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Clinical outcomes of adults with intracranial grade 1 and 2 ganglioglioma

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Highlights

- Low grade adult intracranial ganglioglioma has an excellent prognosis.
- We report a recurrence rate of<10% for low-grade adult intracranial ganglioglioma.
- GTR offers a>50% chance of seizure-freedom.
- The lack of recurrence with GTR means MRI surveillance is not needed beyond 5years.

- Longer-term follow-up is required after STR due to the low risk of recurrence.

Abstract

Ganglioglioma is a rare primary brain tumour that most frequently occurs in children and young adults. They are generally low-grade and have a favourable prognosis, but there is limited literature to guide the optimal management. The aim of this study was to investigate the clinical outcomes of adults with intracranial ganglioglioma, and to determine the frequency and duration of radiological follow-up. Thirteen adult patients with CNS WHO grade 1 and 2 ganglioglioma were identified retrospectively from case records at a tertiary neurosurgical centre between 2010 and 2022. Patient characteristics, magnetic resonance imaging (MRI) features, and clinical outcomes were obtained. Surgery was classified as gross total (GTR) or subtotal (STR) resection. 84.6% (n=11) of patients had temporal lobe tumours, with most (69.2%) presenting with seizures, at a median age of 29.0 years. GTR and STR were achieved in nine and five patients, respectively. No patients received adjuvant radiotherapy. During the median follow-up period of 8.9 years there was no radiological recurrence after GTR, and only one recurrence after STR at 65 months that did not require treatment. There was no patient mortality. Two patients continued to have seizures at last clinical follow-up. Low grade adult intracranial ganglioglioma has an excellent prognosis, with a recurrence rate below 10% in this series. Long-term surveillance is not necessarily required if GTR has been achieved and patients can be considered for discharge after annual MRI for 5 years. In patients where only STR is achieved, annual MRI is required although the progression/recurrence rate remains low and asymptomatic.

Introduction

Gangliogliomas represent 0.4% of all CNS tumours and 1%–7.6% of all primary brain tumours [1]. The CNS WHO classification system describes three grades. CNS WHO grade 1, 2 and 3 tumours make up 86%, 9% and 5% of all gangliogliomas, respectively, while <1% show features consistent with glioblastoma (WHO grade 4) [2]. It is most often the glial component of ganglioglioma which exhibits malignant progression, and rarely the neuronal component (into neuroblastoma) [2]. They occur in children and young adults [1], [3], and the median age at diagnosis is 32 years [4]. Gangliogliomas are slow-growing, typically present with seizures, and the commonest location is the temporal lobe [5]. Tumours are well-circumscribed and often partially cystic with an enhancing mural nodule [6].

Gross total resection (GTR) is the optimal treatment, and the role of chemotherapy or radiotherapy following sub-total resection (STR) remains unclear [4]. Gangliogliomas have an excellent prognosis, with a recent study reporting the 2-, 5-, and 10-year overall survival for patients with low-grade ganglioglioma to be 100%, 88%, and 84%, respectively [1]. The incidence of progression of low-grade tumours and transformation to a higher grade is 16–35% [1].

Favourable prognostic factors include GTR, younger age at diagnosis, female sex, temporal lobe location, and seizures [2], [4], [7], [8]. GTR confers a increased likelihood of seizure-freedom, compared to STR [8].

As adult gangliogliomas are so rare, there is limited literature to guide the management of such patients. Typically, these patients are followed up with serial MRI scans after surgical resection. Like with any cancer diagnosis, adult ganglioglioma patients may experience “scanxiety” – defined as the distress and/or anxiety occurring before, during, and after cancer-related imaging/scans [9]. Considering the low recurrence rates typically observed with these tumours, “scanxiety” could potentially be avoided by safely discharging patients earlier. Additionally, with every follow-up, there is an additional cost for both the patient (i.e. time off work, travel to hospital) and the healthcare service provider (i.e. MRI scan, hospital appointment, clinician time).

The aim of this study was to investigate the clinical outcomes of adults with adult intracranial ganglioglioma, and to determine the required radiological follow up frequency and duration.

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Methods

Adult patients with a pathologically confirmed CNS WHO grade 1 or 2 ganglioglioma between January 2010 and December 2022 were identified from pathology records for inclusion in the study. CNS WHO grade 3 gangliogliomas were excluded. Patients were treated at a tertiary neurosurgical centre. Patient demographics, clinical presentation, treatment, and follow-up were obtained from retrospective review of case notes and electronic patient records. The pathology diagnosis was based on the WHO …

Results

Thirteen patients met the inclusion criteria: nine patients had CNS WHO grade 1 ganglioglioma, and four had CNS WHO grade 2 ganglioglioma. BRAFV600E mutation was observed in three cases, was not present in seven cases and the test failed in three cases. The median age at diagnosis was 29.0years (range 17–44years). There were seven females and six males.

Nine patients presented with seizures – seven were new onset, and two had idiopathic long-standing epilepsy (Table 1). Four patients had …

Discussion

In this retrospective analysis of adult intracranial ganglioglioma treated at a single institution we have shown that seizures are the most common presenting symptom (69.2%), there is a predilection for the temporal lobe (84.6%), and the recurrence rate is low (<10%). Adult gangliogliomas have an excellent prognosis and based on our results, patients undergoing GTR do not require long-term monitoring and follow-up.

In this study of thirteen patients with ganglioglioma, GTR was achieved in …

Conclusion

This study provides a contribution to the existing literature on adult intracranial gangliogliomas. GTR is the recommended surgical aim where it is safe and feasible. When GTR is achieved, patients may be considered as cured and will potentially become seizure free. Long-term follow up is not warranted in cases where GTR is achieved – annual MRI and clinical follow-up for three years post-operatively is sufficient, and avoids patient "scanxiety", as well as unnecessary financial costs to both …

CRediT authorship contribution statement

Dana L. Hutton: Writing – review & editing, Writing – original draft, Data curation. **Janhavi Kulkarni:** Writing – review & editing, Writing – original draft, Data curation. **Khaja Syed:** Data curation. **Ian Scott:** Data curation. **Michael D. Cearns:** Writing – review & editing, Supervision, Resources, Methodology, Investigation, Data curation, Conceptualization. **Samantha J. Mills:** Resources, Data curation. **Michael D. Jenkinson:** Writing – review & editing, Supervision, Resources, Methodology, …

Data availability

The data from this study is not publically available. Please contact the corresponding author to enquire about access to the data set used for this article. …

Declaration of competing interest

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. …

Recommended articles

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