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Giant Occipital Intradiploic Epidermoid – Case Report and Review of Literature

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Abstract- Aim: Intradiploic epidermoid cysts are rare cranial tumors, with the reports on the giant variety being few to date. We report a case of a giant occipital intradiploic epidermoid and review the literature on modalities of management and outcome of giant intradiploic epidermoids. Method: We report a 68-year-old female presenting with a giant occipital intradiploic epidermoid. We did occipital craniectomy excising the lesion sans the dural adhesion to the torcular region. Postoperatively the patient had abatement of symptoms and had an uneventful follow-up. We also reviewed the pertinent literature on giant intradiploic epidermoids. Results: A total of 47 cases were found, with details of the presentation, management, and outcome available for 37 of them. Male dominance with an average of presentation of 46 years was observed. Headache and swelling both were equally reported as the most common symptom and occipital bone was the most common location of the tumor. All cases underwent craniectomy with tumor excision and no recurrence was seen till the last follow-up available. Conclusion: Giant intradiploic epidermoids are rare tumors. Their clinical presentation and tumor characteristics might be different than smaller intradiploic epidermoids, however, they tend to carry a good prognosis if surgical excision is attempted with respect to the location and its proximity to venous sinuses.

Keywords:- *Intradiploic Epidermoid, Large Intradiploic Epidermoid, Giant Intradiploic Epidermoid.*

I. INTRODUCTION

Epidermoid cysts are extremely slow-growing tumors and comprise about 1 % of the intracranial tumors[1][2][3][4]. They usually occur intradural, however, an extradural occurrence is seen in approximately a quarter of the cases. Common extradural locations seen are the scalp, diploic space of calvarium, sphenoid, skull base, orbit, and paranasal sinuses[3][5][6][7][8].

Intradiploic epidermoid cysts (IEC) are fairly uncommon and account for about 16%-25% of all the cranial epidermoids[4][5]. Giant IECs are even rarer and forms a distinct subgroup owing to their indolent growth pattern, exceptionally large size at presentation, tendency to adhere to the dura, and occasionally intradural extension[3][5]. Moreover, malignant transformation, intracranial hypertension, seizures, and focal neurological signs have been described more commonly in patients with large IECs[9][10]. These characteristics of the giant group of the IECs can limit their surgical excision, predisposing them to a relatively different post-operative behavior and long-term consequences.

We report our experience of a case of giant IEC of the occipital bone. Additionally, we review the literature on all the giant (> 5 cm) IEC case reports and series till now.

II. CASE PRESENTATION

A 68-year-old female presented to our tertiary care center with the complaint of occipital headache for 2 years with gradually increasing intensity and with visual disturbances for 2 weeks in the form of visualization of colored halos. There were no neurological deficits on examination with normal visual acuity and normal visual field. A contrast-enhanced computed tomography (CT) of the head showed a solid cystic extra-axial lesion in the diploic space of the occipital bone eroding its outer and inner table and had sclerotic margins (Fig. 1). Magnetic resonance imaging (MRI) was suggestive of a 91 x 64 x 82 mm sized expansile lesion within the occipital bone appearing hyperintense on the T2-weighted sequence (Fig. 2). The lesion was abutting the torcula, however, all venous channels were patent. We performed a midline occipital craniectomy around the cyst removing the outer table and excising the typical pearly white pultaceous material (Fig.3: A). However, the inner wall was adherent to the dura near the venous sinus due to which we left it behind (Fig.3: B). Postoperatively the patient improved symptomatically. There was no recurrence till the last follow-up at 6 months.

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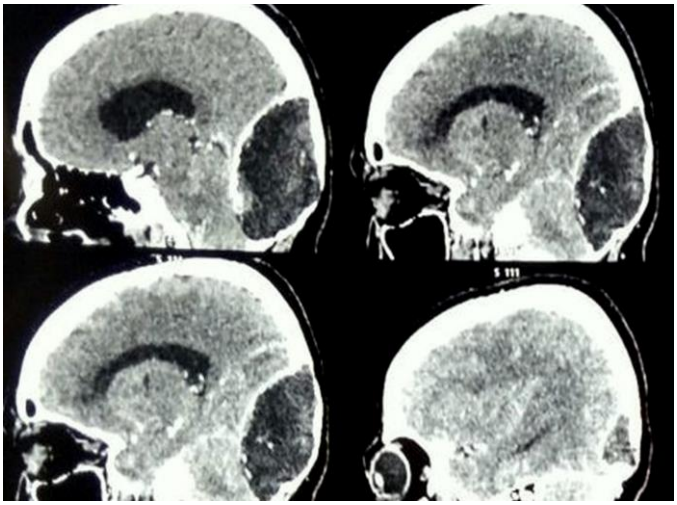

Figure 1: CT brain showing enlargement of the diploic space with erosion of inner table and scalloping.

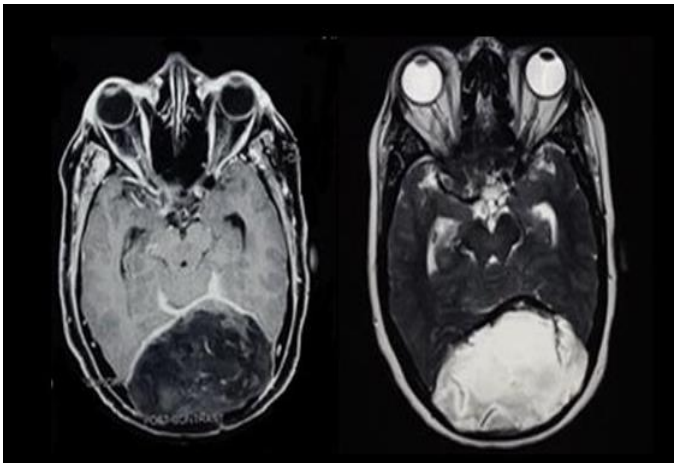

Figure 2: MRI brain T1W with contrast showing non-enhancing circumscribed lesion in the occipital bone along with T2 hyperintensity of the lesion in the second image.

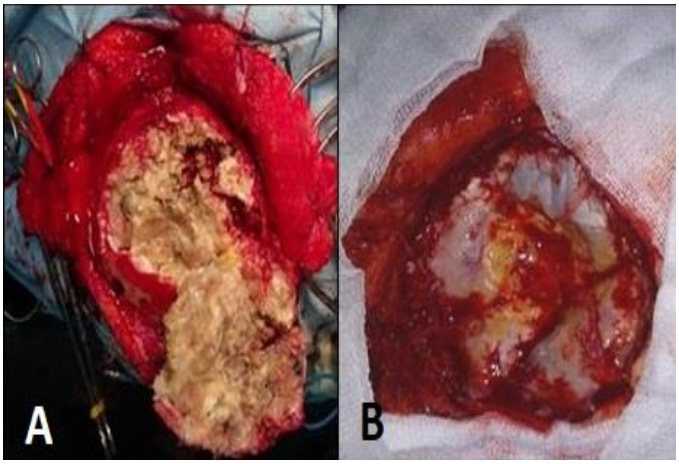

Figure 3: Intraoperative images A. Photograph of the giant epidermoid after removing the outer table, showing pearly white pultaceous material and after excision; B. Post-resection cavity.

III. REVIEW OF LITERATURE

Since there are no well-defined criteria to call an IEC giant-sized, we used the criteria of size to be more than 5 cm in any one dimension and the location to be intradiploic. For our literature review, we searched the PubMed database using the terminologies "Giant intradiploic epidermoid", 'intradiploic epidermoid" "giant cranial epidermoid' and "diploic epidermoid". A total of 47 cases were found, with details of the presentation, management, and outcome available for 37 of them (Table 1). The size of the lesion was confirmed based on the CT or MRI findings mentioned.

IV. RESULTS

Inclusive of all the case reports and series, in total 47 cases of IECs have been reported till now. However, 10 cases were reported in series of mixed small and giant IECs without separate description on the giant cases, hence, were not available for review[1][11].

Out of the 37 cases available for review, the Male: Female ratio was 25:12. The mean age was 46 years, and the range was 13 to 81 years. The most common cranial site of occurrence noted was occipital with a total of 11 cases, followed by 9 in the frontal region, 5 in parietal, 3 in frontoparietal, 3 in the orbital walls, 2 in the temporal, and 1 case each in the parietooccipital, sphenoid, fronto-tempero-sphenoid and mastoid region. Our case also is a giant IEC located in the occipital region. In the presentation, headache and swelling were the two most common symptoms, and both were reported in 14 patients each. Other symptoms related to the local mass effect were ataxia and vision diminution in occipital and orbital lesions, proptosis in sphenoid or orbital lesions, depression, and hemiparesis in parietal lesions, confusion, and anosmia in basifrontal lesions. Other clinical manifestations seen were diplopia and seizures.

All the cases underwent surgical management. 11 of the case report patients had no follow-up reported, whereas the remaining had a mean follow-up of 14.72 months (range: 2 months to 48 months), and in the case series, the follow-up period ranged from a month to 3 years. None of the cases showed recurrence till the last follow-up reported.

Table 1: Review of Giant IECs: Case reports and series

Author (Year of publication)	Age (years)	Sex	Symptoms	Location	Follow -up	Outcome on f/u
Rengachary et al.[7] (1978)	62	M	HA	Occipital	1 Y	Improved, No recurrence
Constans et al.[9] (1985)	58	M	HA, Hemiparesis	Parietal	8 m	Improved, No recurrence.
	59	M	HA	Temporal	8 m	Improved, No recurrence.
Gupta et al.[12] (1987)	13	M	Swelling, Proptosis	Sub-temporal	No f/u	-NA
Dias et al.[13] (1989)	59	M	HA, Diplopia	Occipital	18 m	Improved, No recurrence.
	29	M	HA, Diplopia	Fronto-parietal	30 m	Improved, No recurrence.
	68	F	HA, Confusion, Anosmia	Basi-frontal	18 m	Improved, No recurrence.
Guridi et al.[14] (1990)	47	M	Swelling	Occipital	No f/u	-NA
Sargent et al.[8] (1993)	69	F	Proptosis	Greater wing of the sphenoid	18 m	Improved, No recurrence.
Di Benedetto et al.[15] (1996)	65	F	Swelling	Frontal	16 m	Symptom-free, No recurrence.
Rumelt et al.[16] (1997)	32	M	Proptosis	Fronto-tempero-sphenoidal	18 m	Improved No recurrence.
Jaiswal et al.[2] (2000) ***(Case series)***	19 -61 (8 patients)	7 M, 1 F	Swelling, pain, cerebellar signs	Frontal (3), Frontoparietal (1), Occipital (1), Parieto-occipital (1), Mastoid (1)	1m to 3Y	No recurrence
Narlawar et al.[17] (2002)	35	F	Swelling	Parietal	No f/u	-NA
Blanco et al.[18] (2001)	22	F	Proptosis	Orbit -lateral wall and roof	18 m	Improved
Maiuri et al.[3] (2004)	60	M	HA	Sub-occipital	4 Y	Symptom-free, No recurrence
Iranmanesh et al.[19] (2006)	22	M	Hypoglobus, Ptosis, HA.	Orbital roof	No f/u	-NA
Cho et al.[5] (2007)	69	M	Swelling, Seizure	Fronto-parietal	1 Y	Symptom-free, No progression of residual tumor
Prat Acín et al.[20] (2008)	44	F	Swelling, HA	Occipital	No f/u	-NA
Duan et al.[21] (2009)	35	F	Incidental diagnosis	Occipital	13 m	Symptom-free, No recurrence
Kumaran et al.[22] (2010)	55	M	HA	Occipital	No f/u	NA
Enchev et al.[23] (2011)	54	M	HA, Ataxia	Occipital	No f/u	-NA
Krupp et al.[24] (2012)	81	M	Swelling	Frontal	No f/u	-NA
Hasturk et al.[25] (2013)	69	M	Swelling	Frontal	No f/u	-NA
Samdani et al.[26] (2013)	24	F	Swelling	Frontal	No f/u	-NA
Živković et al.[10] (2014)	68	M	Depression	Parietal	6 m	Improvement of depression, No recurrence
Lawrence et al.[27] (2015)	47	M	HA, Swelling, Confusion	Parietal	3 m	Symptom-free, No recurrence
Gollapudi et al.[28] (2018)	14	F	Swelling	Frontal	No f/u	-NA
Oommen et al.[29] (2018)	46	F	HA, Swelling	Occipital	2 m	Symptom-free, No recurrence
Ma et al.[30] (2019)	61	F	Swelling, Dizziness, Facial numbness	Parietal	15 m	Symptom-free, No recurrence
Zhang et al.[6] (2021)	64	M	Proptosis, vision loss	Orbital roof, Frontal	2 m	No recurrence

Abbreviations: M- Male, F- Female, HA- Headache, Y- years, m- months, -NA- Not applicable

V. DISCUSSION

Our case represents the giant subgroup of IECs like in other reports. The literature review on the giant IECs demonstrates their common characteristics and clinical course and outcome.

IECs are uncommon tumors with indolent growth patterns. The most accepted theory of their origin is of dysontogenetic etiology, which is believed to be related to the displacement of ectodermal remnants at the time of neural tube closure[31]. Acquired forms can occur secondary to the traumatic or iatrogenic displacement of epidermal tissue into the connective tissue[20][23][31].

The earliest case of IEC was reported by Müller in 1838[32]. And, the earliest report on Giant IEC is by Cushing published in 1922[33]. The latest extensive review on the IECs was done by Arko et al.[34] which included 167 patients, whereas, the last case series on Giant IECs was reported in the year 2000 by Jaiswal et al.[2] which included 8 patients. Ours is the first extensive review on all the giant IECs reported till now.

Arko et al. in their review reported IECs to present among patients between 20 and 59 years of age, with a mean age of presentation as 38.1 years[34]. In our review, for giant IECs the mean age of presentation was 46 years, range 13 to 81 years, with a predilection for diagnosis beyond 3rd to 4th decade. The older age of presentation in the giant group could have been due to very slow growth leading to delayed presentation. However, other factors like decreased awareness, limited access to medical care, and poor socioeconomic status can also lead to delayed presentation.

Male predominance was seen in our study too, with 67.5% (25) and 32.5% (12) cases being men and women, respectively. It is not much different than IECs overall with men: women ratio of 60.5%: 39.5%[34].

IECs commonly involve the frontal (30.5%), parietal (29.3%), occipital (28.7%), and temporal (18.6%) bones[34]. Reports on giant IEC describe them to be either predominantly supratentorial or infratentorial[4][23], with a predilection for the frontal and parietal skull bones[2]. In our review, the occipital bone was the most common site, followed by the frontal. Even in our presented case, the lesion was occipital with extension equally supra and infratentorial, centered around the torcula. The reason for the occipital bone becoming the commonest location for giant-sized IEC could be delayed detection because of the location being out of sight, least perception cosmetically and delayed palpability due to growth under the bulk of suboccipital muscles and occipital hair in older men.

Epidermoids usually present as a painless lump, while headache being the second most common symptom[34]. In our review, headache and swelling, both were equally reported. Again, the reason for swelling being not the commonest presentation could be out of sight location, least perception cosmetically, and delayed palpability due to growth under the bulk of suboccipital muscles and occipital hair, for the occipital lesions. Giant IECs can commonly present with neurological deficits secondary to the local mass effect. Frontal and sphenoid bone epidermoids are commonly associated with proptosis, while occipital bone epidermoids can present with ataxia. Other clinical manifestations seen are signs of raised intracranial pressure, diplopia, and seizures. Rarely some cases may present with seizures due to the intradural extension[5][11]. Our patient had visual hallucinations, which were due to the mass effect on bilateral occipital lobes.

IECs are usually benign tumors, however, malignant changes have been reported in the epithelial lining of intradiploic epidermoid cysts[35]. Some authors suggest that IECs that have undergone multiple operations and repeated inflammation are prone to malignant transformation[5]. This has been one of the reasons for an emphasis on the complete removal of giant IECs[2][3][5][14].

CT scan is the most useful first-line investigation to identify the intradiploic nature of the lesion, as well as its extracranial and intracranial extension[5]. As with this case, intradiploic epidermoid tend to produce expansive non-contrast enhancing lesions. IEC usually involves both diploic tables, however, giant diploic epidermoids are more commonly associated with the destruction of the inner table and intracranial extension[1][9]. MRI can help differentiate epidermoid from other bony lesions, showing hyperintensity on T2-weighted and FLAIR sequences, isointense to CSF on T1 weighted imaging, restriction on diffusion-weighted imaging, and non-enhancement on contrast imaging[25]. Hemorrhage or proteinaceous contents inside the cyst may appear hyperintense on the T1 weighted sequence[25]. Contrast enhancement may be seen on the periphery due to perilesional inflammation and malignant transformation should be suspected in case of contrast uptake by the lesion[25]. MRI can also be useful to identify the intradural and intraparenchymal extension[5][11].

The standard treatment for IEC is total excision along with its capsule. Authors have advocated taking into consideration the patient's age, comorbidity, dural and parenchymal extension, relation to sinuses, and potential neurological damage[14]. If required adhered dura can be excised followed by a duroplasty. In our case, the dense attachment of the cyst near the torcula prevented complete excision. The reason for the dense attachment could be the long duration and giant size of the epidermoid. Fortunately, there was no intradural extension in our case. Timely removal of the lesion is advocated to have a diagnosis and to avoid potential future complications, including intracranial mass effect, intraparenchymal growth, intralesional abscess formation, bleeding, and malignant change. In suspected and proven malignant transformation, extensive excision, radiotherapy, and chemotherapy are advised[35][36].

Removal of the tumor can be difficult if intradural and especially intraparenchymal extension[5]. Like in our case, in difficult locations, limited excision should be attempted to preserve the integrity of the dural venous sinuses, and eloquent parenchyma. Neuronavigation could be useful if an invasion of critical structures is suspected. Cranioplasty is frequently done if there is a large bony defect post excision[23]. Total excision

of these cysts carries a good long-term prognosis[9][14]. A recurrence rate of 8.3–25.0% was seen in cases with incomplete removal[35].

VI. CONCLUSION

Giant intradiploic epidermoid are rare. They tend to carry a good prognosis if surgical excision is attempted with relation to location and proximity to venous sinuses.

CONFLICT OF INTEREST

No special device has been used in this case and no conflict of interest.

FUNDING

No funding was received for this research.

INFORMED CONSENT

Informed consent was obtained from the patient included in the study.

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