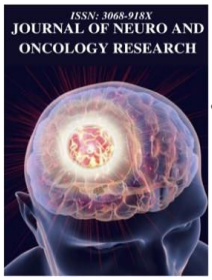

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Case Report

Papillary Glioneuronal Tumor of the Left Parieto-Occipital Lobe Presenting with Diplopia in an 11-Year-Old Girl

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Abstract

Papillary Glioneuronal Tumor (PGNT) is a rare mixed glioneuronal neoplasm classified as CNS WHO grade 1 and is most often reported in young adults. Pediatric cases, particularly in the parieto-occipital region with predominant visual symptoms, are uncommon. We reported on an 11-year-old girl who presented with headache, right periorbital pain and diplopia for 10 days. Examination revealed restriction of the right temporal visual field, uncrossed diplopia in upward gaze and right lateral rectus paresis. Preoperative MRI demonstrated a large left parieto-occipital solid-cystic intra-axial lesion with an enhancing solid component and associated mass effect. The patient underwent left parieto-occipital craniotomy and excision of the lesion in lateral position with head fixation in a Sugita clamp. A single burr-hole-based parieto-occipital craniotomy was performed. Intraoperatively, a solid-cystic tumor was noted; the solid component was greyish, firm to hard, not suckable and moderately vascular, while the cystic component contained yellowish fluid. Gross total excision was achieved. Cytology of the cyst fluid showed cerebral cyst fluid with no abnormal cells. Histopathology revealed a heterogeneous tumor with papillary structures having fibrocollagenous cores and a single to pseudostratified lining. A biphasic pattern of glial and neuronal components was seen with stromal hyalinization and calcification. Immunohistochemistry showed GFAP and S100 positivity in papillary lining cells and synaptophysin positivity in cells with neuronal morphology; p53 expression was absent. MIB-1 labeling index reached approximately 10% in areas with highest expression. The final diagnosis was papillary glioneuronal tumor (CNS WHO grade 1). Postoperatively, the patient was stable and diplopia resolved. A routine postoperative MRI on day 12 showed a heterogeneously enhancing residual-appearing lesion in the left high parietal/parasagittal parieto-occipital region. In view of residual disease, adjuvant radiotherapy was advised. This case adds to the limited pediatric literature on PGNT and highlights an unusual parieto-occipital presentation with diplopia and visual field deficit.

Keywords: Papillary Glioneuronal Tumor; Pediatric; Parieto-Occipital; Diplopia; Glioneuronal Tumor

Introduction

Papillary Glioneuronal Tumor (PGNT) is a rare mixed glioneuronal neoplasm categorized as CNS WHO grade 1. It is typically supratentorial and most frequently described in young adults, with headache and seizures as common presenting symptoms [1-3]. Pediatric cases are rare and occipital or parieto-occipital location with prominent visual symptoms is infrequently reported [4]. We present a pediatric left parieto-occipital PGNT with a clinical presentation dominated by diplopia and visual field restriction.

Cases and Methods

History

An 11-year-old girl, previously healthy, presented with headache, right periorbital pain and double vision for 10 days. The patient was apparently alright 10 days ago when she first developed headache. The headache was insidious in onset, dull aching in nature and gradually increased in intensity. Around the same time, she developed right periorbital pain that worsened on looking to the right or upwards. A few days later, she noticed uncrossed diplopia, more on upward gaze and rightward gaze, improving on closing either eye. There was no history of seizures, loss of consciousness, limb weakness, fever, recent trauma or bowel/bladder complaints. No significant past history was present.

Examination

The patient was conscious and oriented.

- Right temporal visual field restriction
- Uncrossed diplopia on upward gaze
- Right lateral rectus paresis on extraocular movement testing other cranial nerves and the rest of the neurological examination were unremarkable.

Imaging

Pre-op MRI: Large Left parieto-occipital solid-cystic intra-axial lesion with a prominent cystic component and an enhancing solid component, with associated mass effect (Fig. 1).

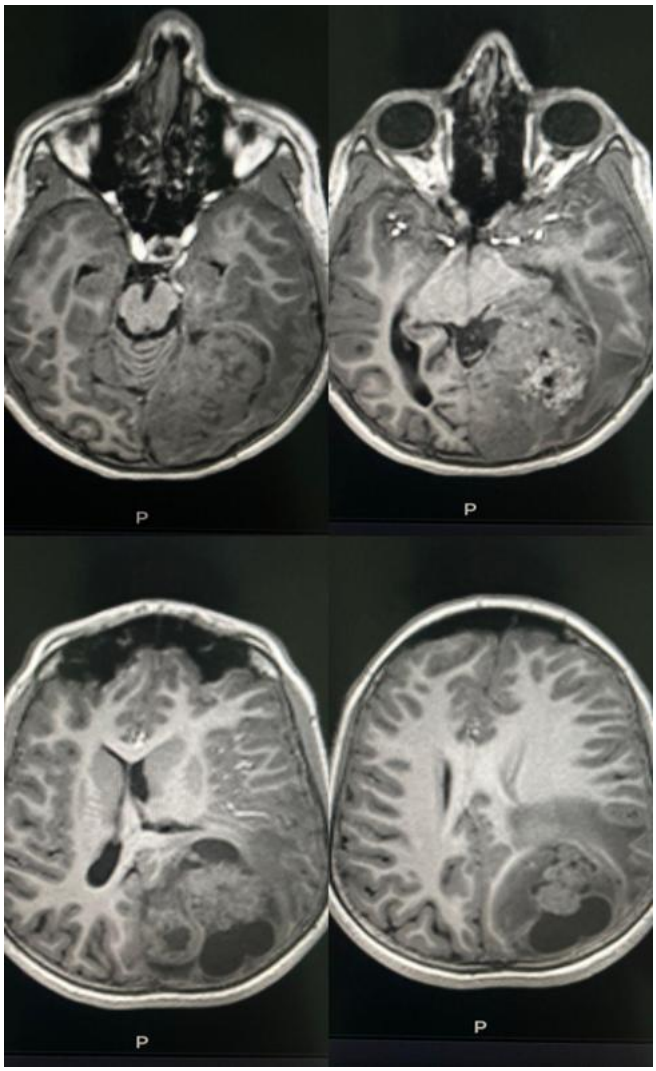


Figure 1: Pre-op MRI images.

Post-op MRI: MRI on postoperative day 12 showed a residual enhancing lesion in the left high parietal/parasagittal parieto-occipital region, T2/FLAIR hyperintense, T1 isointense, with heterogeneous post-contrast enhancement (Fig. 2).

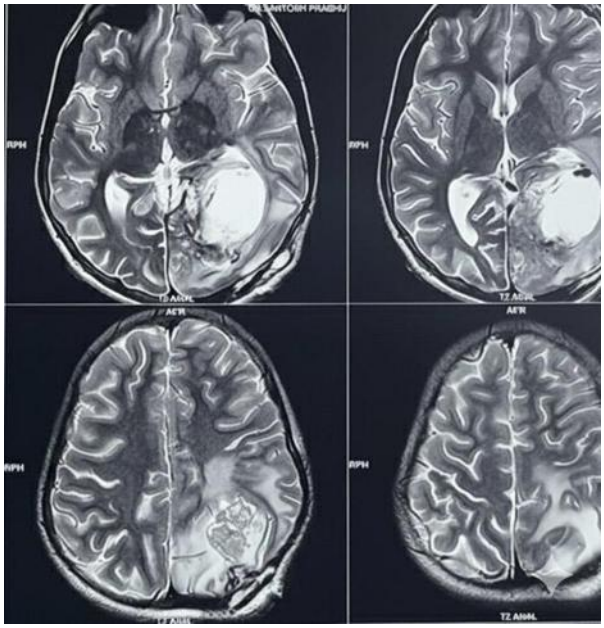


Figure 2: Post-op MRI images.

Surgery

The patient underwent excision of the left occipital space-occupying lesion. Surgery was performed in lateral position with head fixation in a Sugita clamp. A left parieto-occipital single burr-hole craniotomy was done.

A solid-cystic tumor was encountered. The solid component was greyish, firm to hard, not suckable and moderately vascular. The cystic component contained yellowish fluid. Gross total excision was achieved (Fig. 3).

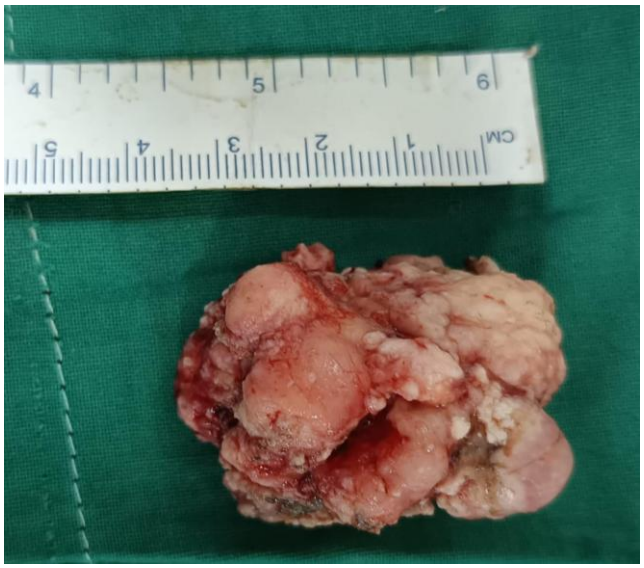


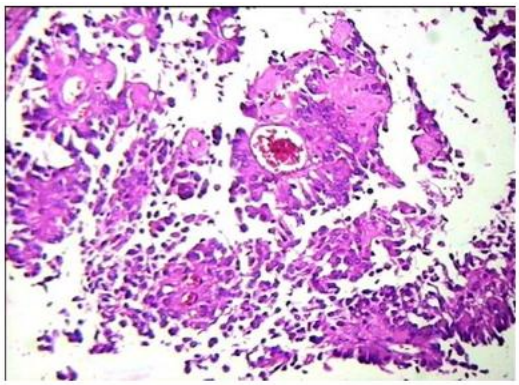
Figure 3: Surgery was performed in lateral position with head fixation in a sugita clamp.

Pathology

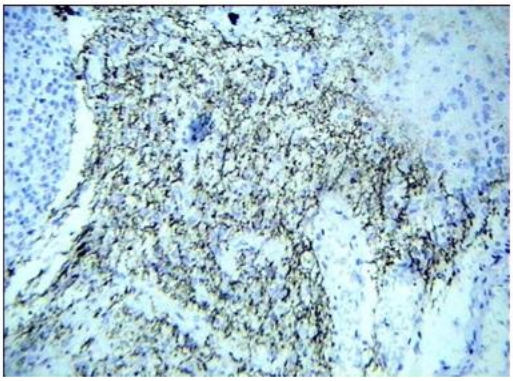
Cytology of the cyst fluid showed cerebral cyst fluid with no abnormal cells. Histopathology revealed a heterogeneous tumor showing papillary structures with fibrocollagenous cores and a single to pseudostratified lining of tumor cells. The glial component showed scant cytoplasm; in places this was pilocytic in loose fascicles. The neuronal component showed uniform

round vesicular nuclei with moderate eosinophilic cytoplasm; a few larger neurons were noted. The stroma was hyalinized with extensive calcification in areas. Stromal vessels showed prominent hyalinization.

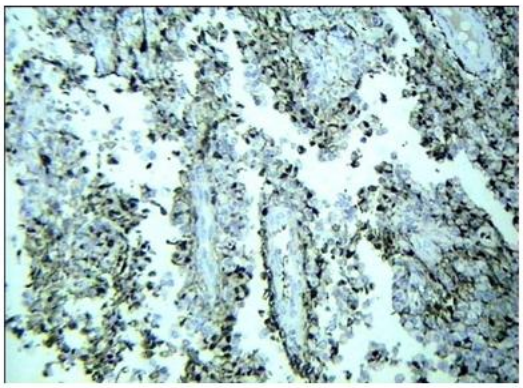
Immunohistochemistry showed GFAP and S100 positivity in papillary lining cells and synaptophysin positivity in cells with neuronal morphology. Tumor cells did not show p53 expression. MIB-1 labeling index was approximately 10% in hotspots.
Final diagnosis: Papillary glioneuronal tumor (CNS WHO grade 1) (Fig. 4).



HPE



Synaptophysin Positive



S100 POSITIVE RIMMING OF PAPILLAE A

Figure 4: Histopathology/IHC images.

Follow-up

Postoperatively, the patient was stable and diplopia resolved. In view of suspected residual lesion on postoperative imaging, adjuvant radiotherapy was advised.

Discussion

PGNT is a rare mixed glioneuronal neoplasm with generally indolent behavior and favorable outcomes following maximal safe resection [1,5,6]. The majority of published cases involve young adults and supratentorial locations, with headache and seizures as dominant symptoms [7]. Pediatric presentations remain uncommon [4]. Our case is unusual due to the pediatric age, parieto-occipital location and presentation with visual field deficit and abducens palsy-related diplopia. The solid-cystic morphology with an enhancing solid component is consistent with reported imaging patterns of PGNT and emphasizes the importance of histopathology and immunohistochemistry for definitive diagnosis [7]. Histology and immunophenotype demonstrated classic biphasic glioneuronal differentiation. The MIB-1 hotspot index reaching ~10% is relatively higher than expected for many grade 1 tumors, supporting close surveillance and individualized adjuvant decision-making. The early postoperative imaging impression of residual lesion further supported the recommendation for radiotherapy.

Conclusion

Papillary glioneuronal tumor of the parieto-occipital region in a child presenting with diplopia and visual field deficit is rare. Maximal safe resection remains the primary treatment. Suspected residual lesion may warrant consideration of adjuvant radiotherapy with close follow-up.

Conflict of Interest

The authors declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

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Data Availability Statement

The clinical data used in this case report consist of private patient medical records and are not publicly available due to patient confidentiality. De-identified data may be made available from the corresponding author upon reasonable request.

Ethical Statement

Written informed consent was obtained from the patient's parents/guardian for publication of this case report and accompanying images.

Informed Consent Statement

Written informed consent was obtained from the patient's parents/guardian for publication of this case report and accompanying images

Authors' Contributions

All authors contributed to the study conception and design. The first draft of the manuscript was written by Rajashree Hariprasad and all authors commented on previous versions of the manuscript. Revisions were done by Arpita Padhy, Wefaq Alshami, Neela Zalmay and Annika Daug. Validation: Dr. Brian Lee and Dr. Frances Chow. Supervision: Dr. Hari Prasad Kunhi Veedu. All authors read and approved the final manuscript.

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