Radiology Case Reports 20 (2025) 170–176

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Case Report

Intradiploic epidermoid cyst in the skull: Case report and systematic review ☆

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ARTICLE INFO

Article history:
Received 17 March 2024
Revised 18 September 2024
Accepted 19 September 2024

Keywords:
Intracranial
Skull
Epidermoid
Cysts

ABSTRACT

Intradiploic epidermoid cysts are rare, benign tumors, accounting for less than 1% of all intracranial tumors. Due to their scarcity, we are reporting a case of an intradiploic epidermoid cyst and reviewing several similar cases in the literature to provide a better description of this tumor and analyze its clinical and radiological features. We conducted a search on the PubMed database for studies published between January 2010 and February 2023, including studies of nontraumatic and noniatrogenic calvarial cysts. We found a total of 34 studies and 41 patients, with an average patient age of 42.5 years. Common symptoms included painless masses, headaches, and seizures. The occipital, frontal, and parietal regions were the most common locations for these tumors. Some patients experienced complications such as otitis media, infection, or extension into the orbit and brain. Surgical resection was performed in all cases except for 2 patients with contraindications to surgery. Histological examination confirmed the diagnosis of an intradiploic epidermoid cyst. These cysts are rare skull lesions and can potentially cause neurological deficits due to their mass effect. If incompletely resected, they may also undergo malignant transformation. No single institution has accumulated enough cases to describe this rare tumor accurately. Therefore, through this review, we aim to highlight this tumor's epidemiological, clinical, and radiological features.

Introduction

Epidermoid cysts are benign tumors occurring between the 2 tables of cranial bones, accounting for less than 1% of all intracranial tumors [1]. They are typically found in the cerebellopontine angle, petrous apex, and suprasellar region, with the diploe being a less common and sporadic location [2]. First described by Muller in 1838, these cysts have since been reported sporadically in small groups of patients, originating

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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https://doi.org/10.1016/j.radcr.2024.09.102

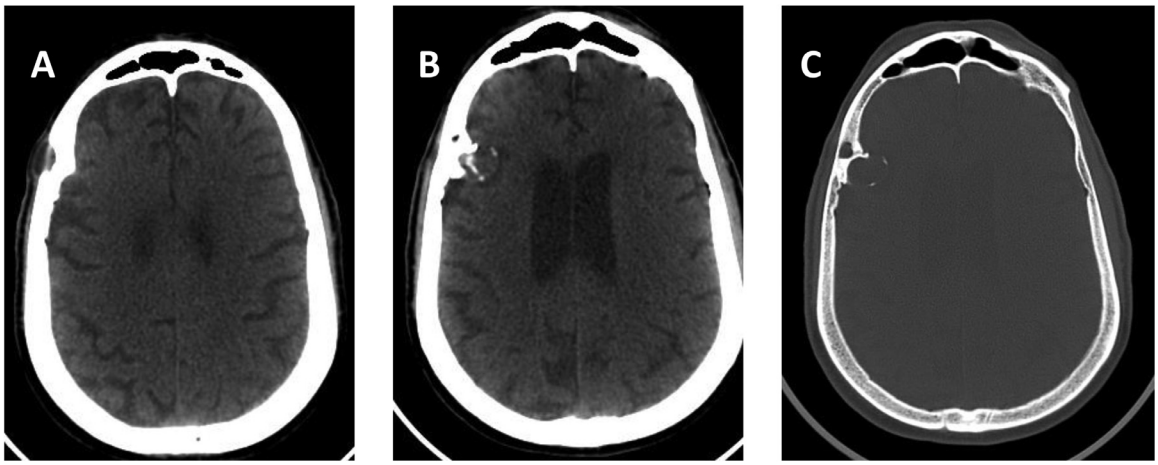

Fig. 1 – Computed Tomography scan axial images on parenchymal (A) and bone reconstruction (C) showing an extra-axial round-shaped lesion, hypodense, with margined calcifications located on the calvarial frontal right bone, associated with condensation of the inner table and erosion of the outer table.

from ectodermal cell remnants within the cranial bones during embryonic development [3]. Neurological signs vary depending on the tumor's location, size, and mass effect on adjacent anatomical structures [4]. Diagnosis is usually suggested through imaging and confirmed by histology. For a good prognosis, accurate radiological assessment and complete excision of the tumor are mandatory. Due to the scarcity of epidermoid cysts, we reviewed numerous cases in the literature to better describe this tumor and analyze its clinical and radiological features.

Clinical report

A 72-year-old man with a medical history of hypertension and diabetes presented to the emergency department with dysarthria, headache, and seizure. On admission, he exhibited loss of consciousness with a Glasgow Coma Scale score of 9/15. Laboratory analyses showed no abnormalities. A computed tomography scan (CT-scan) revealed an extra-axial, round-shaped, hypodense lesion with margined calcifications over the right frontal area, associated with condensation of the inner table of the frontal bone and erosion of the outer table, measuring 19 × 16 mm (Fig. 1). Further examination with magnetic resonance imaging (MRI) of the brain revealed a well-demarcated extra-axial lytic lesion measuring 20 mm × 15 mm in the right frontal bone with erosion of the inner table. The lesion appeared hyperintense on T1-weighted and T2-weighted imaging and FLAIR, with no postcontrast enhancement.

Additionally, the lesion exhibited a hyperintense signal on diffusion-weighted imaging (DWI) with restriction of apparent diffusion coefficient (ADC), causing slight focal brain compression. Another well-circumscribed, subcutaneous frontal lesion adjacent to the described lesion was identified, showing isosignal on T1-weighted imaging and hypersignal on T2-weighted imaging and FLAIR, with no postcontrast enhancement. This lesion also demonstrated a hyperintense signal on DWI with restriction of ADC, measuring 19 × 9 mm (Fig. 2). The patient was scheduled for craniotomy with tumor excision; however, he experienced a pulmonary embolic episode. Considering his age, the absence of neurological deficits, the embolic episode, and the risks associated with the total removal of this giant tumor followed by cranioplasty, the patient did not undergo surgery.

Systematic review

Study selection and search strategy

We searched the PubMed database on February 12, 2023, to identify studies meeting the specified criteria. We included all studies published between January 2010 and February 2023, with available full texts in either French or English. The MeSH (Medical Subject Heading) terms used are Intracranial, Skull, Epidermoid, and cyst. Our focus was epidermoid cysts arising in the diploe of the cranium and on nontraumatic calvarial cysts, excluding those of iatrogenic origin and articles primarily addressing surgical management. Some articles were excluded bases on title, abstract or full text screening, resulting in the inclusion of a total of 34 studies (Fig. 3). Patient information was systematically collected and consolidated to provide data on age, gender, presenting symptoms, and radiological findings. While most authors reported on patient age, sex, and symptoms, radiological findings varied across studies.

Results

A total of 34 studies and 41 patients were included in the literature review (Table 1), The age of presentation varied greatly among patients with most presenting between 21 and 60

Table 1 – Results of our study selection.

Study	cases	Age	sex	Symptoms	Location	Surgery	Complication
Khalid et al., Pakistan, 2021 [5]	1	18	M	Subcutaneous swelling	Parietal	yes	Infection
Kuwano et al., Japan, 2020 [2]	1	49	F	earache fever effusion	Occipital	yes	Otitis media
Turk et al., Turkey, 2018 [6]	6	39 30 34 23 43 76	5F 1M	Soft scalp mass Headache	2 Occipital 3 Parietal 1 frontal	yes	Nothing
Dąbrowski et al., Paland, 2018 [3]	1	56	M	Dizziness Balance disturbances Memory disorders	Temporal and occipital bone	yes	Nothing
Krupp et al., Germany, 2012 [7]	1	81	M	Painless swelling	Frontal and parietal	biopsy	Nothing
Dong Hee Lee, Korea, 2011	1	25	M	Hearing loss	Temporal	yes	Nothing
Choo et al., Korea, 2021 [8]	1	57	F	Headache	Occipital	yes	Nothing
Catapano et al., USA, 2022 [9]	1	71	M	Dizziness Otorrhea	Occipital	yes	Infection
Oommen et al., India, 2018 [1]	1	45	F	Headache	Occipital	yes	Nothing
Nakamoto et al., Tokyo, 2013 [10]	1	56	F	Suspicion of metastasis	Temporal	yes	Nothing
Hasturk et al., Turkey, 2013 [11]	1	69	M	Mass	Frontal	yes	Nothing
Zheng et al., China, 2018 [12]	1	54	M	Headache confusion	Frontal	yes	Nothing
Tica et al, Romania, 2021 [4]	3	12 71 78	F	Mass Headache Dizziness Balance disorder	2 Frontal 1 Parietal	Yes/yes/ no	Nothing
Turkoglu et al.,Turkey, 2010 [13]	1	60	M	Mass Headache Nausea Dizziness	Occipital	yes	Nothing
Akbaba et al., Turkey, 2012 [14]	1	4	M	Non-healing painless ulcer on the left upper eyelid	Frontal	yes	Fistulization to eyelid
Mandelbaum et al., USA, 2017 [15]	1	16	M	Mass	Frontal	yes	Nothing
Gollapudi et al.,India, 2018 [16]	1	14	F	Mass	Frontal	yes	Nothing
Khan et al., Pakistan, 2011 [17]	1	50	M	Headache	Occipital	yes	Nothing
Dhull et al., India 2014 [18]	1	14	M	Nothing	Occipital	yes	Nothing
Lawrence et al., UK, 2016 [19]	1	47	M	Headache confusion	Parietal	yes	Nothing
Yildiz et al., Turkey, 2015 [20]	1	1	F	Mass	Frontal	yes	Nothing
Antoniades et al., Greece, 2021 [21]	1	58	M	Exophthalmia	Frontal	yes	Intra-orbital extension
Moreira-Holguin et al., Mexico, 2015 [22]	2	42 46	M	Intracranial hypertension	Frontal Occipital	yes	Nothing
Pagkou et al., Italy, 2021 [23]	1	24	M	Intracranial hypertension	Occipital	yes	Diplopia and torcular compression
Gadgil et al., USA, 2013 [24]	1	47	M	Headache	Parietal	yes	Hemorrhage post traumatic
Oumniya Abouhanine et al., Morocco, 2020 [25]	1	69	M	Headache	Temporal	yes	Recurrence
Arko et al., USA, 2016 [26]	1	47	F	Seizure	Parietal	yes	Nothing
Kumaran et al., India, 2010 [27]	1	55	M	Headache	Occipital	yes	Nothing
Wu et al., China, 2013 [28]	1	37	M	Seizure	Occipital	yes	Nothing
Weingarten et al., USA, 2020 [29]	1	7	M	Mass	Frontal	yes	Nothing
Yılmaz et al., Turkey,2016 [30]	1	27	M	Mass	Occipital	yes	Nothing
Burnham et Lewis, USA, 2016 [31]	1	55	M	Incidental	Frontal	yes	Orbital and cranial extension
Law et al., China, 2015 [32]	1	37	M	Unsteady gait	Occipital	yes	Nothing
Our Case	1	72	M	Headache Seizure	Frontal	No	Pulmonary embolic

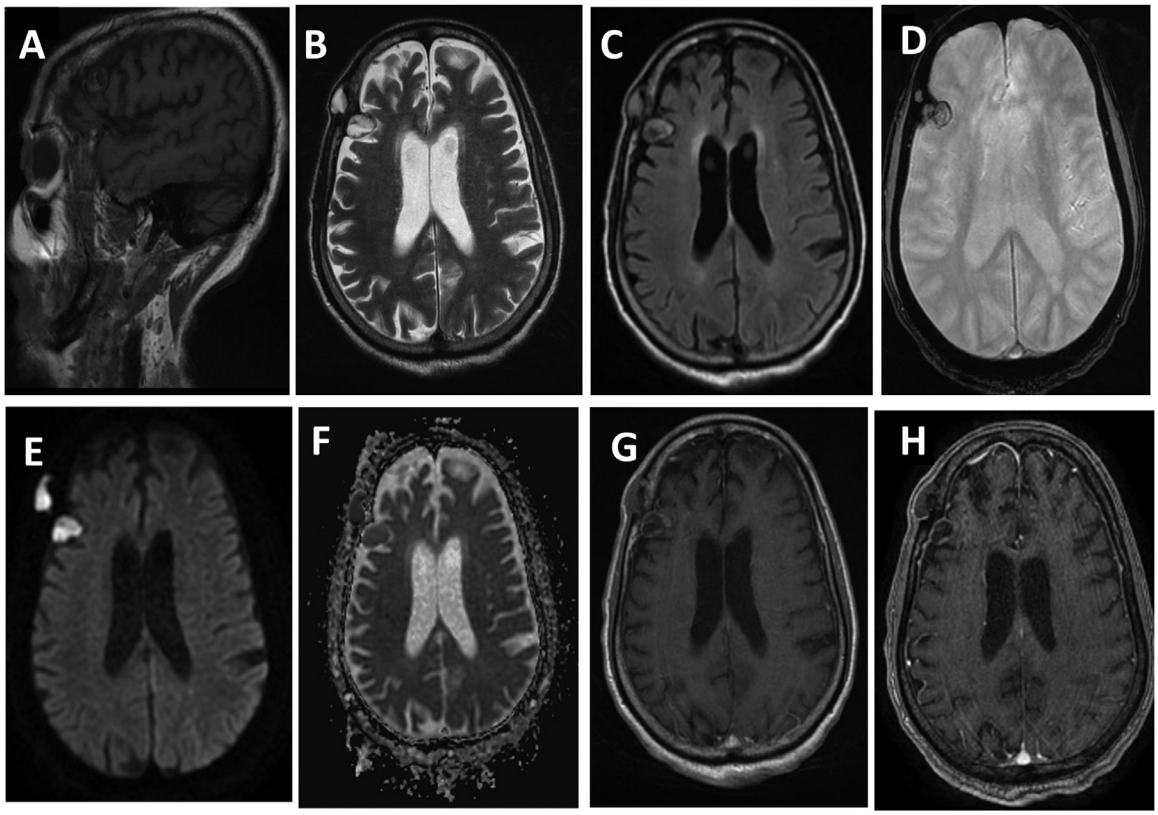

Fig. 2 – Cerebral MRI on sagittal and axial images showing 2 adjacent well-demarcated extra-axial lytic lesions in the right frontal bone with the erosion of the inner table, appearing as hyperintense on T1 WI (A), hyperintense T2 WI (B), inhomogeneous hyperintense on FLAIR sequence (C). Additionally, they exhibit a blooming artifact on the T2* sequence, indicating the presence of calcification (D). Moreover, the lesions appear hyperintense on diffusion-weighted imaging (DWI) and show restriction of Apparent Diffusion Coefficient (E-F). Furthermore, there is no enhancement observed after Gadolinium administration (G-H).

years. The mean age of our patients was 42.5 with a standard deviation 21 years (minimum 1 year, maximum 81 years).

Between our patients we had 15 females (36,6 %) and 26 males (63,4 %).

The literature review comprised a total of 34 studies involving 41 patients. The patients age at presentation varied widely, with the majority presenting between 21 and 60 years old. The mean age of our patient cohort was 42.5 years, with a standard deviation of 21 years (range: 1-81 years). Out of our patients, 15 were female (36.6%) and 26 were male (63.4%).

The symptoms varied among patients. The most common findings were a painless mass, headache, or seizure. However, some patients were asymptomatic or presented with complications such as otitis or infection.

The site of the intradiploic epidermoid cysts was evaluated. The occipital bone was involved in 15 patients (36.6%), the frontal bone in 13 patients (31.7%), the parietal bone in 8 patients (19.5%), and the temporal bone in 3 patients (7.3%). Two patients had cysts in multiple locations: one in the temporal and occipital bones, and one in the frontal and parietal bones.

All our patients presented with identical radiological appearances. The epidermoid cysts appeared as well-defined expansive lesions, hypodense on CT-scans, with thinning or erosion of the inner or outer table of the calvarium, and possible peripheral calcifications. On MRI, the cysts showed iso- or hyposignal on T1-weighted images and hypersignal on T2-weighted images. The lesions exhibited hyperintense signals on DWI with restricted ADC. No enhancement was observed in the lesion or peripherally after contrast administration. Some lesions displayed heterogeneous signals with areas of hyperintensity on T1-weighted images or hypointensity on T2-weighted images.

Nine (22%) of our patients presented with complications: diplopia and torcular compression in 1 patient, fistulization to the eyelid in 1 case, bleeding after a traumatic episode in 1 case, infection in 2 cases, intraorbital or intracranial extension in 2 cases, otitis media with the erosion of the mastoid in 1 case, and recurrence in 1 patient. Malignant transformation was not found in any of our patients.

All patients underwent surgical resection except for two who had contraindications to surgery; one of these patients underwent a biopsy. Histological examination showed a squamous-lined cyst with keratinization, confirming the diagnosis of an intradiploic epidermoid cyst. The follow-up for all our patients was uneventful.

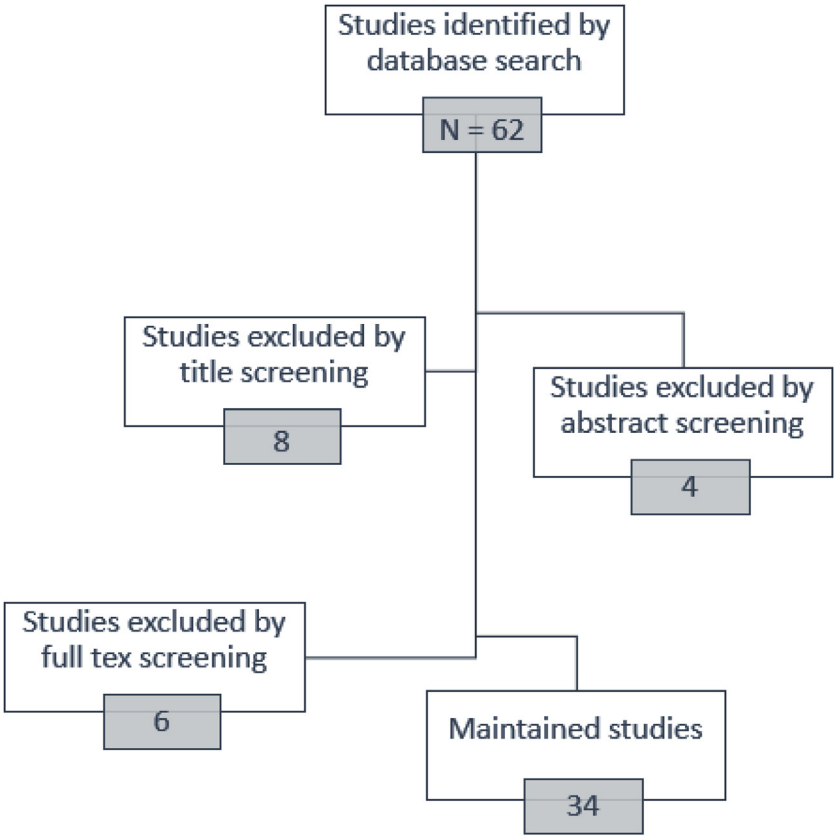

Fig. 3 – Algorithm of study selection. We conducted a search of the PubMed database and identified 62 studies. We excluded articles related to iatrogenic-origin cysts and those primarily addressing surgical management. In the end, we included 34 studies.

Discussion

Intracranial epidermoid cysts are rare brain tumors, with intradiploic location being particularly uncommon. Two mechanisms of pathogenesis are suggested: they may originate from ectodermal epithelial remnants from embryonic development or trauma-trapping epidermal tissue in the diploe [1,3].

Various cases have been reported in the literature, highlighting different presentations and management approaches. Turk et al. discussed the association between these cysts and older age, proposing that age-related cranial bone changes might predispose individuals to these lesions [6]. In another report, Krupp et al. described a giant intradiploic cyst with extensive osteolytic changes, emphasizing the aggressive nature these cysts can sometimes exhibit [7]. Other studies have documented cases with unusual features, such as intracranial extension, spontaneous fistulization, and even coexistence with other pathologies like hematomas or craniosynostosis, as seen in cases by Choo et al., Nakamoto et al., and Yildiz et al. [8,10,20]. Authors such as Hasturk et al., Zheng et al., and Turkoglu et al. have reported on giant cysts that posed surgical challenges due to their size and proximity to critical anatomical structures [11–13]. Unique presentations, such as hemorrhagic cysts and those with orbital or intraorbital extension, have also been reported, adding to the variability in clinical manifestation of these cysts [21,23,27]. Rarely, they can mimic other conditions, such as metastases or fibrous dysplasia, complicating diagnosis as illustrated by Dhull et al. and Mandelbaum et al. [15,18]. As the literature demonstrates, treatment often involves surgical excision to relieve symptoms and prevent recurrence, as in cases presented by Pagkou et al., Kumaran et al., and Law et al., who highlighted the effectiveness of complete resection in reducing recurrence risk [23,27,32]. These cases collectively contribute to the growing body of knowledge on intradiploic epidermoid cysts, underscoring the importance of recognizing their diverse clinical and radiological characteristics to ensure accurate diagnosis and appropriate management [14,17,19,24,28–31].

Our review confirms that these cysts predominantly affect males in their third or fourth decade of life, consistent with existing literature. For example, Arko et al. reported a mean patient age of 38 years and a male predominance [26]. These slow-growing lesions often present with symptoms related to tumor size, such as headaches, seizures, or neurological deficits. Frequently, they are asymptomatic and diagnosed incidentally.

In terms of location, our findings align with the literature, noting a prevalence in the occipital and frontal bones. Arko et al. also identified the frontal, parietal, and occipital bones as common sites. On CT, these cysts appear as low-density le-

sions with a sclerotic border and no enhancement. MRI is preferred for imaging, typically showing low or iso signal on T1-weighted images and high signal on T2-weighted images, with variable FLAIR signal, depending on the protein concentration. DWI is particularly useful for differentiation epidermoid cysts from other differential diagnoses [4,9,26].

Our case report differed in MRI findings, showing a heterogeneous hyperintense signal on T1-weighted images. Differential diagnoses include dermoid cysts, osteolytic metastasis, cystic tumors, arachnoid cysts, calvaria cavernous hemangiomas, aneurysmal bone cysts, fibrous dysplasia, and plasmacytoma [1].

Complications of intradiploic epidermoid cysts can include spontaneous rupture, leading to aseptic meningitis, as well as infection, otitis, and recurrence. Malignant transformation should be suspected if rapid growth or new neurological symptoms arise, potentially due to cyst rupture or chronic inflammation [33,34]. Our review found no cases of malignant transformation.

The treatment of choice is early surgical excision, including removal of the cyst capsule. Large cysts may require cranioplasty, and complete excision is associated with a favorable long-term prognosis. Incomplete resection can lead to recurrence or malignant transformation [5,16,22].

Our study's limitations include focusing solely on non-traumatic and noniatrogenic cysts documented in the literature.

Conclusion

Intradiploic epidermoid cysts are rare and benign skull lesions. Computed tomography (CT) and magnetic resonance imaging (MRI) of the brain help diagnose these cysts and evaluate their surgical resectability. Total removal of these cysts is associated with a very good long-term prognosis and helps prevent complications such as infection, malignant degeneration, or recurrence.

Ethics statement

Ethics approval was not obtained because this manuscript is a case report.

Patient consent

Written informed consent was obtained from the patient.

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