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Brain angiometastasis from a non-seminomatous germ cell tumor: A case report

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ABSTRACT

INTRODUCTION: Brain metastasis from non-seminomatous germ cell tumors (NSGCT) is rare. Herein, we describe the second reported case of brain metastasis from a NSGCT with high-flow arteriovenous (AV) shunts, and propose a novel surgical treatment plan.
CLINICAL CASE: The patient was a 34-year-old male who presented with hemiparesis and hemianesthesia. Magnetic resonance angiography revealed three vascular lesions with afferent vessels and efferent vessels. Angiography displayed two high-flow AV shunts. During angiography, the patient experienced sudden neurological deterioration and consequently underwent surgery. During surgery, a lesion with large AV shunts was observed, with arterialized drainage veins, pedicled arterial vessels affluent to the nidus, and an absent pial plane. The surgical technique was adapted to lesion morphology using special bipolar forceps. Histological and immunohistochemical tests confirmed that the lesion was a NSGCT.
DISCUSSION: NSGCTs are clinically more aggressive than seminomas. Lesions with an AV shunt and glioma combination are designated as angiogliomas. Therefore, we termed the lesion in the present case as an "angiometastasis," which was formed from numerous AV shunts. The use of presurgical embolization has been reported to improve long-term survival in patients with intra-axial hypervascular tumors with AV shunts.
CONCLUSION: We here propose a novel strategy for the management of hypervascular brain metastasis from NSGC, consisting of angiography, tumor embolization, and the use of an angiometastatic surgical technique with special bipolar forceps. This case report may help neurosurgeons make better surgical decisions in the management of highly vascularized brain metastasis.

1. Introduction

Brain metastases from non-seminomatous germ cell tumors (NSGCTs) are rare, occurring in only 0.5%–1% of NSGCT cases [1–3]. Patients presenting with cerebral metastases are classified as having a "poor prognosis" according to the International Germ Cell Consensus Classification [4]. Currently, the treatment recommendation for this type of tumor is based on results of case series and clinical studies and expert opinions [1–3]. To the best of our knowledge, only one published case has reported on the unusual presentation of highly vascular brain metastasis of a germ cell tumor [5]. Currently, all case reports involving an association between arteriovenous (AV) shunts and tumors have involved tumors of glial origin. Herein, we report the second case of brain metastasis from an NSGCT with high-flow AV shunting, revealed by angiography. We describe the morphology of the brain NSGCT metastasis as well as a novel surgical treatment strategy. This work has been reported in accordance with the SCARE criteria [6].

2. Presentation of case

The patient was a 34-year-old male with the following surgical history: radical left orchiectomy at the age of 33 with a histopathologic report of germ cell mixed tumor and pulmonary metastasectomy. His alpha-fetoprotein level was 1.54 ng/mL, and B-human chorionic gonadotropin (B-HCG) level was <1.00 mUI/L.

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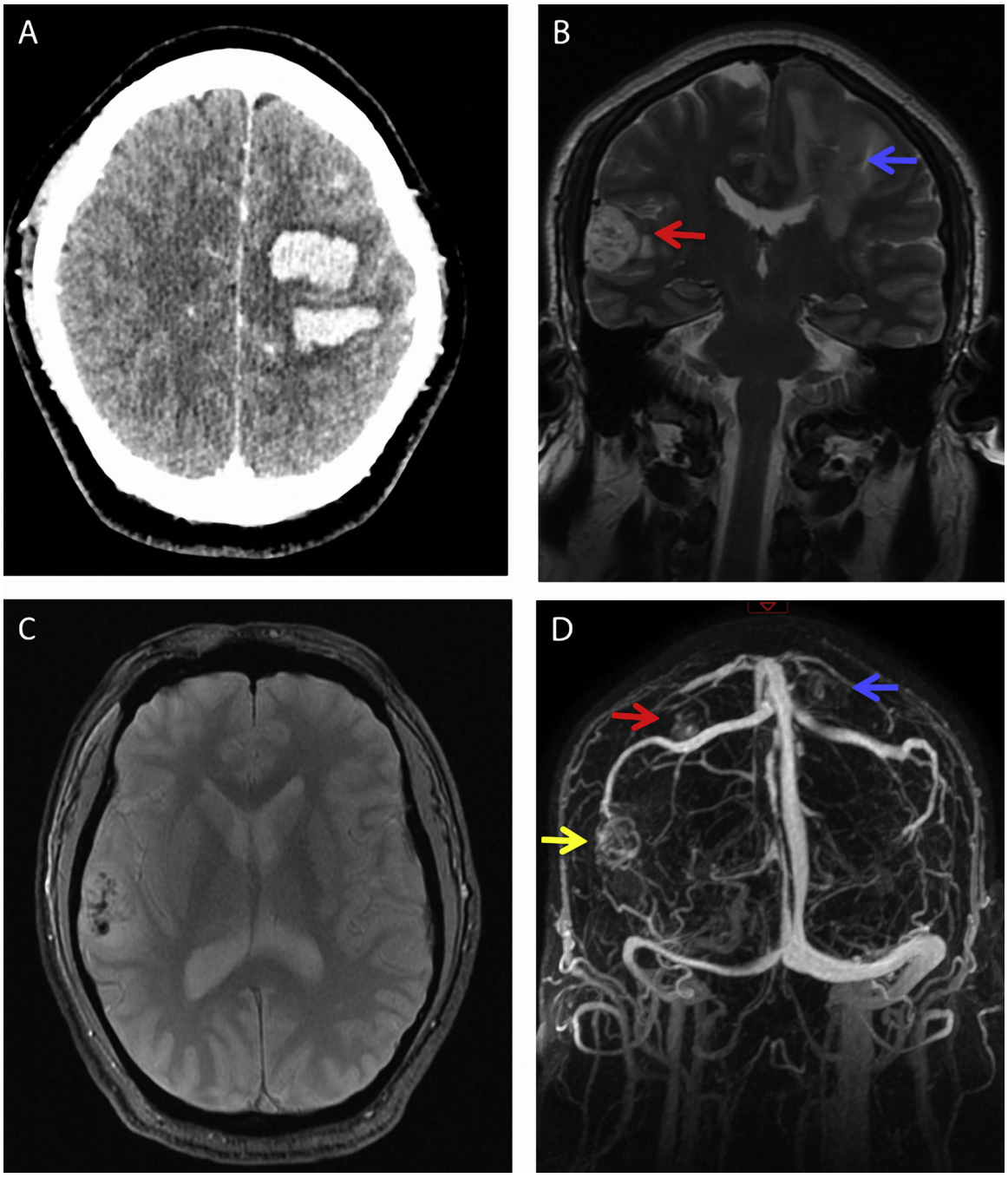

Fig. 1. A) Simple axial computed tomography showing a hemorrhage located in the left hemisphere from the semioval center to the middle frontal, pre-, and post-central gyri. B) Magnetic resonance imaging (MRI) T2 showing two lesions: 1) a heterogeneous lesion in the right superior and middle temporal gyri (red arrow); and 2) a hemorrhagic lesion with edema in the pre- and post-central gyri on the left side (blue arrow). C) MRI T2 gradient-echo showing a lesion located in the temporal lobe, with tubular forms lacking signal. D) MRI showing three vascular lesions (yellow, red, and blue arrows) with afferent vessels (arteries) and efferent vessels (veins).

The patient received additional maintenance polychemotherapy. Laboratory findings showed no further abnormalities.

During examination, the patient was awake and alert with right hemiparesis and hemianesthesia. Computed tomography (CT) performed on admission displayed a hemorrhage in the left frontal lobe. T2-weighted coronal magnetic resonance imaging (MRI) revealed three lesions in the right temporal lobe and left frontal lobe. MRI with T2-weighted gradient-echo sequence revealed a tubular formation with no signal in the temporal lobe. Magnetic resonance angiography (MRA) revealed three vascular lesions with afferent and efferent vessels (Fig. 1). Cerebral angiography displayed two AV shunts (Fig. 2). During angiography, the patient experienced sudden-onset neurological deterioration. CT scan showed a new hemorrhagic lesion in the temporal lobe, with severe cerebral edema (Fig. 2). The hemorrhagic lesion was removed via decompressive craniectomy. During surgery, the lesion was observed to have large AV shunts, arterialized drainage vein, and pedicle arterial vessels affluent to the nidus. The lesion was managed as follows: 1) its borders were exposed (this was challenging because the pial plane was absent); 2) progressive circumferential dissection of the lesion was performed and affluent arterial vessels coagulated and cut, achieving hemostasis was challenging as the feeding vessels reflected their neoplastic infiltration; and 3) the final stage involved drainage of the veins that were coagulated

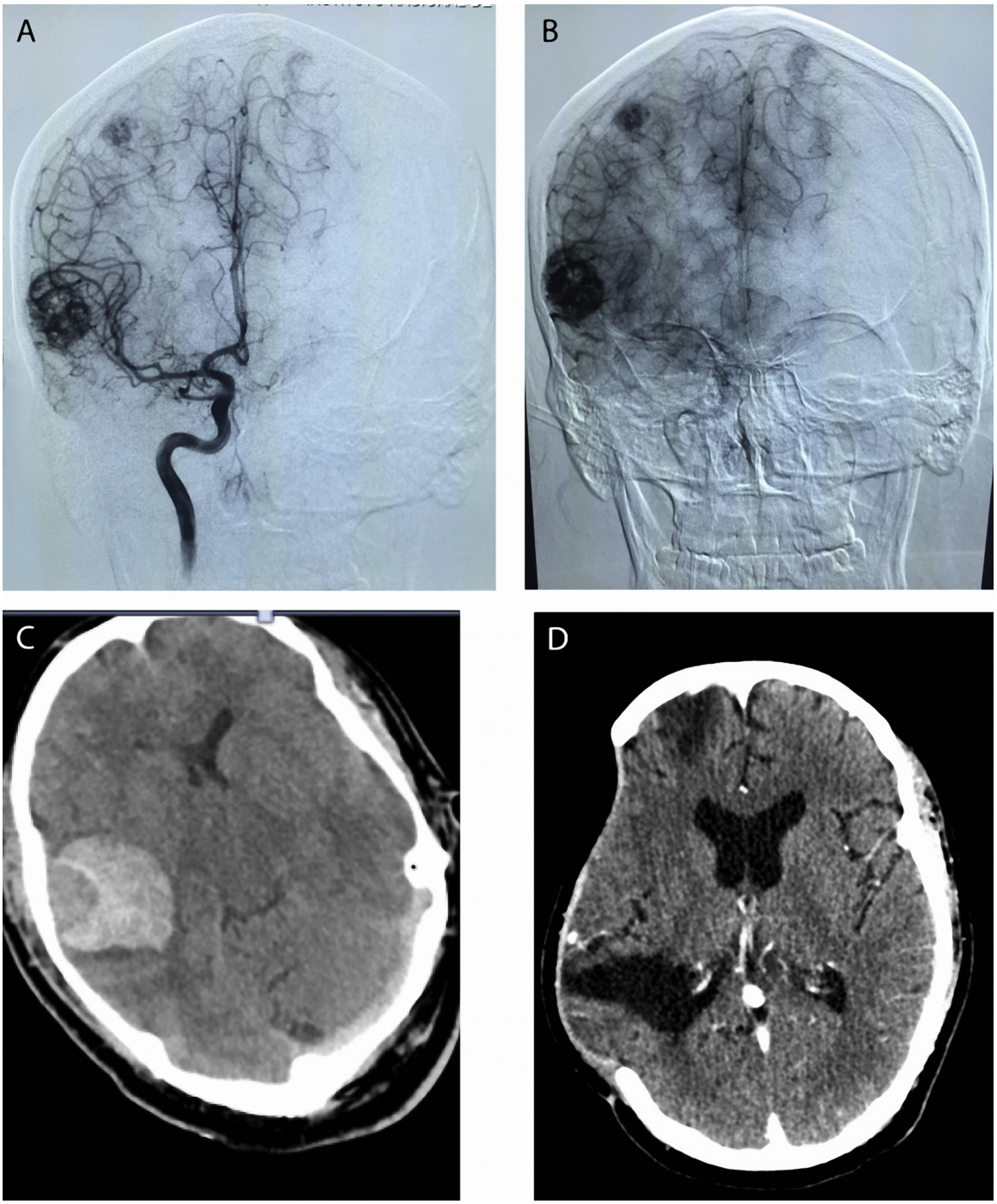

Fig. 2. A) Posteroanterior view of cerebral angiography showed a tangle of serpiginous vessels inside the lesions; 1) one lesion was located in the temporal lobe which was supplied by the anterior and middle temporal artery of the middle cerebral artery; and 2) the second lesion was supplied by the angular artery. B) The late phase of the angiogram showed venous drainage in both lesions. C) Computed tomography (CT) showed a new hemorrhage in the lesion of the temporal lobe with an important mass effect, ventricular compression, severe cerebral edema, and midline deviation. D) Postoperative CT showed a decompressive craniectomy with a complete lesion resection.

and excised. We termed this surgical technique the "angiometastasis technique." Histological and immunohistochemical analyses confirmed that the lesion was a NSGCT (yolk sac tumor) (Fig. 3).

The patient showed good recovery in terms of his overall status in the first month after surgery, and his muscular force was partially recovered and radiotherapy indicated. In the fifth postsurgical month, the remaining brain metastases grew and bled, which caused a fatal intracranial hypertensive with rostrocaudal deterioration and death.

3. Discussion

NSGCTs are clinically more aggressive than seminomas and are considered high-risk when brain metastasis is involved [7]. Although brain MRI is not commonly performed in patients with pulmonary metastasis of NSGCT, the results of several studies have indicated that brain MRI should be performed in such cases [5,7,8]. We believe that performing MRI at an early stage may lead to early detection, which can subsequently improve patient outcome.

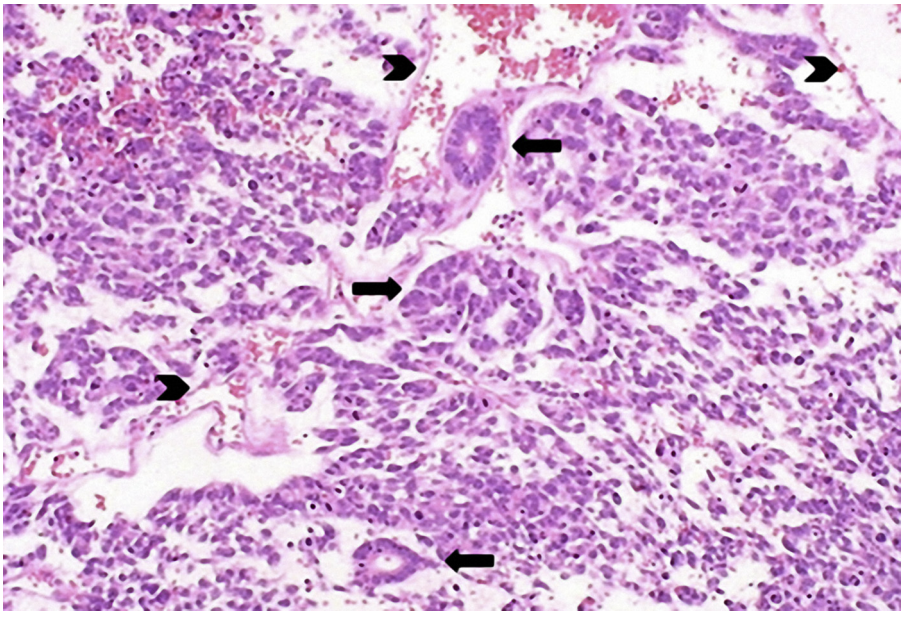

Fig. 3. Round to polygonal cells with nuclear pleomorphism arranged in decohesive nests or irregular anastomosing cords. Note the presence of gland-like tubular structures and numerous blood vessels (arrowhead). Neoplastic cells showed immunoreactivity to SALL-4 (transcription factor). Histological and immunohistochemical findings were consistent with metastatic yolk sac tumor (H&E stain, original magnification = 100×).

The following common characteristics suggest brain metastasis: 1) lack of normalization of tumor markers during chemotherapy; 2) lung metastasis; 3) presence of embryonal or chorionic carcinoma; and 4) highly elevated B-HCG levels (>40,000 U/L) [1,9,10]. In the present case, tumor marker levels were within normal range; however, the patient showed signs of neurological deficit, indicating brain metastasis. Detection of asymptomatic brain metastasis is critical because it can have a strong impact on patient prognosis. In these circumstances, we propose screening more rigorously.

3.1. Hypervascular tumors

Timothy et al. [5] described an unusual presentation of NSGCT metastasis. The patient underwent surgery for intracerebral hemorrhage that was initially thought to be due to an arteriovenous malformation. Results of immunocytological analysis of the brain lesion indicated choriocarcinoma, and imaging results were not well documented. In the present report, we have described a second case of highly vascular brain metastasis from NSGCT. We have presented the relevant angiography imaging results to elucidate the morphology of the lesion, which consisted of AV shunts, feeder vessels, vascular nidus, and drainage vessels of the brain metastasis. This imaging information was critical for planning the initial approach because the angiography scans suggested an AV malformation.

Lesions involving a combination of AV shunt and glioma are designated as angiogliomas [11]. We termed the lesion in the present case "angiometastasis." The lesion was formed by numerous AV shunts, abnormal vasculature, tumor infiltration of the vessels, and tortuous vessels with high-grade bleeding.

Massive hemorrhage primarily occurs in malignant tumors that are highly vascularized [12]. Hemorrhage in brain metastasis from NSGCT could be due to: a) polychemotherapy protocols, which occasionally result in tumor lysis [8]; b) AV shunts; c) abnormally hyalinized vessels; and d) presence of numerous thin-walled vessels [12].

The physiology of the underlying vascular neoformation and AV shunt, which presents in cerebral metastasis, is not well understood. Vascular endothelial growth factor is the most potent angiogenic factor involved in the growth and development of brain metastasis, creating subsequent abnormal AV connections [13].

3.2. A novel surgical plan

New surgical techniques for the management of brain metastasis include: 1) intraoperative MRI; 2) awake surgery; 3) assist-port surgery (BrainPath); 4) neurophysiologic monitoring; 5) intracavitary brachytherapy; 6) laser interstitial thermal therapy; and 7) fluorescence-guided neurosurgery (fluorescein and 5-aminolevulinic acid) [13,14].

The use of presurgical embolization has been reported for the improvement of long-term survival in patients with intra-axial hypervascular tumors involving AV shunts [15]. Endovascular therapy for gliomas consists of the following: 1) neoadjuvant embolization and devascularization; 2) direct intra-arterial drug delivery; and 3) disruption of the blood–brain barrier for improved drug delivery [8]. Currently, there is no general consensus regarding the management of highly vascular brain metastasis. Therefore, we propose the following three-step management plan for the treatment of brain metastasis from NSGCT with high-flow AV shunts. Step 1: Perform cerebral angiography, with the possibility of endovascular therapy to embolize the metastasis. This novel approach should be adopted with caution, as intra-axial tumors are more challenging to embolize because their feeding vessels are more distal and narrow. Step 2: Use the interchangeable tips on bipolar forceps for better control in the neo-angiogenesis region of the tumor's feeding arteries, to allow for a reduction in bleeding when embolization is not possible. Step 3: Consider the unusual morphology of the lesion when planning its resection (i.e., AV shunt, vascular nidus, drainage veins, neoplastic infiltration of the vessel walls, and tumor cellular proliferation) [16]. We termed this surgical technique the "angiometastasis technique" (Fig. 4).

4. Conclusion

This is the second reported case of an association of AV shunts with brain metastasis from NSGCT. We believe that our contribution to this uncommon case adds new knowledge, specifically of

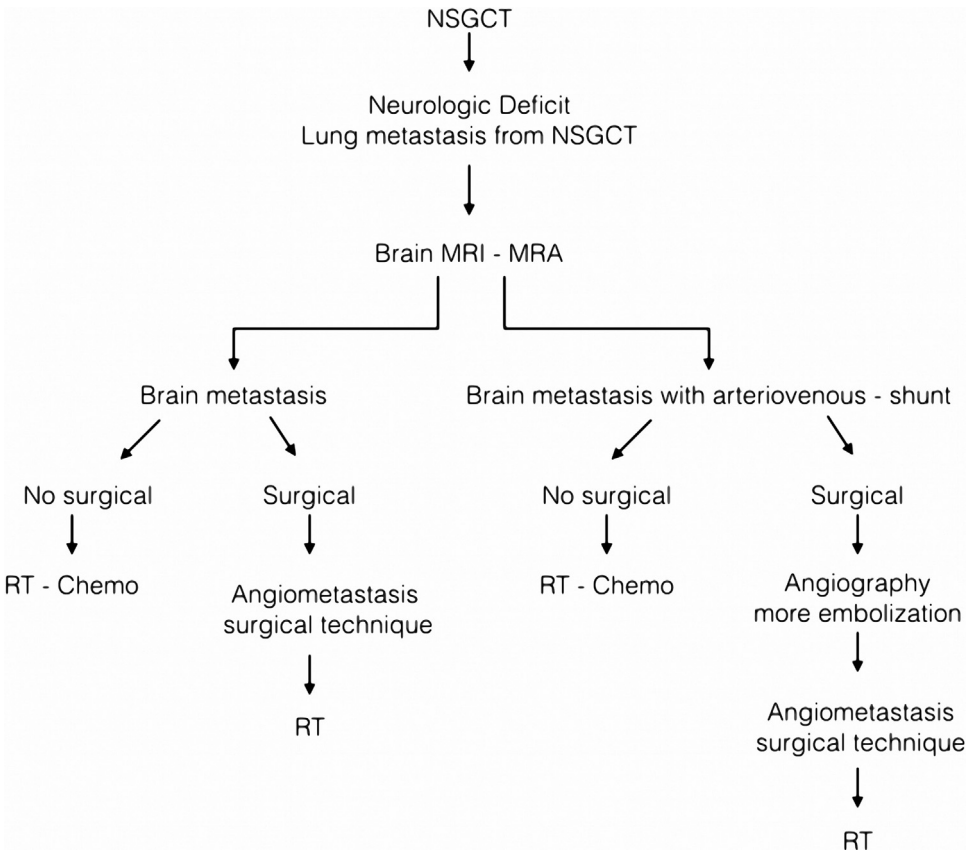

Fig. 4. Flow chart depicting a novel technique with which to manage brain metastasis from non-seminomatous germ cell tumors (NSGCT).

the hypervascular metastasis or "angiometastasis." Its morphology as well as treatment nuances have been previously described in this case report. In the future, brain metastasis treatment should be personalized according to factors related to specific tumor molecular pathways, such as angiogenesis and endothelial proliferation, and results of imaging studies.

Conflicts of interest

We do not have nothing to declare. No conflict of interest.

Funding

We do not have nothing to declare. No sources funding.

Ethical approval

The ethics committee of the Institute approved the submission of the case report. (number 42842-1).

Consent

I confirm that we have patient consent for the publication of this case report.

Author contribution

Alejandro Monroy Sosa, Gervith Reyes Soto, Bernardo Cacho Díaz, Ángel Herrera Gómez: Study concept.

Alejandro Monroy Sosa, Allan Hernández Estrada, Martín Granados García, and Ana María Cano Valdez.: Data collection and Data analysis.

Alejandro Monroy Sosa, Gervith Reyes Soto and Bernardo Cacho Díaz: writing the paper.

Guarantor

Alejandro Monroy Sosa.

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