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Short clinical case

Awake surgery for isolated parenchymal degenerating neurocysticercosis – Case report and focused review of misdiagnosis of neurocysticercosis

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ABSTRACT

Differential diagnosis of isolated single neurocysticercosis can be difficult, and management is controversial. We report here an original surgical strategy, and review previous studies reporting misdiagnosis, using the PRISMA guidelines. A 24-year-old man was admitted to our hospital for recent memory impairment, hypoesthesia of the right hand, and recurrent focal seizures without loss of consciousness. Brain MRI revealed a single ring-enhancing parenchymal lesion in the left superior postcentral gyrus, with large perilesional edema. Since exhaustive systemic exploration was negative, surgical resection of the lesion was decided on in a multidisciplinary team meeting. To preserve eloquent brain areas, surgery was performed in awake condition. It allowed complete resolution of clinical manifestations. The diagnosis of neurocysticercosis was confirmed on pathology. This case illustrates the utility of awake surgery in degenerating neurocysticercosis in functional areas, and emphasizes the importance of including it in differential diagnosis of cystic ring-enhancing brain lesions.

1. Introduction

Single ring-enhancing parenchymal lesions of the central nervous system have multiple differential diagnoses, including degenerating neurocysticercosis and tuberculoma. Differentiation between these two conditions can be a real dilemma: many cases of neurocysticercosis are seronegative, being already degenerated or because antibody response is not high enough, while many cases of tuberculoma are isolated, with no other mycobacteria sites [1–3]. Cystic brain metastases or primary tumor can also be incriminated, and misdiagnosis between neurocysticercosis and neoplasia has frequently been reported in the literature [4–43]. Managing such equivocal clinical conditions can be difficult, notably in case of large lesions located in eloquent brain areas. Surgery in awake condition can enable safe lesion resection and definitive diagnosis. Here we demonstrate, for the first time, the role of this technique in the management of isolated neurocysticercosis. We also performed a review of studies investigating misdiagnosis.

2. Case report

A 24-year-old soldier, native of Madagascar but living in France for 5 years, was admitted to the emergency department after recurrent aware focal seizures. The patient was right-handed, with no neurological history. He had had the first aware focal sensory seizure (paresthesia of the right upper limb, subsequently irradiating to the rest of the right hemi-body)two months earlier, with no triggering factor. On first examination, he had right hand hypoesthesia and was subfebrile (37.8 °C), but without signs of meningitis.

Brain MRI disclosed a solitary multilobular cystic ring-enhancing lesion, located at the gray/white matter junction of the superior left postcentral gyrus. The center of the lesion was hypointense on T1-weighted images, hyperintense on T2-weighted images, with increased apparent diffusion coefficient, suggesting necrosis and ruling out pyogenic abscess. There was large perilesional edema and heterogeneous enhancement of the lesion periphery on the contrast-enhanced T1-weighted images. MR

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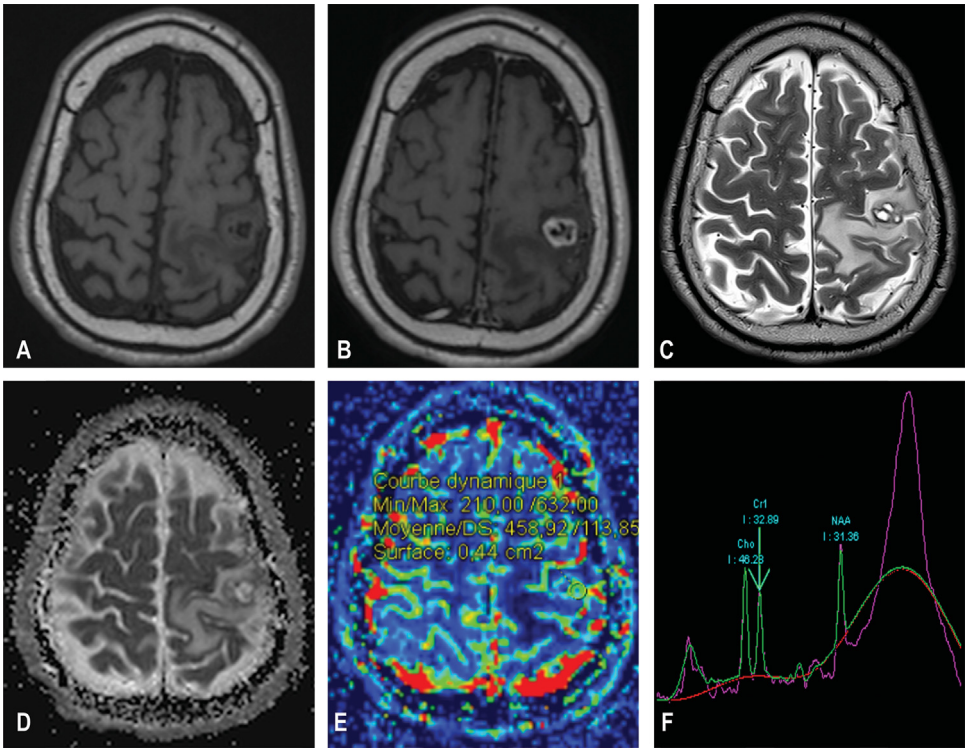

Fig. 1. Preoperative MRI findings. A. T1-weighted imaging showing single lesion in the superior left postcentral gyrus. B. T1-weighted gadolinium-enhanced imaging showing irregular rim enhancement of the lesion. C. T2-weighted imaging showing a multilobular cystic structure of the lesion, and large perilesional edema. D. Diffusion-weighted imaging showing increased apparent diffusion coefficient of the lesion. E. MR perfusion-weighted imaging showing slightly increased perfusion of the lesion. F. Long Echo Time MR spectroscopy showing increased choline peak with preserved choline/N-acetyl aspartate ratio.

spectroscopy found marked elevation of the lipid/lactate peak and depression of the creatine and N-acetyl aspartate peaks. MR perfusion-weighted imaging revealed slight neo-angiogenesis, which was difficult to interpret because of the proximity of the brain cortex. Altogether, the MRI aspect of the lesion was rather in favor of non-pyogenic infection, but a neoplastic lesion was not entirely ruled out (see Fig. 1).

Daily antiepileptic treatment was started and evidence of parasitic infection, systemic disease or secondary brain tumor was explored. Thoraco-abdominopelvic CT did not find any progressive

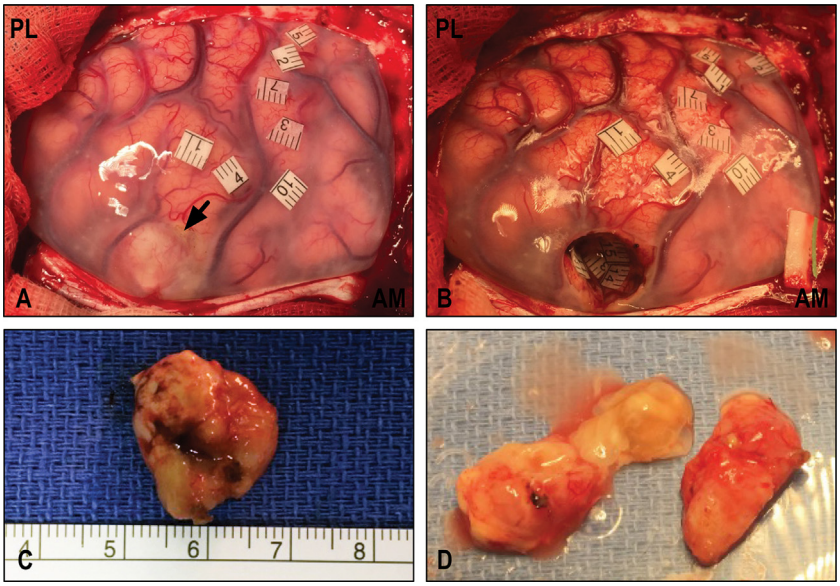

Fig. 2. Intraoperative photographs and gross examination of the lesion. A. Photograph of the brain surface, taken after cortical mapping and before lesion resection. The lesion is visible on the cortical surface as whitish area (arrow). Direct electrical stimulation led to dysarthria (tag 5), speech arrest/negative motor response of the right upper limb (tags 3 and 10), arrest of the fine motor skills of the right hand (tags 2 and 7), and paresthesia of the right hand (tags 1 and 4). B. Photograph, taken after lesion resection, showing the previously identified cortical functional areas and the responsive sites found at the bottom of the surgical cavity. Paresthesia and arrest of the regular movement of the right arm were systematically induced by the stimulation of each of the subcortical eloquent sites indicated by sterile number tags. C. Macroscopic aspect of the removed parasitic abscess. D. Gross examination of the abscess after cutting in half. A thick wall surrounds cystic cavity, filled with gelatinous content. AM: anteromedial; PL: posterolateral.

or infectious lesions. The results of all other investigations–lumbar puncture, ophthalmologic examination and biological tests (blood culture and parasitological stool examination, serology for human immunodeficiency virus, hepatitis B or C virus, syphilis, taenia infection, and interferon-gamma-release assay) came back negative. Exhaustive neurocognitive assessment revealed mild deficits of visual spatial processing speed, working memory, executive functions and attention.

Given the clinical manifestations, the atypical radiological features and impossibility of ruling out tumor, and the volume of the lesion, surgical resection was decided on. Because of the location of the lesion in the primary somatosensory area, surgery was performed in awake condition in order to detect, and thus conserve, functional cortical and subcortical structures. Once the craniotomy was completed and the dura mater opened, the lesion was partially visualized on the brain surface (see Fig. 2A). It was delineated on intraoperative ultrasound and landmarked with sterile empty tags. Direct electrical stimulation was then performed, using a bipolar electrode probe with 5-mm inter-electrode gap (NIMBUS Stimulator, Newmedic, France), delivering a biphasic electric current (pulse frequency 60 Hz, 1 ms pulse width). Functional disturbances were evaluated online by a speech therapist and responsive sites (i.e., areas in which repetitive stimulation provoked at least three identical functional disturbances) were marked with sterile number tags. At current amplitude of 3.75 mA, the primary somatosensory and motor cortices were first identified by recording any sensory disturbance and by asking the patient to continually count up to ten while performing regular movements with the right arm. Then picture-naming (DO80) [44] and right arm movements were used as intraoperative tasks until the end of resection. Once the exposed brain surface was entirely mapped, lesion removal was started. A firm, whitish lesion, well delineated but strongly adherent to the arachnoid mater, was found and completely removed. Since perilesional tumor infiltration could not be ruled out, resection was extended into the modified adjacent brain tissue. Continuous subcortical stimulation was performed, using the same current amplitude of 3.75 mA, and surgery was suspended after identification of eloquent areas in the depth of the operative cavity (see Fig. 2A and B). Intraoperative photographs were systematically taken before and after resection.

The removed lesion was cut in half, and a cystic cavity with gelatinous content, surrounded by a thick wall, was found (see Fig. 2C and D). The histopathological examination revealed a degenerating parasitic cyst, containing a cysticercus with wavy cuticle external layer covered by microtriches and associated with aggregated subcuticular cells, and internal layer composed of loose myxoid tissue. The cyst was surrounded by a large inflammatory cell infiltrate (plasma cells, lymphocytes, macrophages, and rarer eosinophils, neutrophils and giant cells) in a background of fibrosis and gliosis (see Fig. 3).

On postoperative radiographs of the limbs, no calcified lesions were found in the soft tissue. Treatment with albendazole (400 mg twice daily), simultaneous to corticosteroid therapy, was performed for two 2 weeks. The neurological deficit recovered progressively, and neurocognitive assessment at 1 year was normal. At the 1-year follow-up, brain MRI found total edema resorption without any new or residual infectious lesion. There had been no further epileptic seizures were recorded, and from the 13th postoperative month antiepileptic treatment was gradually stopped, allowing the patient to return to normal social and occupational life.

3. Discussion

Neurocysticercosis is the most common parasitosis of the central nervous system worldwide [45]. It is caused by the larval stage of the tapeworm *Taenia solium* after fecal-oral contamination with

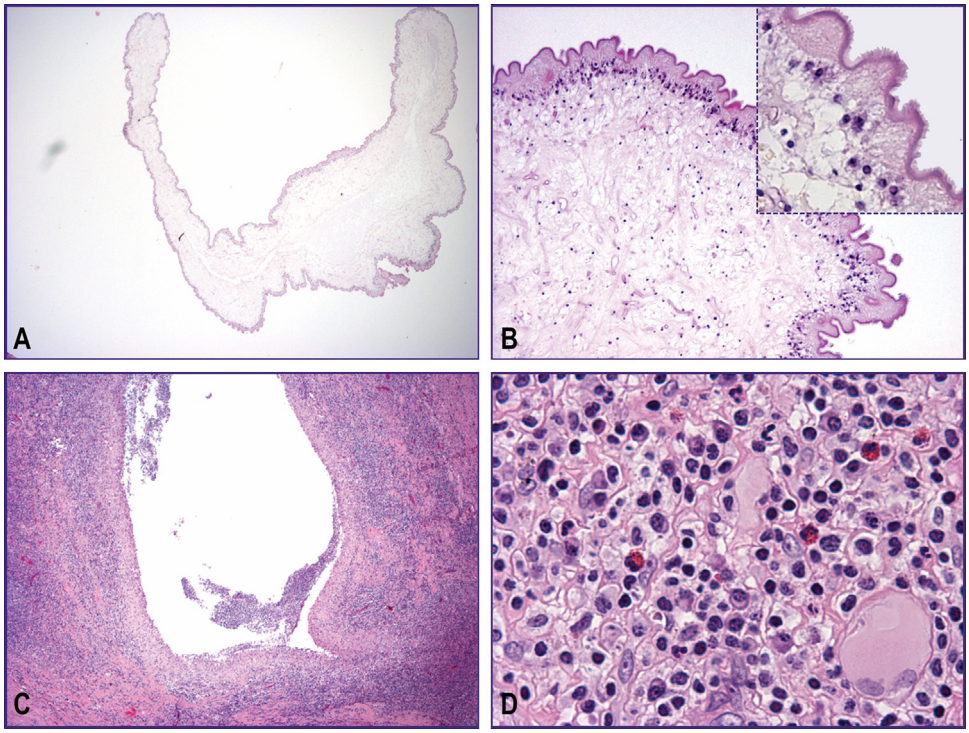

Fig. 3. Histopathological findings (hematoxylin and eosin stain). A. Cysticercus parasite composed of wavy cuticle external layer and loose myxoid internal content (original magnification ×20). B. Intermediate- (×100) and high-power (×400, upper right corner) magnifications of the cyscticercus, showing a superficial layer covered with microtriches, an intermediate cellular layer, and an inner loose reticular layer. C. Abscess wall and central cavity, seen at low (×20) magnification. D. High-power (×400) magnification of the abscess wall, showing large inflammatory cell infiltrate containing lymphocytes, plasma cells, eosinophils, neutrophils and giant cells.

Table 1
Literature review of studies reporting neurocysticercosis mimicking neoplasia.

Authors & Date	Age (yrs)/gender	Patient residence/Country of publication	Provisional diagnosis	Neoplastic history	Symptoms	Number of lesions	Location	Size	Imaging characteristics	Cysticercosis serology	Diagnosis
Arismendi-Morillo 2004	NA (NR)	NA (NR)/Venezuela	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
Bouillot 2003	14/NA (NR)	Endemic areas/France	High grade glioma	NA (NR)	Seizure	1	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
Bouillot 2003	29/NA (NR)	Endemic areas/France	High grade glioma	NA (NR)	Seizure, left facial paralysis	1	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
Bousquet 1996	12/M	Reunion Island/France	Retrobulbar optic nerve tumor	No	Left-eye vision loss without exophthalmos	1	Retrobulbar portion of the left optic nerve	NS	CT: hypodense center, hyperdense periphery, ring enhancement, no perilesional edema	NS	Surgical removal
Bruschi 2006	NA (NR)	NA (NR)/Italy	Tumor	NA (NR)	Seizures	1	Right-fronto-temporal	NS	MRI: cystic lesion	NA (NR)	Surgical removal
Colli 1984	22/F	NA (NR)/Brazil	Tumor	NA (NR)	Seizures, right hemiparesis, dysphasia	2	Left fronto-temporo-parietal, and other, more diffuse	70 × 77 mm	CT: cystic lesion	NA (NR)	Surgical removal
Colli 1986	15/F	Brazil/Brazil	Tumor	NS	Headache, dysosmia, hydrocephalic hypertension, cerebellar syndrome	Multiple	Right temporal, medial frontal	NS	CT: cystic lesion	(+) in serum (retrospectively)	Ventriculography
Colli 1986	13/M	Brazil/Brazil	Tumor	NS	Left brachio-facial paresis	Multiple	Left fronto-parietal and right parietal	NS	CT: enhancing lesions, perilesional edema	(+) in serum	Serology, spontaneous disappearance
Coulibaly 2008	70/F	Guadeloupe/France	Metastasis	NS	Gait disturbance, right hemiparesis, bradypsychia	Multiple	Diffuse	Small	CT/MRI: enhancing lesions, no perilesional edema	Western blot (+) in serum and CSF	Biopsy
Dao, 2015	37/F	Congo/Morocco	Metastasis	Breast adenocarcinoma	Headache, gait disorder	1	Left internal temporo-occipital.	57 × 51 mm (CT), 32 × 30 mm (MRI)	CT: hyperdense, ring enhancement, perilesional edema	(−) in serum	Biopsy
Kaw 1994	NA (NR)	NA (NR)/NA (NR)	Meningioma	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	Biopsy
Kim 2006	67/M	NS/Korea	Hemangioblastoma	NS	Dizziness, nausea, vomiting	1	Left cerebellar	55 × 40 × 40 mm	MRI: cystic lesion with thin wall, internal septations and mural nodule, ring enhancement except of the associated nodule, subtle perilesional edema	(+) in serum (retrospectively)	Surgical removal

Table 1 (*Continued*)

Authors & Date	Age (yrs)/gender	Patient residence/Country of publication	Provisional diagnosis	Neoplastic history	Symptoms	Number of lesions	Location	Size	Imaging characteristics	Cysticercosis serology	Diagnosis
L'Ollivier 2012	69/M	France/France	Glioma	NS	Asthenia and headache	1	Right frontal	40 × 30 mm	CT/MRI: internal necrotic or cystic septations, ring enhancement, perilesional edema	NS	Surgical removal
Matyja 1998	NA (NR)	NA (NR)/Poland	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
Matyja 1998	NA (NR)	NA (NR)/Poland	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
McDowell and Harper 1990	NA (NR)	NA (NR)/Australia	NA (NR)	NA (NR)	Seizures, headache	Multiple	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
McDowell and Harper 1990	NA (NR)	NA (NR)/Australia	NA (NR)	NA (NR)	Seizures, headache	1	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
Michael 1990	39/F	USA/USA	Tumor	No	Headache	1	Peripheral	NS	CT/MRI: isodense, heterogeneous medium intensity nodule, ring enhancement, large perilesional edema	NS	Surgical removal
Michael 1990	32/F	USA/USA	Tumor	No	Seizure	1	Right frontal	Small	CT/MRI: isodense, medium intensity, perilesional edema	NS	Surgical removal
Mittal 2014	1 ½/M	NS/India	Tumor	no	NA (NR)	1	Right thalamus	43 × 45 mm	CT/MRI: hypodense lesion with eccentric mural scolex, T1 hypointense, minimal enhancement, T2 heterogeneously hyperintense, increased choline peak and decreased N-acetyl aspartate, moderate perilesional edema	NS	New MRI 4 months later
Nandhagopal 2010	6/M	NA (NR)/NA (NR)	NA (NR)	Tuberous sclerosis	NA (NR)	2	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
Ohsaki 1999	19/F	NS (but lived in India for 5 years)/Japan	Tumor	No	Seizures	1	Right parietal	6 mm	MRI: T1 hypointense, ring enhancement, no perilesional edema	ELISA and EITB (-) (after surgical removal)	Surgical removal

Table 1 (*Continued*)

Authors & Date	Age (yrs)/gender	Patient residence/Country of publication	Provisional diagnosis	Neoplastic history	Symptoms	Number of lesions	Location	Size	Imaging characteristics	Cysticercosis serology	Diagnosis
Sabel 2001	47/M	Greece/Germany	Malignant tumor	NS	Speech difficulties, dyscalculia, alexia	1, multilobular	Left frontal	NS	CT/MRI: hypodense center and hyperdense periphery, other small calcified lesions, T1 hypointense, ring enhancement, T2 hyperintense, mild perilesional edema	NS	Surgical removal
Silver 1996	NA (NR)	Born in Korea/US	High grade glioma	No	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	Surgical removal
Song 2013	68/F	NS/China	glioma	NS	Facial numbness, left hearing impairment, headache	1	Left parietal	$42 \times 20 \times 25$ mm	MRI: T1 hypointense, ring enhancement, T2 hyperintense, no perilesional edema	(-) in serum	Surgical removal
Umredkar 2009	17/F	NS/India	Abscess or glioma	NS	HEADACHE, cognitive deficit	1	Left insular	$53 \times 43 \times 35$ mm	MRI: T1 hypointense, ring enhancement, T2 hyperintense, perilesional edema	NS	Surgical removal
Umredkar 2009	40/M	NS/India	Abscess or glioma	NS	Headache, seizures, cognitive deficit	1	Left frontal	NS	MRI: cystic lesion, ring enhancement, perilesional edema	NS	Surgical removal

Table 1 (*Continued*)

Authors & Date	Age (yrs)/gender	Patient residence/Country of publication	Provisional diagnosis	Neoplastic history	Symptoms	Number of lesions	Location	Size	Imaging characteristics	Cysticercosis serology	Diagnosis
Vasiljevic-Vuckovic 2011	30/F	NS/Serbia	Low grade tumor	NS	Seizure	1	Left occipital	35 × 20 mm	MRI: low-grade tumor spectrum, ring enhancement, no perilesional edema	NS	Surgical removal
Wada 2004	74/M	Philippines/Honolulu	Primary or secondary tumor	Gastric carcinoma	Seizures	1	Right frontal	20 × 25 × 30 mm	Irregular lesion, ring enhancement, perilesional edema	NS	Surgical removal
Yong and Warren 1994	NA (NR)	South America/Australia	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
Yong and Warren 1994	NA (NR)	South America/Australia	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
Yong and Warren 1994	NA (NR)	South America/Australia	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
Yong and Warren 1994	NA (NR)	Cyprus/Australia	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)

NA: not available; NR: full article not retrieved; NS: not specified in article; F: female; M: male; CT: computed tomography; MRI: magnetic resonance imaging; CSF: cerebrospinal fluid; EITB: enzyme-linked immunoelectrotransfer blot; ELISA: enzyme-linked immunosorbent assay.

Table 2
Literature review of studies reporting neoplasia mimicking neurocysticercosis.

Authors & Date	Age (yrs)/genderex	Patient residence/Country of publication	Final diagnosis	Neoplastic history	Symptoms	Number of lesions	Location	Size	Imaging characteristics	Cysticercosis serology	Diagnosis
Agapejev 1992	32/M	Brazil/Brazil	Oligodendroglioma	No	Headache, cognitive deficit	1	Supra-sellar and fronto-parietal	NS	CT: hypodense center, hyperdense periphery, ring enhancement, perilesional edema	(+) in CSF and punctured fluid	Surgical removal after therapeutic test (albendazole)
Bago-Rozankovic 2013	55/F	NS/Croatia	Papillary thyroid carcinoma metastases	No	Headache, dizziness	Multiple	Supra- and infra-tentorial	NS	CT/MRI: hypodense center, hyperdense periphery, cystic lesion, solid intraluminal component, ring enhancement, perilesional edema	(+) in serum	Biopsy of brain lesions after therapeutic test (albendazole)
Bick 2012	62/F	NS/US	Breast carcinoma metastases	Breast carcinoma	Seizures	Multiple	Diffuse	NS	CT/MRI: punctate peripheral calcifications, minimal enhancement	NS	NS
Choi 2012	50/M	Korea/Korea	Adenocarcinoma metastases (primitive not found)	No	Headache	Multiple	Prepontine and premedullary cisterns, cerebellar	NS	CT/MRI: cystic lesion, partial ring enhancement, no perilesional edema	(−) in serum and CSF	Partial surgical removal of brain lesions after therapeutic test (praziquantel)
Costa 2014	61/F	Brazil/Brazil	Lung adenocarcinoma metastases	No	Seizures	Multiple	Diffuse	NS	MRI: T1 hypo-intense, ring enhancement, perilesional edema	NS	Biopsy of brain lesions after therapeutic test (albendazole)
Fantini 2017	41/F	NA (NR) (caucasian)/Italy	Uterine cervical cancer metastases	Uterine cervical carcinoma	NA (NR)	Multiple	NA (NR)	NA (NR)	Calcifications	NA (NR)	NA (NR)
Graber 2011	56/M	South America/USA	Pleomorphic sarcoma of malignant fibrous histiocytoma type	No	Right sided weakness	Multiple	Diffuse	NS	CT/MRI: hypodense center, hyperdense periphery, cystic lesion, ring enhancement, perilesional edema for some lesions	(−) in serum	Biopsy of new lesion in muscle
Hoang 2017	64/M	NA (NR)/USA	Lung adenocarcinoma metastases	NA (NR)	Phototopia	Multiple	NA (NR)	NS	NA (NR)	NA (NR)	NA (NR)

Table 2 (*Continued*)

Authors & Date	Age (yrs)/genderex	Patient residence/Country of publication	Final diagnosis	Neoplastic history	Symptoms	Number of lesions	Location	Size	Imaging characteristics	Cysticercosis serology	Diagnosis
Kim 2017	47/M	NS/Korea	Non-small cell lung cancer metastases	No	Dysarthria	Multiple	Diffuse bilateral	?	MRI: smooth/thin-walled cystic lesions, little or no ring enhancement, no perilesional edema	(−) in serum and CSF	Therapeutic test (albendazole)
Lam 2016	60/F	NA (NR) (caucasian)/Canada	Small-cell neuroendocrine carcinoma metastases	No	Ataxia, otalgia	Multiple	Supra- and infra-tentorial	Varying sizes	CT/MRI: cystic lesions, mostly thin-walled, some calcified, fluid levels, ring enhancement, no perilesional edema	(−) serum	Biopsy of brain lesions
Li 2016	68/M	China/China	Anaplastic astrocytoma	No	Right lower limb weakness, headaches	1	Left frontal	6 mm	MRI: cystic lesion, smooth wall, ring enhancement, T1 hypointense, T2 hyperintense	(+) in serum	Surgical removal after therapeutic test (albendazole)
Mota 2011	47/M	NS/Brazil	Lung adenocarcinoma metastases	Lung cancer found concomitantly	Bilateral hearing loss, tinnitus, dizziness, imbalance	Multiple	Supra- and infra-tentorial	NS	CT/MRI: hypodense center, hyperdense periphery, T2 hyperintense, T1 hypointense, no enhancement, high apparent diffusion coefficient, no perilesional edema	NS	Surgical removal of a brain lesion after therapeutic test (albendazole)
Ruppert 2010	55/M	NS/USA	Lung adenocarcinoma metastases	Lung adenocarcinoma	Cerebellar ataxia	Milliary	Supra- and infra-tentorial	NS	MRI: T1 barely detectable	(−) in serum and CSF	Brain autopsy after therapeutic test (albendazole)
Salomao 2006	15/M	NS/Brazil	Anaplastic oligoastrocytoma	No	Seizures	Multiple	Left parieto-temporal, right hypothalamus	NS	CT: hypodense center, hyperdense periphery with mural nodule, perilesional edema, calcified mass in the right hypothalamic	(+) in the punctured fluid	Surgical removal after therapeutic test (albendazole)

Table 2 (*Continued*)

Authors & Date	Age (yrs)/genderex	Patient residence/Country of publication	Final diagnosis	Neoplastic history	Symptoms	Number of lesions	Location	Size	Imaging characteristics	Cysticercosis serology	Diagnosis
Troiani 2011	45/F	NS/Brazil	Breast cancer metastases	Breast cancer	Headache	Multiple	Diffuse	NS	CT: hypodense, cystic lesions some with small mural nodules, T1 hypointense, ring enhancement, more intensely in the region of mural nodules, T2 hyperintense	NS	Surgical removal after therapeutic test (albendazole)
Tuchman 2009	16/M	NS/USA	Choroid plexus papilloma	No	Depression, memory deficit	1	Septum pellucidum	NS	MRI: T1 hypo-intense, ring enhancement with associated enhancing nodule, T2 hyperintense, no perilesional edema	NS	Surgical removal

NA: not available; NR: full article not retrieved; NS: not specified in article; F: female; M: male; CT: computed tomography; MRI: magnetic resonance imaging; CSF: cerebrospinal fluid.

Table 3
Literature review of studies reporting neurocysticercosis mimicking tuberculoma.

Authors & Date	Age (yrs)/gender	Patient residence/Country of publication	Provisional diagnosis	Neoplastic history	Symptoms	Number of lesions	Location	Size	Imaging characteristics	Cysticercosis serology	Diagnosis
Dayananda 2011	20/M	India/India	Tuberculoma	NS	Headache, seizures	Conglomerate	Right frontal	29 × 17 mm	MRI: hypointense periphery, hyperintense center, no diffusion restriction, ring enhancement, perilesional edema	NS	Surgical removal
Maeda 2011	NS/F	India/Japan	Tuberculoma	No	Headache, right forearm paresthesia	Multiple	NS	NS	CT/MRI: scattered calcifications, T1 hyper-intense, enhancement, peripheral edema	Positive (serum)	Surgical removal (mitochondrial DNA PCR)

NS: not specified in article; M: male; F: female; MRI: magnetic resonance imaging.

Table 4
Literature review of studies reporting tuberculoma mimicking neurocysticercosis.

Authors & Date	Age (yrs)/gender	Patient residence/Country of publication	Final diagnosis	Neoplastic history	Symptoms	Number of lesions	Location	Size	Imaging characteristics	Cysticercosis serology	Diagnosis
Lu 2011	41/M	NS/Japan	Tuberculoma	NS	Vertigo, vomiting	Multiple	Supra- and infra-tentorial, ventricles	Small	MRI: T2-hyperintense, ring enhancement, perilesional edema	(+) in serum, (-) in CSF	Failure of therapeutic test, MR spectroscopy
Seth 2010	4/F	India/India	Tuberculoma	NS	Seizures	3	Left frontal, bi-parietal	9.6–13 mm	CT/MRI: enhancing excentric foci, perilesional edema	(+) in serum	Failure of therapeutic test, MR spectroscopy
Tosomeen 1998	26/F	India/Lebanon	Tuberculoma	NS	Seizures	1	NA (NR)	NA (NR)	NA (NR)	NA (NR)	NA (NR)
Verma 2014	18/F	India/India	Tuberculoma	NS	Headache, seizure	Multiple	Supra- and infra-tentorial	Small	MRI: increased choline peak, increased choline to creatinine ratio, normal N-acetylaspartate peak, ring enhancement	(−) in serum	PCR tuberculosis

NA: not available; NR: full article not retrieved; NS: not specified in article; F: female; M: male; CT: computed tomography; MRI: magnetic resonance imaging; CSF: cerebrospinal fluid; PCR: polymerase chain reaction.

the eggs of the parasite. The embryos of the eggs reach the brain after hematogenous spread from the bowel, actively crossing the wall. After a long asymptomatic period, the initially viable cysticerci degenerate and the resulting inflammation causes specific symptoms, such as seizures and focal neurological and/or cognitive deficit, which depend on the topography and the size of the lesion(s).

The difficulty of differentiating neurocysticercosis from tumor or tuberculoma has often been discussed in the literature and will most likely continue to be in the future. Because of the completely different management, such cases can be a real moral dilemma for the clinician. On the one hand, anti-parasitic drugs used in neurocysticercosis have potential side-effects, can be ineffective in bulky lesions, and can delay urgent treatment if the true diagnosis is neoplasia. On the other hand, brain surgery is invasive and associated with specific risks, in particular that of inducing or irreversibly worsening a neurological/cognitive deficit. Such a risk is greater for lesions located in eloquent brain areas and removed in a classical manner (under general anesthesia).

Several studies have reported misdiagnosis of brain neurocysticercosis, and we performed a literature review of these, focusing on the differential diagnoses considered in the present case: tumor (studies listed in Tables 1 and 2) [4–43], and tuberculoma (studies listed in Tables 3 and 4) [46–51]. The PRISMA guidelines used to select articles are presented in Fig. 4.

Twenty-four studies reported 33 cases with neurocysticercotic brain lesions that mimicked neoplasia (see Table 1), and full-text papers were retrieved for 16. For patients with available data, mean age was 34.4 ± 22.7 years (range, 1.5–74 years). Definitive diagnosis of neurocysticercosis was made by biopsy in 3 patients, surgical removal of the lesion in 15 patients (inconclusive biopsy having been performed in 1), suggestive ventriculography (in 1983) in 1, and repeat imaging in a child whose parents refused an invasive procedure.

Tumor lesions initially diagnosed as neurocysticercosis were reported in 16 articles (see Table 2), and 14 full-text papers were retrieved. Sixteen patients were reported; for those with available data, mean age was 48.4 ± 15.3 years (range, 15–68 years). The final diagnosis of neoplasia was confirmed by biopsy in 4 patients (2 after failure of a previous therapeutic test), surgical removal in 7 (all except 1 after unsuccessful therapeutic test), autopsy in 1, biopsy of a muscle lesion in 1, and after failure of therapeutic test with cysticidal drug but without histological confirmation in 1.

Two articles (see Table 3) reported brain neurocysticercosis that mimicked tuberculoma, in a woman (age not specified) and a 20-year-old man, respectively. In both cases, diagnosis was confirmed after resection of the lesion.

Four articles (3 full-texts retrieved) reported tuberculoma that mimicked neurocysticercosis (see Table 4) in 4 patients. Mean age was 22.3 ± 13.4 years (range, 4–41 years). Diagnosis was made after failure of therapeutic test and through MR spectroscopy (2 cases), or polymerase chain reaction on cerebrospinal fluid (1 case).

When the provisional diagnosis was neurocysticercosis, clinicians were more likely to be working in regions endemic for cysticercosis (9 of the 17 cases with final diagnosis of tumor, and 3 of the 4 cases of tuberculoma), while only 9 of the 32 cases with initial diagnosis of tumor or tuberculoma occurred in endemic regions. When the provisional diagnosis was neurocysticercosis and the final diagnosis was tumor, lesions were most often multiple (13 of the 16 cases). Conversely, when the provisional diagnosis was tumor, the number of lesions was 1 or 2 in 20 of the 33 cases (unspecified number in 9 of the 33 cases).

Given the frequent misdiagnosis of parenchymal brain neurocysticercosis, diagnostic criteria have been proposed to help clinicians avoid delayed or inappropriate management [52]. However, often neither brain imaging nor serum and cerebrospinal fluid cysticercosis tests are sufficient for definitive diagnosis. Moreover, positive cysticercosis tests have been reported even when performed on a tumor biopsy sample [41]. For all these reasons, current management of neurocysticercosis remains contradictory and an individual approach is highly recommended [53]. Antiparasitic treatment may be used for viable cysticerci, but is generally not recommended for degenerative cysts. Because of the risk of additional brain damage, surgery is classically not considered, except for neurocysticercosis-induced hydrocephalus, with or without intraventricular or cisternal cysts. However, in case of large parenchymal cyst or uncertain diagnosis, surgical resection can also be indicated [54]. Bulky lesions can not only have major functional impact

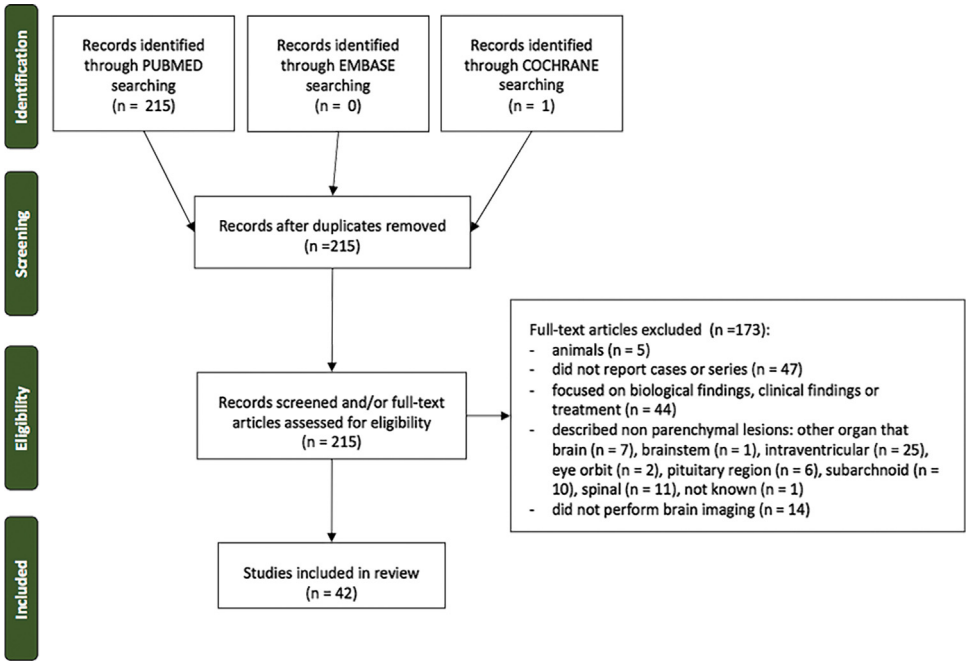

Fig. 4. PRISMA chart for literature review.

but also be life threatening, and removal can resolve the patient's symptoms. If surgery is planned, conserving brain functions is of paramount importance, notably for lesions in eloquent areas. Direct electrical stimulation during awake surgery is the gold standard for detecting functional brain areas [55] and can be of great interest here. Traditionally used in the management of diffuse brain tumors and epilepsy, this technique is increasingly proving useful for other types of brain condition. In the present case, diagnosis could not be confirmed preoperatively despite a large set of laboratory and imaging investigations. In the absence of a lesion for which antiparasitic drugs could be indicated (cysticerci with viable appearance), no therapeutic test was performed. A wait-and-see attitude was rejected, given the large volume of the lesion, its unknown growth potential, and the existing neurological deficit and epilepsy. Despite the location of the lesion in a highly eloquent brain area, resection was safely performed using intraoperative mapping, demonstrating the usefulness of awake surgery in the management of degenerating parenchymal neurocysticercosis.

Informed consent

Informed consent was obtained from all participants included in the study.

Disclosure of interest

The authors declare that they have no competing interest.

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