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Surgical Treatment of Intradiploic Epidermoid Cyst Treated as Depression

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SUMMARY

Introduction Extradural intradiploic epidermoid cysts are rare, representing less than 0.25% of all primary intracranial tumors. They can be neurologically silent and can only present psychiatric symptoms like depression, cognitive or personality changes.

Case Outline A 68-year-old male with two year long history of depressive mood, lack of motivation, helplessness, hopelessness and poor response to antidepressive drug therapy was described. CT scan showed a well-defined mass in the parietal scalp with destruction of the scull. He underwent intracranial tumor resection. Surgical resection and cranioplasty were performed. Pathology confirmed intradiploic epidermoid cyst.

Conclusion Total removal of these cysts and repeated washing of the cavity with 0.9 % saline may prevent recurrence and aseptic meningitis and may improve mental state of the patient. We also emphasize the need for neuroimaging studies in a patient with atypical changes in mental status, even without neurological signs or symptoms.

Keywords: epidermoid cyst; brain tumor; depression

INTRODUCTION

The cysts originate during weeks 3–5 of gestation from the ectodermal cellular remnants that arise from the incomplete cleavage of the neural ectoderm from the cutaneous ectoderm. Epidermoid cysts have been described as non-neoplastic cysts and represent approximately 1% of all primary intracranial tumors. They may be intradural (usually extra-axial) or extradural (usually arising in the diploic space of calvaria). Intradural cysts most frequently involve the posterior cranial fossa, especially the cerebellopontine angle (CPA).

Extradural intradiploic epidermoid cysts, like epidermoid cysts in other cranial locations, are rare, accounting for less than 0.25% of all primary intracranial tumors [1, 2]. They can be located in any part of the skull, and occur from the first to the seventh decade [3]. These lesions are usually discovered incidentally and may remain asymptomatic for many years. They can be often manifested only through the changes in mental state and remain undiscovered for many years if they grow intracranially and produce brain compression or undergo malignant change [4].

Intracranial tumors may give rise to symptoms simulating depression, anxiety states, hypomania and schizophrenia [5]. Most often, it is slow-growing benign tumors that are responsible.

Epidermoid cysts usually grow insidiously at a linear rate, and can result in slow onset of neurological and psychiatric symptoms. Patients can present with depression, anxiety, cognitive or personality changes, psychosis, apathy/abulia [5, 6]. Psychiatric symptoms, such as depression or mania, may be initial presenting symptoms in some cases of brain tumors [7-11].

In this report, we describe the clinical, radiologic and pathologic aspects of a 68-year-old male with an epidermoid cyst of the parietal bone.

CASE REPORT

A 68-year-old male patient was admitted to Neurosurgery Department, Clinical Hospital Center Zemun, with minor weakness of the right side of his body and subcutaneous mass on the left parietal scalp.

A year and a half ago, before admission, he consulted a psychiatrist due to depressive thoughts and problems with memory and motivation. The patient had frequent headaches, depressed mood most of the day, diminished interest in almost all activities, insomnia, increased appetite and diminished ability to think or concentrate and remember. There was no organic problem in his medical history, no head traumas, and there was no family history of neuropsychiatric diseases. He also mentioned the dysesthesia on the left side of the skull. The patient was treated after being diagnosed as psycho-organic syndrome (mild

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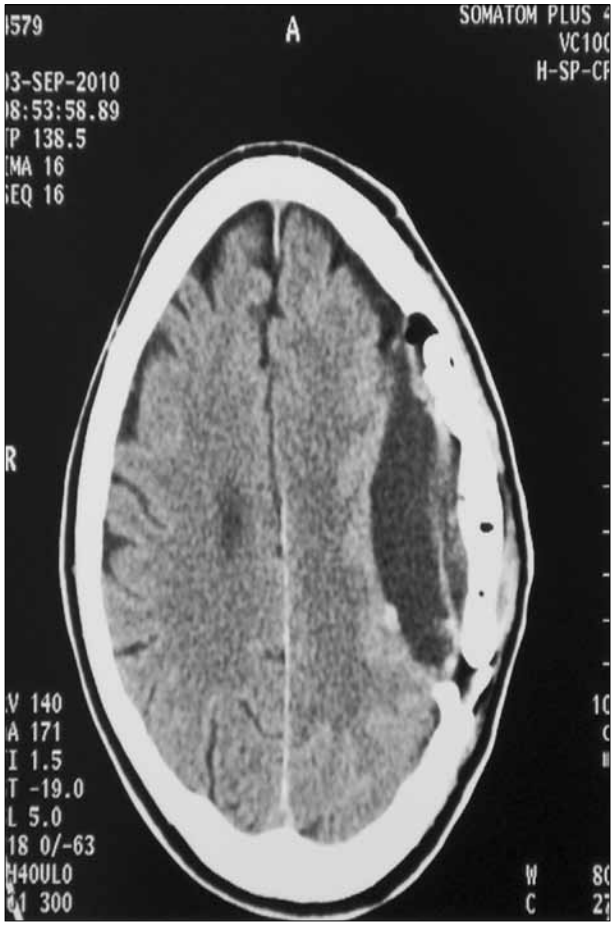

Figure 1. CT scan showing well-defined mass (6×7 cm) in the parietal scalp with destruction of the skull and compressed brain tissue

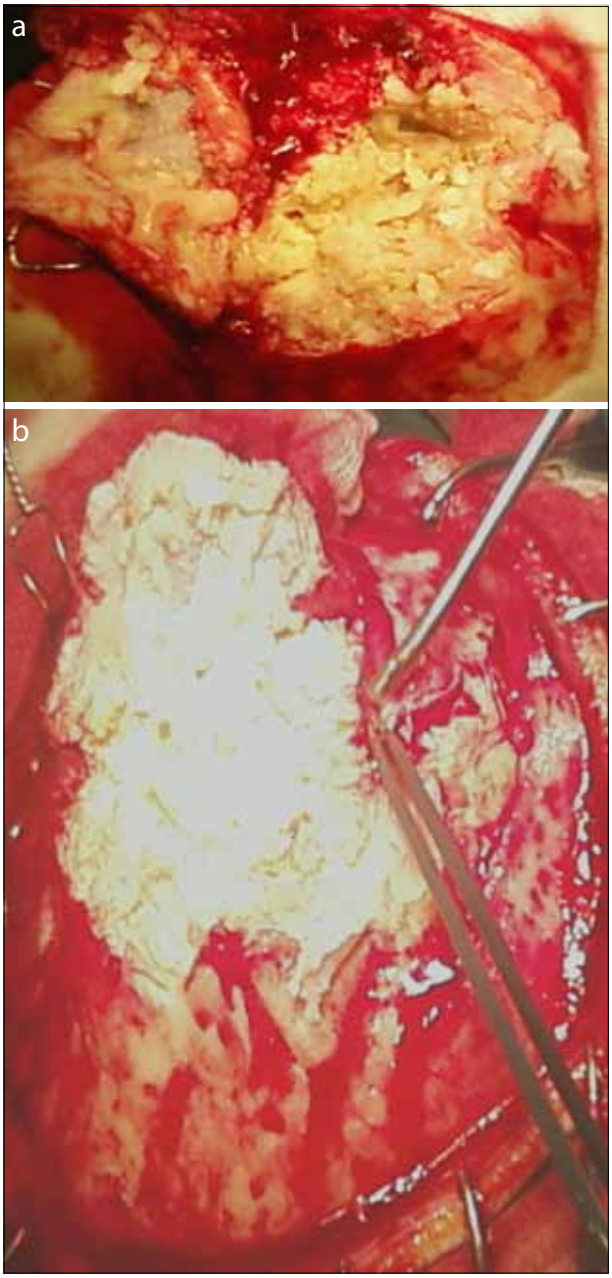

Figure 2. Intraoperative images: a) the tumor was avascular, lightly colored, and soft; b) the parietal bone was partially eroded and damaged

cognitive disorder) and major depression with tanakan, sertraline and lorazepam. After one-year treatment with antidepressive drugs there was no change in his symptomatology. An electroencephalogram (EEG) was done and it was normal. He was examined by the psychiatrist one month before hospitalization because his symptoms progressively worsened and he got weakness of the right side of his body and difficulties with speaking.

CT scan was indicated and showed a well-defined mass (6×7 cm) in the parietal scalp with the destruction of the skull (Figure 1) and compressive effect on the left ventricle. Epidermoid cyst originating from diploe and eosinophilic granuloma was considered. Preoperative cerebral angiography was undertaken to check the blood supply of the mass and eliminate vascular anomalies: abnormality was not observed. No further neuroradiological examination was needed because the present finding showed clearly extracerebral mass with parietal bone erosion. The patient denied any kind of trauma to this region. His routine hematological and biochemical parameters were normal.

There were no well-defined margins in the deep portion and the mass was totally removed under general anesthesia. Intraoperatively, epidermoid tumor was exposed upon the skin incision beneath the galea aponeurotica pericranial layer. The tumor was light and white colored and was associated with parietal bone erosion (Figure 2). After the dissection of tumor margins, it was intraoperatively shown that the tumor infiltrated all parietal bone tissue with its intracranial extension to the dura. Dura matrix was intact. Surgical finding confirmed that the tumor originated from diploe. Complete removal of the tumor and cranioplasty were carried out (Figure 3).

Frozen and paraffin sections showed that the cystic structure was lined by squamous epithelium containing laminated keratin material. Pathologic findings confirmed the suspicion of intradiploic epidermoid cyst.

Postoperative recovery was uneventful, and the patient was discharged nine days after surgery without neurological signs and antidepressive medications were not restarted following the surgery. In the early postoperative period, an improvement in his psychic condition was evident. He

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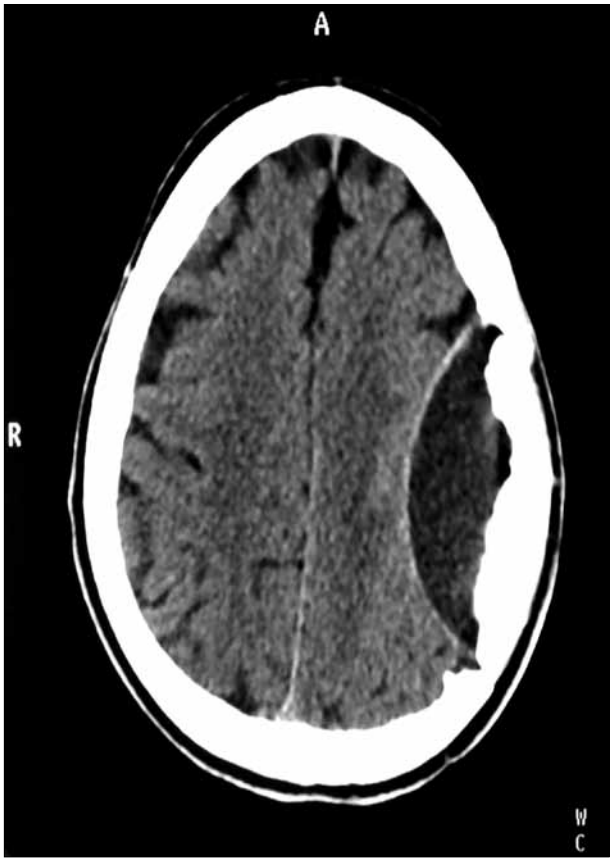

Figure 3. CT scan after six months showing no reccurence of epidermoid cyst

had not symptoms of frequent headaches and manifested depressive thoughts. His motivation was improved, he started to sleep and eat better and his activities in every day life improved. During the following six months after the surgery there was no evidence of tumor recurrence.

DISCUSSION

Epidermoid tumors were first described in 1829 by the French pathologist Cruveilhier. Since Cushing [12] first described a large diploic epidermoid cyst in 1922, only a few cases of giant calvarial intradiploic epidermoids have been reported.

These rare lesions arise from displaced ectodermal cells during the closure of the neural tube in weeks 3–5 of embryonic life. The lesions grow very slowly along natural cleavage planes. By the time of diagnosis, they usually involve several regions; therefore, it is difficult to locate their exact origin. Approximately 40% to 50% of intracranial epidermoid tumors are localized to the CPA, making it the most common intracranial location. Extradural intradiploic epidermoid cysts, like epidermoid cysts in other cranial locations, are rare, representing less than 0.25% of all primary intracranial tumors [1, 2]. Epidermoid neoplasms are more common in men than in women, with the onset of symptoms occurring between the ages of 20 and 50 years [2]. Although both diploic tables are frequently involved, giant diploic epidermoids are associated with the extensive destruction of the inner table and prevalent intracranial growth [13, 14].

The diagnosis of epidermoid cyst was suggested by imaging (skull radiographs, CT scan, MRI) and confirmed by histology. CT scan allows for good assessment of both skull involvement and intracranial extension and reveals the exact site, limits, and characteristic bone defects of these lesions [3]. The typical CT aspect is a large homogenous hypodense unenhancing mass, with or without calcifications, typically showing a density range of –20 to +20.

Differential diagnosis should include dermoid cyst, hydatid cyst, arachnoid cyst, cholesterol granuloma, eosinophilic granuloma, aneurysmal bone cyst, and meningioma [3]. It is particularly common to misdiagnose an epidermoid cyst as a dermoid cyst, as the difference between them is mainly histological.

The definite diagnose can be achieved by surgical removal and histopathological confirmation.

The indications for surgery include cosmetic effect, prevention of progression of psychiatric symptoms and neurological deficit, treatment of osteomyelitis, and resection of cysts with malignant degeneration [1]. Most cranial epidermoids are small and do not extend intracranially, but progressive growth may result in large cranial defects or compression of the brain and vascular structures [15, 16]. Removal of these tumors and subsequent cranioplasty, despite their large size, are recommended [17], particularly for very large intradiploic epidermoid cysts associated with significant bony defects [1]. Total removal of these cysts is associated with a very good long-term prognosis [18, 19]. Recurrence is likely if the cyst wall is not completely removed, with a recurrence rate of 8.3–25.0% [20]. We were able to remove the cyst and capsule completely in our patient. Repeated washing of the cavity with 0.9% saline prevented aseptic meningitis and recurrence. A postoperative antibiotic regimen was implemented to prevent infection.

This case also illustrates the need for a prompt thorough assessment when patients present any atypical psychiatric symptoms or changes in their mental state. Brain imaging should be undertaken.

Our 68-year-old male patient was treated in a psychiatric department for a year and a half before the brain tumor was diagnosed. It is unclear in our case whether his psychiatric symptoms were caused by large epidermoid cyst or he developed tumor at a later stage. Atypicality of presentation, poor response to treatment or waxing of symptoms should lead to suspicion of brain tumor. It is possible that magnetic resonance imaging/CT scan with contrast may have detected the mass earlier.

As in our case, the mass was associated with the massive bone destruction and intracranial extension with the compressive effect on the brain, which caused depression and psycho-organic syndrome in the first place and later on the weakness on the left side of his body and dysphasia. A lot of tumors are manifested only with changes in the mental state and usually with uncommon symptoms. The most common is depression, lack of motivation and memory deficits. One of the alarming signs of the tumor presence is resistance to antidepressive drug therapy. Treatment of

diagnosed depression is sometimes problematic in patients with brain tumors. The timing of the initial prescription of the medications relative to the diagnosis of the tumor is unknown. The side effects of antidepressant medications on patients with brain tumors are not well featured. Almost all antidepressant medications may lower seizure threshold [21, 22]; which antidepressant would be least likely associated with the increased seizure activity in this situation is not clearly defined. The usual side-effects of particular anti-depressant medications are at risk for being magnified by the presence of the brain tumor. Perhaps most importantly, the efficacy of anti-depressant medications in this patient population is unknown.

Intradiploic epidermoid cysts are benign lesions of the skull that may undergo malignant transformation. It is important to consider this diagnosis in a patient who presents with a slowly progressive scalp mass that demonstrates a lytic lesion on the x-ray. Precise radiological assessment and complete removal of the tumor and its capsule are essential for a good long-term prognosis. Repeated washing of the cavity with 0.9% saline may prevent aseptic meningitis and recurrence. This case also illustrates the need for prompt assessment when patients present with any atypical psychiatric symptoms or changes in the mental state. Brain imaging should be undertaken in such cases.

REFERENCES

1. Prall JA, Lloyd GL, Breeze RE. Traumatic brain injury associated with an intradiploic epidermoid cyst: case report. Neurosurgery. 1995; 37:523-5.
2. Hakyemez B, Aksoy U, Yildiz H, Ergin N. Intracranial epidermoid cysts: diffusion-weighted, FLAIR and conventional MR findings. Eur J Radiol. 2005; 54:214-20.
3. Bikmaz K, Cosar M, Bek S, Gokduman CA, Arslan M, Iplikcioglu AC. Intradiploic epidermoid cysts of the skull: a report of four cases. Clin Neurol Neurosurg. 2005; 107:262-7.
4. Tamura K, Aoyagi M, Wakimoto H, Tamaki M, Yamamoto K, Yamamoto M, et al. Malignant transformation eight years after removal of a benign epidermoid cyst: a case report. J Neurooncol. 2006; 79:67-72.
5. Madhusoodanan S, Danan D, Brenner R, Bogunovic O. Brain tumor and psychiatric manifestations: a case report and brief review. Ann Clin Psychiatry. 2004; 16:111-3.
6. Moise D, Madhusoodanan S. Psychiatric symptoms associated with brain tumors: a clinical enigma. CNS Spectr. 2006; 11:28-31.
7. Bunevicius A, Deltuva VP, Deltuviene D, Tamasauskas A, Bunevicius R: Brain lesions manifesting as psychiatric disorders: eight cases. CNS Spectr. 2008; 13:950-8.
8. Litofsky NS, Resnick AG. The relationships between depression and brain tumors. J Neurooncol. 2009; 94:153-61.
9. Arnold SD, Forman LM, Brigidi BD, Carter KE, Schweitzer HA, Quinn HE, et al. Evaluation and characterization of generalized anxiety and depression in patients with primary brain tumors. Neuro Oncol. 2008; 10:171-81.
10. Jarquin-Valdivia AA. Psychiatric symptoms and brain tumors. Arch Neurol. 2004; 61:1800-4.
11. Zivkovic N, Berisavac I, Markovic M, Benovic R, Samardzic M, Popovic I. Psychiatric manifestations of brain tumors. Materia Medica. 2010; 26:173-6.
12. Cushing H. A large epidermal cholesteatoma of the parietotemporal region deforming the left hemisphere without cerebral symptoms. Surg Gynecol Obset. 1922; 34:557-66.
13. Arana E, Latorre FF, Revert A, Menor F, Riesgo P, Liano F, et al. Intradiploic epidermoid cysts. Neuroradiology. 1996; 38:306-11.
14. Constans JP, Meder JF, de Divitiis E, Donzelli R, Maiuri F. Giant intradiploic epidermoid cysts of the skull. Report of two cases. J Neurosurg. 1985; 62:445-8.
15. Rengachary S, Kishore PR, Watanabe I. Intradiploic epidermoid cyst of the occipital bone with torcular obstruction. Case report. J Neurosurg. 1978; 48:475-8.
16. Skandalakis JE, Godwin JT, Mabon RF. Epidermoid cyst of the skull: report of four cases and review of the literature. Surgery. 1958; 43:990-1001.
17. Jaiswal AK, Mahapatra AK. Giant intradiploic epidermoid cysts of the skull. A report of eight cases. Br J Neurosurg. 2000; 14:225-8.
18. Constans JP, Meder JF, De Divitiis E, Donzelli R, Maiuri F. Giant intradiploic epidermoid cysts of the skull. Report of two cases. J Neurosurg. 1985; 62:445-8.
19. Guridi J, Ollier J, Aguilera F. Giant intradiploic epidermoid tumor of the occipital bone: case report. Neurosurgery. 1990; 27:978-80.
20. Yanai Y, Tsuji R, Ohmori S, Tatara N, Kubota S, Nagashima C. Malignant change in an intradiploic epidermoid: report of a case and review of the literature. Neurosurgery. 1985; 16:252-6.
21. Baldessari RJ. Drug therapy of depression and anxiety disorders. In: Brunton LL, editor. Goodman and Gilman's the Pharmacological Basis of Therapeutics. 11th ed. New York: The McGraw Hill Companies, Inc; 2006. Ch. 17 [on-line edition].
22. Hall G, Fitzgerald DJ. Psychiatric emergencies. In: Stone CK, Humpries RL, editors. Current Diagnosis and Treatment: Emergency Medicine. 6th ed. Texas: The McGraw Hill Companies, Inc; 2008. Ch. 47 [on-line edition].

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Хируршко лечење интрадиплоичне епидермоидне цисте лечене као депресија

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КРАТАК САДРЖАЈ

Увод Екстрадуралне интрадиплоичне епидермоидне цисте су ретке и чине мање од 0,25% свих интракранијалних тумора. Болесници с овим цистама могу бити без неуролошких тегоба и испољити само психијатријске симптоме, као што су депресија, когнитивне или промене личности.

Приказ болесника Приказан је мушкарац стар 68 година који се две године пре пријема лечио због депресивног расположења, недостатка мотивације и беспомоћности и код којег је забележен лош одговор на терапију антидепресивима. Скенер ендокранијума је показао јасно дефинисану масу паријетално са деструкцијом кости лобање. Урађена је екстирпација интракранијалног тумора са краниопластиком. Патохистолошки налаз је потврдио интрадиплоичну епидермоидну цисту.

Закључак Екстирпација епидермоидне цисте и прање кавума физиолошким раствором у концентрацији од 0,9% може да спречи рецидив и појаву асептичног менингитиса, а може и побољшати психичко стање болесника. Такође наглашавамо потребу за раном неурорадиолошком дијагностиком код особа са нетипичним променама менталног статуса, чак и без неуролошких симптома и знакова обољења.

Кључне речи: епидермоидна циста; тумор на мозгу; депресија

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