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Primary Intracranial Ewing Sarcoma: Case Series and Review of Literature

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Abstract: ***Primary intracranial extraosseous Ewing sarcoma is one of those rare and elusive tumours that often hides behind the mask of more common intracranial lesions, making timely diagnosis particularly challenging. In my view, what makes this condition so striking is not just its aggressive nature but also the ease with which it can be misinterpreted as central primitive neuroectodermal tumours or even benign findings such as hematomas. This paper presents three diverse cases two in children and one in an adult where the tumour manifested in different ways, from dural-based masses infiltrating bone and muscle to deep-seated lesions mimicking diffuse gliomas. It is evident that radiological appearances alone could not provide clarity, and only a combination of histopathology, immunohistochemistry (especially CD99 expression), and molecular insights into the EWSR1 rearrangement made the diagnosis possible. That said, the treatment outcomes varied depending on the extent of surgical resection and the integration of radiotherapy or chemotherapy, which highlights the importance of a multidisciplinary approach. Taking this further, one cannot ignore the sobering reality that despite advances in surgical precision and supportive care, prognosis often remains guarded, with recurrence and neurological complications being frequent hurdles. This suggests that while the current multimodal strategies bring hope, there is still much ground to cover in understanding the biological behaviour of this tumour. In essence, these cases not only reinforce the diagnostic complexity of intracranial Ewing sarcoma but also remind us of the pressing need for tailored treatment protocols that balance aggressive intervention with long-term quality of life.***

Keywords: Ewings sarcoma, round cell tumours

1. Introduction

Ewing sarcoma family of tumours (ESFT) are small round cell tumours with similar histopathology, immunophenotype, and genetic features. They are aggressive, malignant tumours with a high tendency to bleed.[1] ESFT can be divided into: Ewing sarcoma of the bone (ESB), Extraskeletal Ewing sarcoma (EES), Peripheral primitive neuroectodermal tumour (pPNET).[2] About 85–90% of patients with ES/pPNET show a chromosomal translocation t(11;22) (q24;12), leading to the fusion of the gene EWSR1 and the ETS family gene FLI1.[3] PNETs and Ewing's sarcoma are part of the same morphological spectrum, with pPNET showing neuroectodermal differentiation.[4] The main difference between pPNETs and Ewing's sarcoma is the degree of neuroectodermal differentiation. Ewing's sarcoma shows absent or limited neuroectodermal differentiation, whereas pPNETs demonstrate more neuroectodermal features.[5] Primary Intracranial Extraosseous Ewing Sarcoma (ES) is a rare and aggressive malignant tumor that typically affects children and adolescents.[6-10] James Ewing first described this tumor in 1921, differentiating it from lymphoma and other cancers.[11] Ewing sarcoma is the second most common malignant bone tumor in children. It often develops in the long bones' cortex; however, it can also happen in the ribs and vertebrae.[12] Common locations for extraosseous Ewing's sarcoma (EES) include: the soft tissues and bones of the lower extremity, paravertebral region retroperitoneal regions.[13-14] EES rarely presents as a primary intracranial lesion.[12] Primary Intracranial Extraosseous ES accounts for only 1–4% of all extraosseous Ewing's sarcoma.[14-16] When found in the CNS, it most commonly arises as a solitary lesion from the dural surface of the brain or the spinal cord. It can be misdiagnosed as central primitive neuroectodermal tumor (PNET), other intracranial lesions, or even an epidural hematoma. The distinction between Ewing sarcoma and PNET is important because they have different treatments and prognoses.[17] However, these two tumors share the same characteristic cytogenetic features and are categorized into the same tumor family.[18] They are both small, round cell neoplasms and are highly malignant. We report 3 cases of Primary Intracranial Ewing Sarcoma treated at our center.

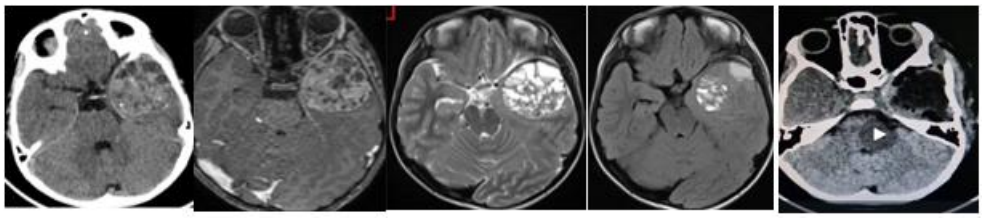
2. Case Details

Case 1:
8y/Male child presented with a history of headache in the last 15 days, MRI revealed a 6.1×6.2×5.2 cm round-shaped extra-axial mass-like lesion Left temporal which extends into the intracranial space- into the temporal lobe and extracranial- into the maxillary and ethmoidal sinus, displaying heterogeneous enhancement. The patient underwent left temporal craniotomy and a dural-based lesion, which was heavily vascular, extending into the skull base to the infratemporal fossa by destruction of skull base bone. The tumour is pinkish, spongy, with ill-defined planes to the surrounding brain structure. Intra dural lesion was seen, it was

moderately vascular, densely adherent to the dura, non-suckable, pinkish.

Histopathological examination revealed uniform small round to oval undifferentiated cells with hyperchromatic nuclei and scant basophilic cytoplasm. Immunostaining revealed positive reactivity and a membranous pattern for CD99 the diagnosis of dural-based ES.

Postoperatively, the patient was free of neurological deficits. Postoperative CT and MRI revealed no evidence of remnant enhancing lesion with implying complete tumour removal patient underwent conventional radiotherapy.



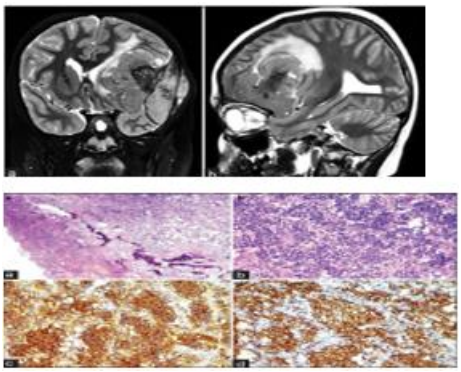
Case 1 Images- MRI showed a 6.1×6.2×5.2 cm left temporal extra-axial mass extending intracranially into the temporal lobe and extracranially into the maxillary and ethmoidal sinuses, with T1 hypointensity, variable T2 signals, and heterogeneous gadolinium enhancement. CT revealed a heterogeneous hypo- to iso-dense mass without calcification or bony erosion

Case 2:
7y/Male child presented a swelling in the left temporal region with associated headache for 20 days. History of projectile vomiting and diplopia. The neurological examination revealed Left lateral rectus palsy and right-sided paucity.

The patient underwent emergency left FTP craniotomy. Intraoperatively temporalis muscle and temporal bone were infiltrated with tumor, and it was highly vascular, non-suckable, and hard. Part of the muscle, bone, and dura were removed along with the tumor. Gross total resection was done.

Histopathological examination revealed uniform small round to oval undifferentiated cells with hyperchromatic nuclei and scant basophilic cytoplasm. Immunostaining revealed positive reactivity and a membranous pattern for CD99, the diagnosis of bone-based ES.

Postoperatively, the patient was free of neurological deficits. Postoperative CT and MRI revealed no evidence of remnant enhancing lesion with implying complete tumour removal patient underwent chemotherapy.



Case:2 images
Large well-defined, lobulated, extra-axial mass lesion measu ring ~8 × 5 × 7.5 cm along the left frontal and parietal region s
(a) - Overlying bone shows tumour infiltration. (b) Monomo rphic tumour cells arranged in nests and lobules with focal cy toplasmic clearing. (c) - Diffuse bright membranous immuno -positivity of CD99 antibody. (d) - Diffuse bright membrano us immuno-positivity of NKX2.2 antibodies

Case 3:
A 30-year-old female presented with a history of dizziness for 1.5 months and left hemi cranium headache for 1 month with features of raised intracranial pressure, with no history of bladder or bowel involvement, no cerebellar involvement, and no autonomic nervous system involvement.

MRI BRAIN revealed Ill ill-defined T2/FLAIR hyperintense, T1 hypointense intra-axial lesion centered in the left insula with a size of 7x7 cm involving adjacent frontoparietotemporal lobes, causing their expansion. No bloomin' on SWI/ diffusion restriction. Post-contrast images were not available in the provided films. Imaging findings s/o neoplastic etiology - diffuse infiltrative glioma.

The patient underwent left frontotemporal craniotomy assisted by neuronavigation. Middle frontal cortisectomy. The tumor shows a Greyish, firm tumor. Not well demarcated from surrounding tissue. Subtotal resection of the tumour.

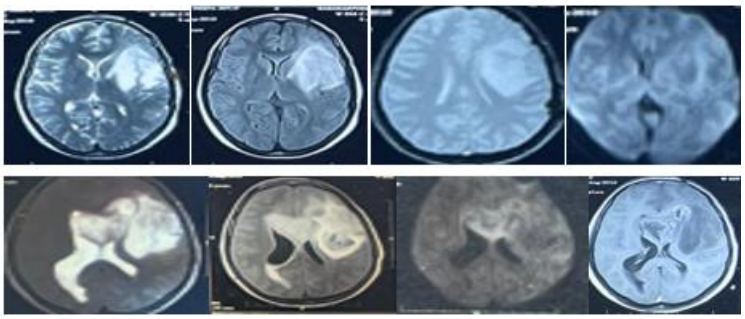
Post op CT and MRI revealed residual lesion. she was presented to emergency after months GTCS 3-4 episodes/month for 3 months and left hemicranial headache for 14 days with features of raised intracranial pressure and MRI Brain reveled there is progression of the lesion with involvement of corpus callosum and cpntralateral periventricular white matter. Areas of diffusion restriction with patchy enhancement and necrotic areas are present in the left temporal region.

The patient underwent emergency re-exploration. Partial frontal lobectomy performed (inferior frontal gyrus preserved). Tumour Characteristics and Extent Lesion was extending posteriorly to the corpus callosum with extensive invasion. Medially limited by falx; anteriorly by frontal lobe. The lesion was described as pale, soft, spongy, and moderately vascular. No well-defined plane between the tumour and normal brain parenchyma.

Histopathological examination revealed uniform small round to oval undifferentiated cells with hyperchromatic nuclei and scant basophilic cytoplasm. IHC of NKX2.2 showed strong and diffuse positivity in tumor cells, features that are suggestive of Ewing Sarcoma.

Late post op period, she had an episode of seizure with altered sensorium. NCCT was performed, suggestive of hydrocephalus with periventricular ooze. She underwent a right medium-pressure ventriculoperitoneal shunt.

During the remaining post-operative period, she gradually improved and was discharged in satisfactory condition.



Case 3 images: MRI showed a 3.7×2.7×2.6 cm left insular lesion, T2 hyperintense, T1 hypointense, with edema, MCA encasement, and central enhancement with peripheral hypo enhancement, MRI showed a left insular T1 hypointense, T2 hyperintense lesion extending into the corpus callosum and frontal white matter, with perilesional edema, diffusion restriction, and heterogeneous contrast enhancement

3. Discussion

Ewing sarcoma (ES) was first described by James Ewing in 1921.[12] ES commonly affects children and young adults. In 1969, Tefft et al. described a series of 5 patients with soft tissue tumours that histologically resembled ES but did not have any apparent bone involvement. This entity is now known as extra skeletal Ewing sarcoma (EES).[14] ES has a peak incidence at 15 years of age. EES commonly occurs in the second and third decades of life, with a slight male predilection. The mean age of presentation of meningeal Ewing sarcoma is 20 years, with a standard deviation of 13 years. 70% of ES cases occur in the first 2 decades of life.

The frequency of ES in the western hemisphere has been noted at 1–3 per million. ES commonly arises in the cortex of long bones but can also occur in other locations, like the ribs and vertebrae. Primary intracranial ES, or CNS-EES, is very rare. When in the central nervous system (CNS), it most commonly presents as a solitary lesion originating from the dural surface of the brain or the spinal cord.[12] Primary intracranial ES is usually intraparenchymal. The defining characteristic of the Ewing family of tumours is the presence of specific chimeric transcripts due to a chromosomal translocation. The translocation t (11;22) (q24; q12) is considered pathognomonic for ES and occurs in about 85%–90 % of ES patients.[19-20]

This translocation causes fusion of the gene EWSR1 on chromosome 22 with the ETS family gene FLI1 on chromosome 11. Other translocations seen in ES and intracranial ES, although less frequently, include: t(21;22)(q22;q12), fusing EWSR1 with the ERG gene on chromosome 21 (17,20); t(7;22)(p22;q12), fusing EWSR1 with the ETV1 gene (1,21); t(17;22)(q21;q12), fusing EWSR1 with the E1AF gene.[21] These chromosomal translocations lead to the formation of chimeric proteins, most notably EWS-FLI1 or EWS-ERG, that are thought to contribute to the development of ES.[22] The exact mechanism by which the fusion proteins lead to tumorigenesis is not fully understood, but it is thought that they disrupt normal cellular processes such as cell growth, differentiation, and apoptosis. The EWS-FLI1 fusion gene codes for transcripts that may be detected by RNA-based polymerase chain reaction assay, and the chimeric protein product is capable of transforming NIH 3T3 cells in vitro.[23] Occasionally, EWSR1 fusions with non-ETS gene family members are also seen. Immunohistochemistry is also used to distinguish intracranial ES from central PNET. Immunostaining for CD99, encoded by the MIC2 gene, is usually positive in CNS-EES and negative in most central PNET. However, CD99 is not a specific marker and can also be expressed in some other CNS tumours. Distinguishing between central PNET and peripheral PNET or intracranial ES is important because these tumour types have different clinical behaviours, treatment, and prognoses. Primary intracranial Ewing sarcoma/peripheral primitive neuroectodermal tumor (pPNET-ES) and central primitive neuroectodermal tumor (cPNET) are distinct entities requiring different treatment approaches and carrying different prognoses.[24-25] Although both tumours consist of undifferentiated small round cells, several key differences exist:

Histogenesis:

- CPNETs are derived from progenitor cells found in the central nervous system (CNS), such as subependymal matrix cells or the cerebellum's external granular layer. Conversely, it is believed that pPNET-ES originates from mesenchymal or neural crest cells that are not part of the central nervous system.

Molecular Characteristics:[26]

- pPNET-ES is characterized by the presence of the EWSR1 gene rearrangement, most commonly the t(11;22)(q24;q12) translocation. This translocation results in the fusion of the EWSR1 gene with the FLI1 gene, leading to the formation of the EWS-FLI1 fusion protein, a chimeric transcription factor that plays a crucial role in tumour development and progression.

- cPNETs do not exhibit the EWSR1 gene abnormality or express the MIC2 gene. Instead, cPNETs frequently show isochromosome 17q and MYC gene amplifications.

Immunohistochemistry:[26]

- pPNET-ES demonstrates strong membranous expression of the cell surface glycoprotein CD99, encoded by the MIC2 gene, in almost all cases.
- cPNETs are typically negative for CD99 staining.
- Treatment:[17]
- pPNET-ES: Treatment involves surgical resection followed by adjuvant multiagent chemotherapy and focal radiotherapy to the primary site and any areas of metastatic disease.
- cPNET: Treatment also involves a multimodal approach with surgery, chemotherapy, and radiotherapy. However, the specific protocols differ, as cPNET tends to spread along the neuraxis more frequently. Therefore, treatment typically involves complete craniospinal irradiation followed by a focal boost.

Prognosis:[28] Whilst both pPNET-ES and cPNET are aggressive malignancies, long-term disease-free survival has been reported in some cases of pPNET-ES, which is uncommon in cPNET. When the tumor originates from tissues inside or around the central nervous system, the prognosis for pPNET-ES may be better than that of cPNET. Both tumor forms, however, often have a poor prognosis and a high likelihood of recurrence.

Table 1: Key Differences between pPNET-ES and cPNET

Feature	pPNET- ES	cPNET
Histogenesis	Neural crest or mesenchymal origin	Precursor cells within the CNS
Molecular Characteristics	EWSR1 gene rearrangement (e.g., t(11;22))	Isochromosome 17q, MYC gene amplification
Immunohistochemistry	Strong CD99 expression	Negative for CD99
Treatment	Surgery, focal radiotherapy, and chemotherapy	Surgery, craniospinal irradiation, chemotherapy
Prognosis	Potentially more favourable than cPNET	Generally poor

Table 2: Characteristics and treatment modalities of studies with case series of three or more patients with primary intracranial Ewing sarcoma.

Author	No. of Subjects	NO. of Patients with Metastatic Disease	Mean Age (Range) in Years	Mean Follow-up (range) in Months	Uniform Chemo Therapy Protocol	No. of Patients Treated with CT	Number of Patients with SR	NO. of Patients with Radical SR	No. of Patients with Marginal SR	NO. Of Patients with RT alone	No. of Patients with SR and RT	LR Rate	Survival
						Primary intracerebral							
Chen et al. [12]	14	3	14.1	30.1 (6 84)	No	10	14	7 (50%)	7 (50%)		9	71.4	5-year OS 19%
Colak et al.[13]	4		13.8 (6-26)	32.0 (11-69)	Yes	4	4	3 (75%)		1	3		100%: mean 32 months (11—69)
Yang et al.,[14]	4		10 (5-16)	49.5 (12-126)	No	4	4				4		25% died (36 months) 75% alive: mean 54 (12—126) months
Singh et al.[15]	7		13 (7-21)	26.9 (12-48)	yes	2 (+1 pat. after LR)	7				4	14%	570/0 died, meaning 33.2 months PFSn = 3: 233 months
Jingetal.[16]	8	3	15 (7-23)				8				8		
Keetal. [17]	3		19.3 (15-28)		Yes	3	3	2	1		3	66.7 %	n = 1: years n = 2 with LR: lost to f.u.
Vanden Heuvel et al. [18]	3		21.8 (2.4-61)		No	2	3				1		n = 1: >5 years n = 1: months n = 1: lost to f.u

4. Conclusion

Primary intracranial Ewing sarcoma/peripheral primitive neuroectodermal tumour (pPNET-ES) is an extremely rare, aggressive malignancy. It is more common in children and young adults and is often misdiagnosed as other brain tumours. pPNET-ES requires a combination of radiological findings, histopathological examination, and molecular genetic analysis, including immunohistochemical staining for CD99 and detection of the EWSR1 gene rearrangement for diagnosis. Treatment often involves a multimodality approach, with surgery, radiotherapy, and chemotherapy. The prognosis of this rare malignancy is often poor.

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