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Case Report

A sequential occurrence of neurocysticercosis and concomitant benign and malignant brain lesions: A case report of a 43-year-old Indian male

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ABSTRACT

Background: The occurrence of multiple brain tumors of different cellular origins in a single individual is extremely rare. There is limited documentation regarding the incidence of intracranial neoplasms in individuals with preexisting neurocysticercosis (NCC).

Case Description: We report the case of a 43-year-old male who had been under our care since he first suffered from seizures 2½ years ago when he was diagnosed with NCC. A year after the diagnosis of NCC, he presented to the emergency room with seizures, when he was found to have a new small left frontal meningioma, which was managed conservatively. In the next year, the patient was admitted to the emergency room in a disoriented state, and his imaging revealed a new lesion – a left frontal glioma, for which he was operated. Six months later, another glioma was found in the right frontal region, which was excised surgically. Four months after the second surgery, the patient was brought with intractable seizures when he was diagnosed with cerebrospinal fluid spread of NCC. During this admission, the patient expired due to a pulmonary infection.

Conclusion: This case report presents the sequential occurrence of neurocysticercosis, meningioma, and glioma in an Indian male patient. The occurrence of NCC with brain tumors is rarely reported in the literature; further research is needed to understand the occurrence of multiple brain tumors, especially in the setting of preexisting NCC.

Keywords: Glioma, Meningioma, Multiple, Neoplasms, Neurocysticercosis

INTRODUCTION

The occurrence of multiple primary brain tumors in a single individual is infrequent, especially when these tumors originate from distinct cellular types. Such cases are particularly rare and are often associated with prior radiation exposure or phakomatoses.[13,14] Neurocysticercosis (NCC) is recognized as the most common helminthic infection affecting the brain.[11] There are limited case reports that have noted the presence of brain tumors in patients with NCC.[4,6] The presence of both tumors in a single patient with a history of NCC adds a unique dimension to our understanding of tumor development in this context. This case contributes to the broader knowledge of tumor pathology associated with NCC and highlights the need for further investigation into such rare occurrences.

CASE REPORT

In September 2022, a 43-year-old male presented with recent onset personality change and forgetfulness over the past 20 days. On examination, the patient was not oriented to time, place, or person; his Glasgow Coma Scale (GCS) score was 14 (E4V4M6), and his mini-mental state examination score was 22. The patient also displayed features of acalculia as well as decreased power in the right upper and lower limbs (4/5). No sensory deficits were observed.

His medical history included an episode of NCC, treated 2½ years back with albendazole 15 mg/kg over 4 weeks 1 month; no concurrent medical conditions or relevant family history was reported at presentation. Finally, his medication consisted of antiepileptic medication only.

On his regular follow-up imaging the previous year, a new small hyperintense lesion on T2 was observed, suggestive of meningioma (1.5 cm × 2 cm) [Figure 1]. He was managed symptomatically, as the lesion was close to the motor strip, and the patient was neurologically intact.

On current admission, the ensuing brain magnetic resonance imaging (MRI) [Figure 2] revealed multiple findings as follows:

a. A T2 hyperintense lesion (4.2 × 3.5 cm) in the left frontal region with extensive edema; the lesion showed heterogeneous contrast enhancement on T1 fat-saturated sequences. The complementary magnetic resonance spectroscopy [Figure 3] showed reduced N-Acetylaspartate (NAA) with raised choline (choline/NAA ratio was 5–9 times). The latter findings suggested the presence of new high-grade glioma.
b. In keeping with the previous MRI the year before, an extra-axial T2 hyperintense 2.0 × 2.4 cm lesion in the left posterior frontal region was again demonstrated. The lesion also showed intense homogenous contrast enhancement on postgadolinium sequences, suggestive of meningioma.
c. Multiple T2 hypointense lesions were observed in both cerebral and cerebellar hemispheres bilaterally, all blooming on gradient echo sequence. No abnormal contrast enhancement was seen, suggesting the presence of calcified granulomas.

A surgical plan was made to excise both lesions. A left frontoparietal craniotomy in supine was performed, followed by a small corticectomy in the anterior part of the left middle frontal gyrus. The first tumor (Sample A = suspected to be a glioma) was ill-defined, extending up to the frontal horn of the left lateral ventricle, grayish in color, mildly vascular, and aspiratable.

The second tumor (Sample B = presumed to be meningioma), located in the left posterior frontal region, was approached next and was found attached to the dura, compressing the brain parenchyma. It was found to be grayish and moderately vascular, measuring approximately 2 × 2 cm in dimensions and located about 3 cm lateral to the midline. Due to its dense adherence to the motor strip, only a partial resection

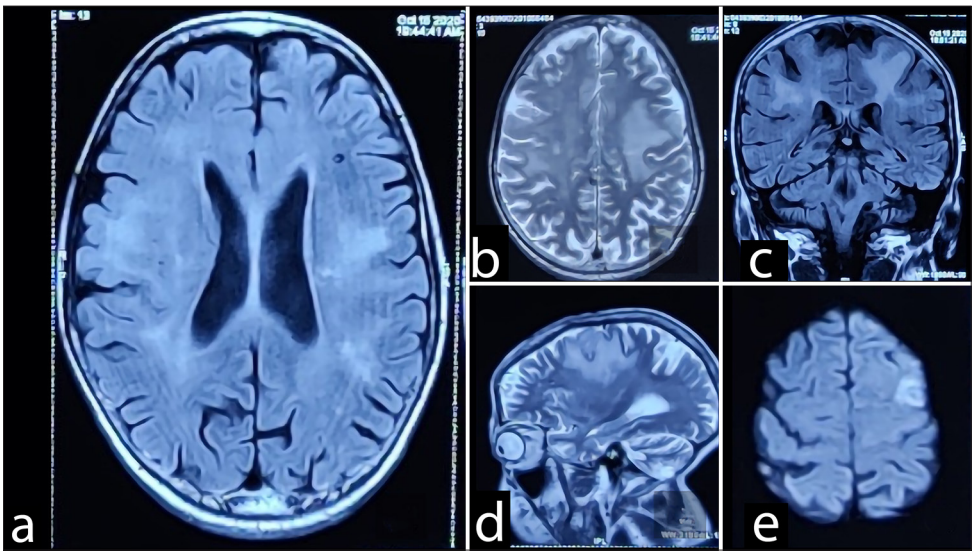

Figure 1: (a) Multiple foci of GRE blooming seen in left cerebellum, bilateral temporal, bilateral basal ganglia, frontoparietal and occipital region. (b) Lesions are iso to hypointense on T1W/T2W/Flair-attenuated inversion recovery (FLAIR) images. (c and d) Perifocal edema seen around some of these, largest in right frontoparietal parasagittal region measuring 7 mm in axial plane. (e) Small meningioma was found on left posterior frontal region.

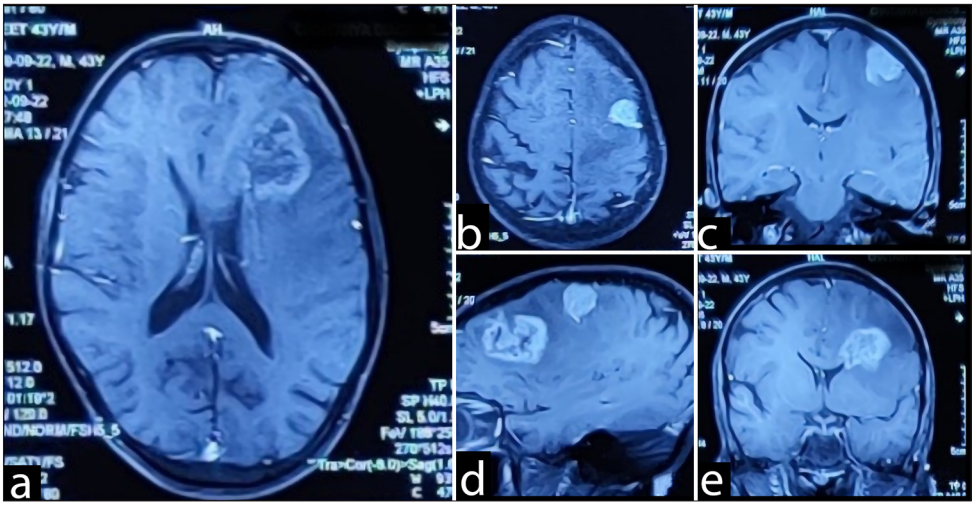

Figure 2: (a) T2 hyperintense lesion (4.2 x 3.5 cm) in the left frontal region with extensive edema with heterogeneous contrast enhancement suggestive of Glioma .(b-e) Another extra-axial T2 hyperintense lesion is in the left posterior frontal region measuring (2.0 x 2.4 cm) with contrast enhancement suggestive of meningioma.

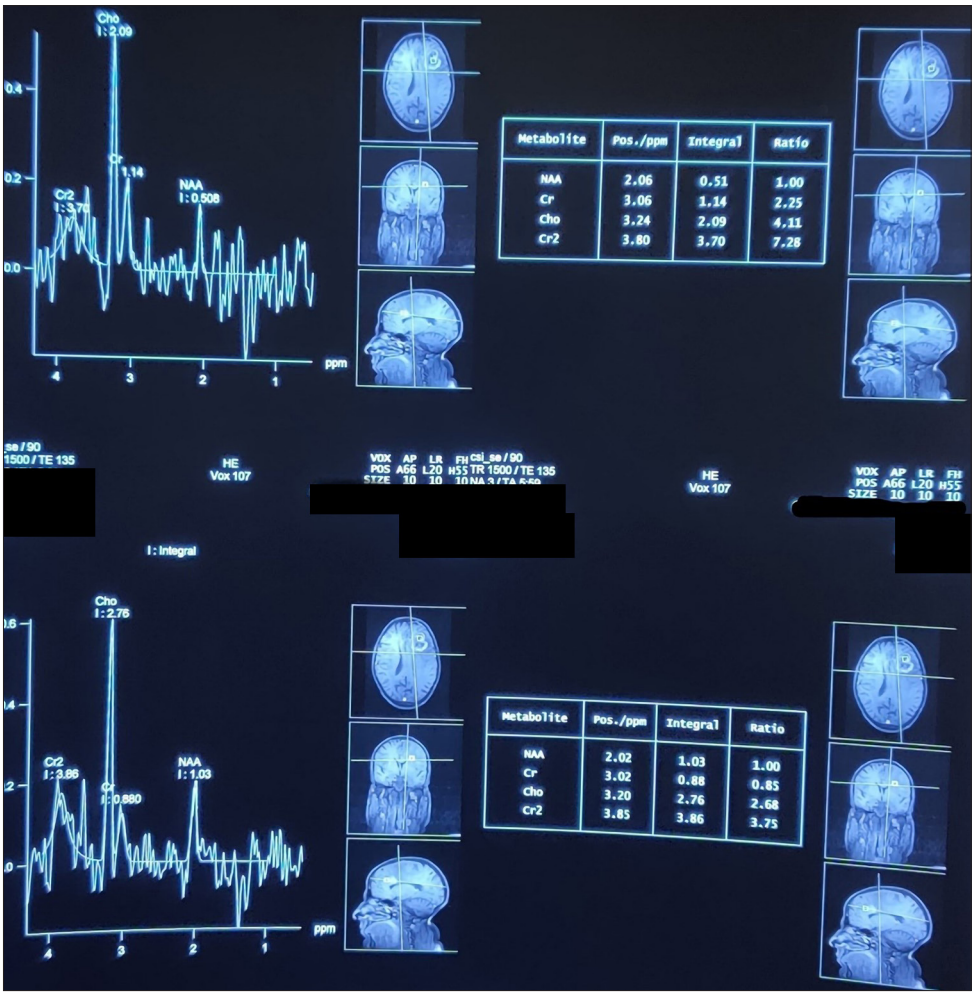

Figure 3: Magnetic resonance spectroscopy shows reduced N-Acetylaspartate (NAA) with raised choline in the left frontal lesion. Choline/NAA ratio is 5–9 times. Findings are suggestive of glioma.

was possible after surgery. The patient was extubated and shifted to the intensive care unit. The postoperative period was uneventful [Figure 4], discharged 5 days postoperatively.

Upon outpatient review a week later, he was doing well with no significant complaints. The subsequent histopathological examination of sample A [Figure 5] revealed atypical neuroglial cells with high nuclear-cytoplasmic ratio on a fibrillary background. Microvascular proliferation and mitotic activity were also reported, although without evidence of necrosis. Tumor cells were immunoreactive for Glial fibrillary acidic protein (GFAP) and immunonegative for P53 and Isocitrate dehydrogensae (IDH). The Ki-67 labeling index was 20%. The morphology and immunoprofile favored a diagnosis of high-grade diffuse astrocytoma (WHO grade 4).

Histopathological examination of Sample B [Figure 6] showed oval cells in small clusters with psammoma bodies and no mitosis or necrosis. Tumor cells were immunoreactive for epithelial membrane antigen (EMA) and progesterone receptors (PR), whereas the Ki-67 labeling index was set at 2%. The morphology and immune profile favored the presence of psammomatous meningioma (WHO Grade 1).

In view of the glioma, the patient underwent standard chemo-radiotherapy (60 Gy in 30 fractions with concurrent temozolomide), followed by temozolomide for the next 6 months.

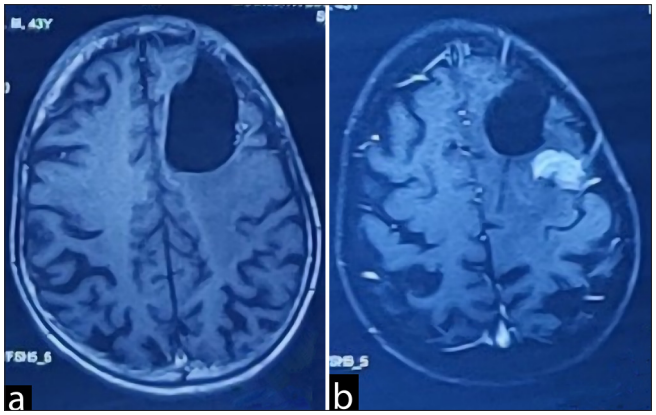

Figure 4: Postoperative MRI revealing (a) adequate excision of left frontal glioma and (b) partial excision of left posterior frontal meningioma.

Long-term follow-up

After an uneventful 6 months, the patient was readmitted to the emergency department with complaints of seizures. On the MRI brain [Figure 7], a T2 hyperintense lesion (3 × 2 cm) in the right frontal region was identified, suspected to be a new focus of high-grade glioma. As a result, a right frontal craniotomy was done; an ill-defined, mildly vascular, gray-colored, aspiratable tumor was excised; the postoperative recovery was uneventful [Figure 8].

Despite a good recovery in the following 4 months, the patient was readmitted to the emergency department with intractable seizures; on admission, the GCS score was 9 (E4V1M) with restricted left eye movements. The ensuing contrast MRI [Figure 9] brain revealed multiple lobulated heterogeneous enhancing lesions in both cerebral hemispheres and the region of the right cerebellum, not seen on previous imaging. The largest lesion was seen in the fourth ventricle; ultimately, cerebrospinal fluid (CSF) spread of NCC was suggested yet with no serology confirmation. The patient was managed medically with antiepileptics, steroids, sedatives, antibiotics, and intravenous fluids. Unfortunately, the patient developed a severe pulmonary infection and expired shortly after.

DISCUSSION

This case report highlights the diagnostic and management challenges posed by the complex interplay between concomitant brain lesions and infectious entities.

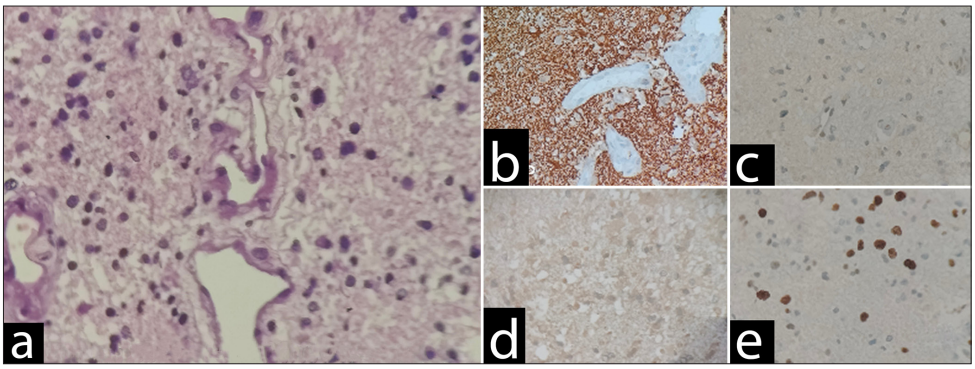

Figure 5: High-grade diffuse glioma with astrocytic morphology and immunoprofile suggestive of IDH wild-type glioblastoma (central nervous system WHO Grade 4) (a) Sections show a diffuse high-grade glioma with astrocytic morphology. There is microvascular proliferation and increased mitosis. However, necrosis is not identified. (b) GFAP: Diffuse and strongly positive. (c) p53: Patchy (wild type). (d) IDH: Negative. (e) Ki67: 20%. GFAP: Glial fibrillary acidic protein, IDH: Isocitrate dehydrogenase (10x magnification).

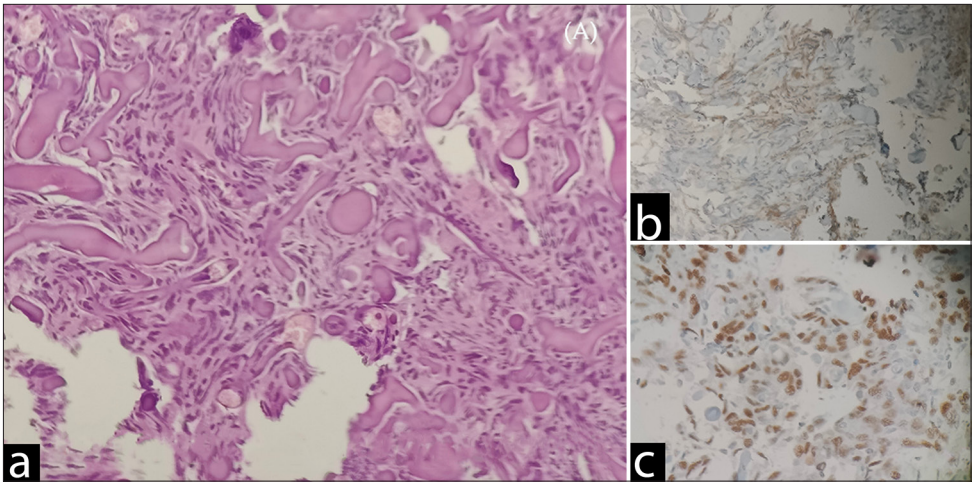

Figure 6: Psammomatous meningioma (central nervous system WHO grade 1) (a) Sections show whorls of meningothelial cells with bland nuclei with nuclear pseudoinclusions in few. Numerous psammoma bodies were noted. Negative for increased mitosis, necrosis, direct brain invasion, and features of atypical or anaplastic meningioma. (b) EMA: Diffuse positive. (c) PR: Weak positive. EMA: Epithethelial membrane antigen, PR: Progesterone receptor.

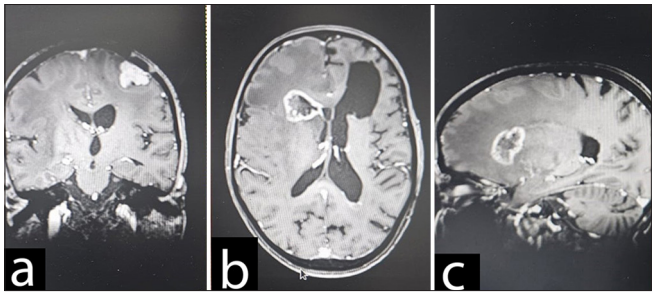

Figure 7: MRI Brain revealing (a) partially excised left posterior frontal meningioma and (b,c) right frontal glioma.

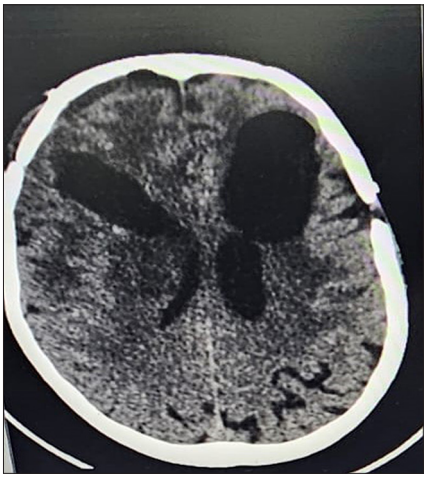

Figure 8: Postoperative noncontrast computed tomography head suggestive of adequate excision of right frontal glioma.

In general, the development of concurring primary brain tumors with different cellular origins remains a rare phenomenon.[13,14] In a paper published by Tunthanathip *et al*.,[17] the group described such occurrences most commonly taking place in the fourth to sixth decades of life, with females being more frequently affected. In their study analysis, meningiomas were frequently associated with pituitary adenoma, followed by gliomas. On the other hand, gliomas were found to be mostly associated with meningioma, often diagnosed simultaneously, as was the case here.

Without clear evidence, mainly two theories have been proposed to explain such unusual findings. The pathogenesis has been ascribed to perilesional tissue irritation, a result of edema associated with the tumor; the irritation is believed to induce arachnoid or astrocyte transformation, subsequently leading to tumorigenesis. In addition, a genetic pathway has been proposed, which includes abnormalities of p53, receptor tyrosine kinases, notch, Wnt (Wingless-related integration site), and other signal transduction mechanisms.[3,15] In addition, Suzuki *et al*. found strong positive expression of platelet-derived growth factor receptors to be a common feature of glioblastoma and meningioma.[16]

In the same fashion, Amatya *et al*. reported that the p53-positivity rate was <5% in all cases of benign meningioma, whereas it was >10% in 19% of cases of atypical meningiomas and 70% of cases of anaplastic meningiomas.[1]

Studies by Nestler *et al*. and Ohba *et al*.,[9,10] on the other hand, failed to come to a concrete conclusion implicating the genetic pathway. One hypothesis explaining the consecutive occurrence of these lesions is that they were coincidental; however, other groups, such as Spallone *et al*., have suggested that adjacent meningiomas and glial tumors might occur due

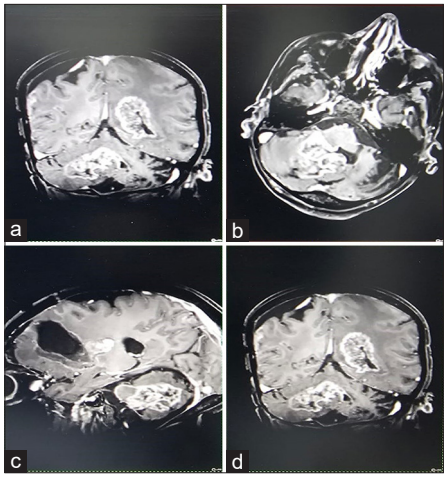

Figure 9: (a) Multiple lobulated heterogeneously enhancing lesions in bilateral cerebral hemispheres, bilateral occipitoparietal region, left high frontal subcortical region, right cerebellar hemisphereprotruding into 4th ventricle. (b and c) Largest lesion is noted in 4th ventricle measuring 4.7x3.9x3.1 cm. (d) Irregular gyral enhancement with associated gyral thickening noted in bilateral occipitoparietal regions, possibility of Cerebrospinal fluid (CSF) spread.

to an underlying tumor irritation, inducing the second tumor in the perilesional region.[15] In our case, MR findings initially suggested a meningioma and a new lesion indicative of high-grade glioma. The characterization of these lesions was supported by histopathological analysis, revealing high-grade diffuse astrocytoma (WHO Grade-4) and psammomatous meningioma (WHO Grade 1), respectively. This dual pathology is unusual but not unprecedented: our review of the literature indicates that while solitary meningiomas and gliomas are common, their co-occurrence remains a rare phenomenon. Beyond tumoral irritation as a precursor for malignancy, some studies have also suggested that multiple primary brain tumors can arise from other shared risk factors such as genetic predispositions or environmental exposures, though in this case, a direct connection between NCC and tumor development remains speculative.[1,3]

Association of brain tumors with NCC

The management approach in this case involved a combination of surgical resection, radiotherapy, and chemotherapy, which aligns with standard protocols for high-grade gliomas and meningiomas. The initial decision to manage the meningioma symptomatically due to its proximity to the motor strip was prudent, given that aggressive resection could have led to significant neurological deficits. However, the need for excising additional lesions along with late NCC-related dissemination highlights the confronting challenges the patient and the multidisciplinary team had to face throughout follow-up.

NCC is known to be the most common helminthic infection affecting humans, with a widely reported inflammatory response.[11] Suggested mechanisms of carcinogenesis in NCC include chronic inflammation, modulation of the host immune system, inhibition of intracellular communication, disruption of proliferation–antiproliferation pathways, induction of genomic instability, and stimulation of malignant stem cell progeny.[4,6-8,12] In the context of host-parasite interaction and ensuing immune responses, a study conducted on surgically resected and autopsied material proposed that the surface glycoprotein of the cysticercal cyst is the antigenic component able to trigger antibody production in CSF; these antigens can be found in large amounts in inflammatory settings, ultimately enhancing the parasite's capability of self-preservation.[5] Scarce evidence exists to document the involvement of other helminthic/parasitic agents in brain malignancy, such as a systematic review done in 2022, only highlighting the prevalence of *Toxoplasma gondii* with brain tumors.[2] Therefore, despite these case reports and studies, it is difficult to draw definitive cause-effect conclusions involving NCC and brain malignancies.

CONCLUSION

Considering the inherently dynamic nature of NCC, close surveillance structured on serial imaging and clinic controls remains of the essence. Indeed, timely imaging with MRI or computed tomography scans allows for effective tracking of disease progression, evaluation of treatment efficacy, and early detection of secondary issues, thereby facilitating optimal patient care. Although existing data are sparse, there is a plausible concern that chronic inflammation and other changes associated with NCC could predispose patients to an increased risk of secondary neoplasms. To address this concern, future studies should focus on large-scale epidemiological investigations to determine if NCC is linked to a higher incidence of brain tumors. Aiming to improve relevant treatment strategies, mechanistic research with large cohorts is warranted to uncover the biological pathways through which NCC might influence tumor development, including those involving prolonged inflammation and/or immune system alterations.

Ethical approval

Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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