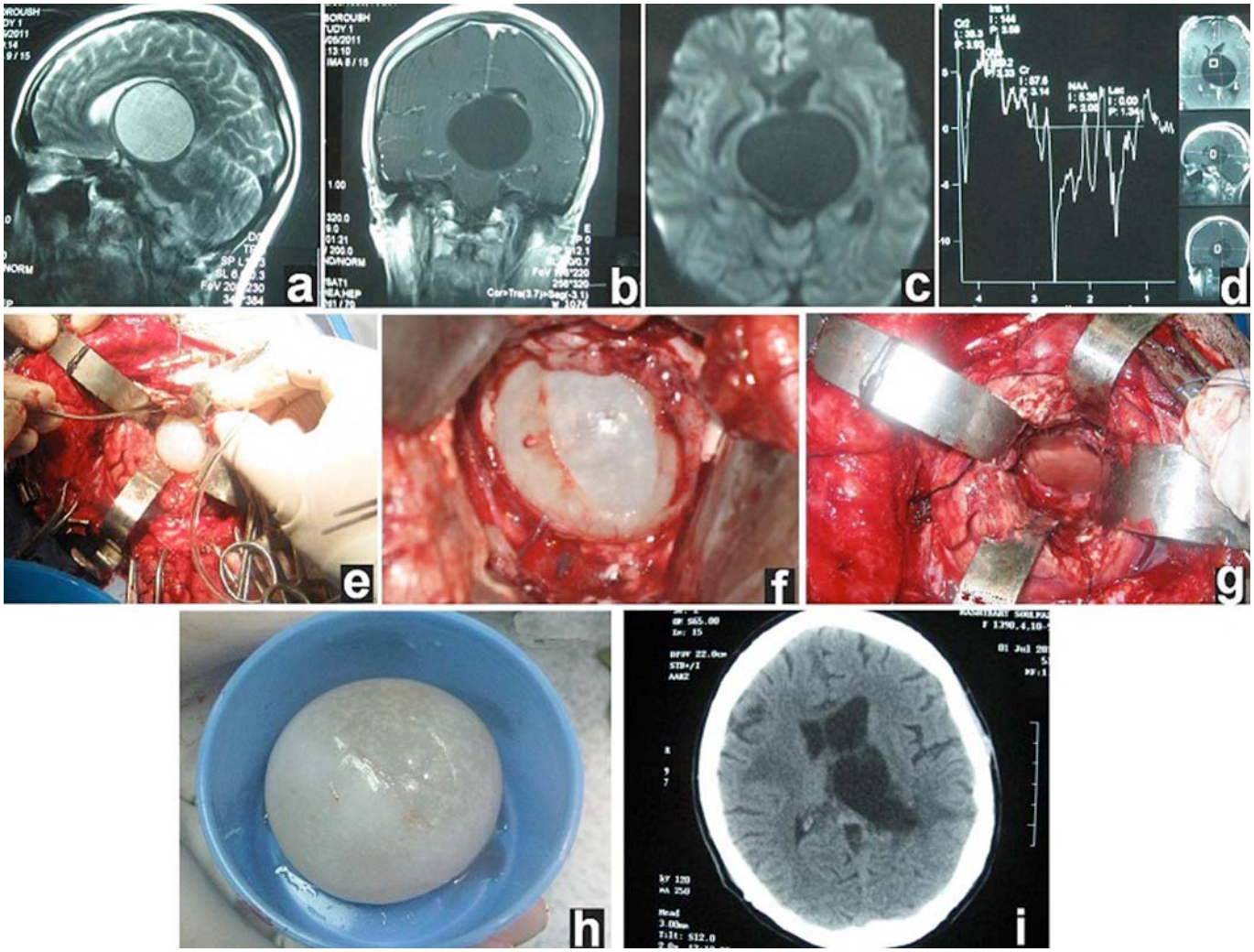

Figure 1: T2-weigted MRI showing a large hyperintense spherical well defined cystic lesion within the third ventricle (**a**). The lesion is hypointense in T1-weigthed image with no post-contrast injection enhancement (**b**). Diffusion weighted sequence reveals no evidence of restriction (**c**). Magnetic resonance spectrography showing diminished level of N-acetyl-aspartate (NAA) (**d**). Hydatid cyst delivery through corticectomy using gravity and hydrostatic forces (**e**, **f**). The empty cavity after cyst removal (**g**). Intact removed cyst (**h**). Post-operative brain computed tomography (**i**).

Sulcal effacement, basal cisternal closure, and mass effect on the tectal plate and midbrain, left thalamus, and callosal splenium were observed (Figure **1a-b**). Diffusion weighted sequence showed no evidence of restriction (Figure **1c**). Magnetic resonance spectroscopy (MRS) revealed diminished level of the neural marker, N-acetyl-aspartate (NAA), along with nonspecific surges in lactate (Lac) and creatine (Cr) waves; a finding which supports the idea that the cyst fluid is mainly composed of non-neural chemicals. Yet, MRS could not provide any further diagnostic data (Figure **1d**).

Considering the mentioned radiologic findings, the patient underwent systemic work-up to rule out hydatid disease. Abdominal ultrasound revealed multiple cysts in peritoneum and liver. The indirect hemagglutination test was positive for hydatid disease. Nevertheless, neither the lungs were involved, nor echocardiography revealed any valvular vegetation.

Operation

The patient was scheduled for surgery in order to remove the cyst intact. In semi-lateral position, with sagittal midline parallel to the floor and the body being secured to the table by tape, a large left parieto-occipital craniotomy was performed. After opening the dura in cruciate pattern, a linear posterior parietal cortical incision was made toward the atrium of the left lateral ventricle. The ventricular cavity was reached at a depth of 3 cm, and the shadow of the cyst became apparent behind the protruded upper wall of the third ventricle (into the left atrium) which was dissected to expose the cyst wall (Figure **1e**). The head was turned toward the floor and the cyst was removed intact using

gravity force and hydrostatic expulsion (Dowling's technique) (Figure **1f-h**). The cyst cavity was irrigated immediately with 3% saline solution (Figure **1g**).

After surgery she was extubated and regained the pre-operative level of consciousness. The patient was taken care of in the neuro-intensive care unit for 24 hours. She did not develop any further neurological deficit and the head CT showed excision of the cyst along with brain relaxation and resolution of hydrocephalus (Figure **1i**). She was prescribed albendazole, 400mg twice a day to control systemic hydatid disease. Rehabilitation program and occupational therapy were scheduled and started during the inpatient hospital stay and continued for the next 12 weeks and the patient's condition gradually improved in terms of communication ability and motor activities.

DISCUSSION

Iran lies in the endemic zone of *echinococcosis* [6]. Intracranial hydatid disease is rare even in endemic areas, and the brain involvement occurs only in 1-4% of cases [7]. Cerebral hydatid cysts are usually located in the watershed zone of the middle cerebral arteries [8]. However, few documented cases of unusually located intracranial hydatid cysts in thalamus [9-10], parasellar region [11], interpeduncular cistern [7], cavernous sinus [12], and brainstem [13-14] have been reported. Ventricular location is even more uncommon. Rare reports of cerebral hydatid disease in the lateral ventricle [15], fourth ventricle [16], and the aqueduct of Sylvius [17] have been published. In this paper, we present an unusual case of hydatid cyst in the third ventricle.

Mechanisms of Intraventricular Hydatid Cyst Formation

The proposed mechanisms for intraventricular cyst implantation include (1) reaching the ventricular cavity *via* the choroid plexus, (2) spillage of a ruptured primary cyst situated near the ependymal lining into the ventricular system, and (3) birth of the cyst into the ventricle from spontaneous ependymal rupture [15].

Treatment

Classically, the treatment of cerebral hydatid cysts is exclusively surgical [14]. The main goal of the operative intervention is to remove the cyst undamaged with its contents, to avoid leakage of hydatid fluid which possibly causes immediate anaphylactic shock, meningitis, or inevitably local recurrence when the contents are spilled [10]. Delivering the cyst intact using Dowling technique is still the standard method [18]. Essential steps of the technique described by Carrera *et al.* [3] are: (1) a large flap; (2) careful handing during all the operative steps avoiding monopolar coagulation; (3) opening of the atrophic cortex overlying the cyst over an area whose diameter should be no less than three quarters of the diameter of the cyst, and (4) letting the cyst come out by just lowering the head of the operating table and instillation of warm saline between the cyst and surrounding brain. In our opinion, the anesthesiologist plays a crucial role in the delivery process by controlled increase of the intracranial pressure. Some state that the use of mannitol is a quite useful tact to let the brain shrink and let go of the cyst [19]. However, trouble arises in this approach when the cyst is located in deep areas. A major limitation to the removal of hydatid cysts in these areas has been the inability to clearly define the safe anatomic route [14]. Furthermore, such cysts are more prone to tearing throughout the procedure. Avoiding this risk, some authors have proposed a cortical incision equal to the cyst diameter, though it may pose a higher range of morbidity [10]. Some others advocate internal decompression by aspiration followed by extirpation of the cyst wall as an alternative option in deep-seated hydatid cysts, though it may raise the risk of recurrence [13].

In this particular case, the patient's condition was so critical that required a life saving intervention to decompress neural structures. Aspiration technique was expected to carry a high risk of recurrence and dissemination, since the scolices could directly spread throughout the whole ventricular system. Hence, the plan was removal of the cyst intact. Transcallosal approach was not considered, as the cyst diameter was large and imposed an extensive callosal incision and retraction, which endangered nearby structures and would impose severe morbidity. So, the lesion was approached from posterior parietal corridor and the cyst was removed with intact contents using hydraulic forces and gravity.

CONCLUSION

This case report shows successful removal of a large deeply seated hydatid cyst of the brain using Dowling's technique. The careful surgical planning and taking extreme caution to deploy all the necessary armamentarium and strategies, the cyst was extirpated without being damaged. However, it would be *non*

sequitur to conclude that any large deep hydatid cyst of the brain could be safely removed by using the Dowling's technique. Still, correct surgical planning and tailoring the treatment strategy to each individual patient are the indispensable cornerstones of a successful therapy in such patients.

ASSURANCES

The study has been approved by the institutional review boards and the patient's parents gave informed consent.

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COMPETING INTEREST

The authors declare that they have no conflict of interest.

CONTRIBUTORSHIP STATEMENT

Z.H; substantial contribution to conception and drafting the manuscript. F.T; revising the manuscript critically. ATM; final revising of the manuscript. SA.F.T; final approval of the version to be published.

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