Case Report

Journal of Case Reports & Imaging

Hemorrhagic Cerebral Metastases Presenting After Complete Resection of Atrial Myxoma: A Case Report with a Favorable Outcome and Review of the Literature

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Abstract

Cardiac myxoma is the most common benign heart tumor. Its complete resection is usually curative; however, sometimes recurs with metastases, exhibiting a malignant potential through mechanisms which remain unclear. The brain is the most frequent metastatic site. With a small number of cases reported in literature, there is currently no standard management for cerebral myxoma metastases, and most of the reported patients have been treated with cerebral surgery, but postoperative chemotherapy and/or radiation have been attempted. We report a case treated for a benign atrial myxoma, which developed multiple brain metastases, in whom a "wait and see" strategy was adopted, delaying surgery approach with a favorable outcome over a prolonged follow-up period.

Keywords: Brain; Cardiac myxoma; Metastases

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Received Date: July 08, 2021

Accepted Date: July 13, 2021

Published Date: July 20, 2021

Citation: Giannoni S, Pizzanelli C, Frosini D, Canovetti S, Mancuso M, et al. (2021) Hemorrhagic Cerebral Metastases Presenting After Complete Resection of Atrial Myxoma: A Case Report with a Favorable Outcome and Review of the Literature. J Case Repo Imag 5: 042.

Case Presentation

A 72-year-old woman was admitted to the emergency room for acute weakness of the right arm and speech impairment.

Ten months before, she underwent resection of an inter-atrial septal cardiac benign myxoma that has manifested embolizing the left upper limb.

Her past medical history also included resection of VIII right cranial nerve for schwannoma at the age of 52 years, with recurrence of this lesion at the age of 55, with hydrocephalus treated with ventriculoperitoneal shunt and complete resection of the lesion and subsequent radiotherapy.

At the neurological examination the patient was drowsy, with fluent aphasia, mild dysarthria, right hemianopsia and a mild weakness of the right arm. As a consequence of the previous neurosurgery, chronic right facial nerve perifer ical palsy and deafness in the right ear were also observed. The acute neurological deficits persisted only a few hours with subsequent complete remission. Transient neurological symptoms and electroencephalographic focal slowing in the left temporo-occipital regions led us to interpret the neurological symptoms as a focal limited epileptic seizure.

In addition to previous neuroradiological features related to the past neurosurgery, the brain CT scan revealed some hyperdense masses of round shapes in the right occipital and left parietal regions (Figure 1, section 1 a-b), interpreted as hemorrhagic metastatic lesions or, alternatively, as abscesses from opportunistic germs. The brain MRI defined these findings better, revealing one nodular lesion in the right occipital pole and a group of numerous and confluent lesions in the left parietal lobe, surrounded by a vasogenic edema, the largestone located in the left supramarginal gyrus and parieto-temporo-occipital carrefour (section 1 c-f). All lesions presented hemorrhagic components in subacute or chronic phases (section 1e-f, g-h), mildly enhancing gadolinium (section 1i-l). There were also signs of leptomeningeal hemosiderosis (section 1 g-h).

In the absence of clinical and biohumoral signs of infection, the neuroradiological features supported the diagnosis of metastatic hemorrhagic lesions. Total body CT scans and F18-fluorodeoxyglucose positron emission tomography were both negative for malignancy, as well as esophagogastro-and colonoscopy. We have therefore assumed that cerebral lesions were secondary to myxoma. Transesophageal echocardiogram demonstrated no recurrence of the tumor.

Given the age of the patient, with an already complex neurosurgical history, the paucisymptomatic presentation of disease, and the site of lesions in eloquent areas, it was preferred not to proceed with surgical options, and to keep a "wait and see" strategy.

Thus, the patient was treated with Dexamethasone 8 mg/die per os for two weeks, which was then slowly tapered down until a low maintenance dose of 0.5-1 mg/die. She was also treated with Clobazam as an antiepileptic drug.

A second total body CT-PET scan, performed 12 months after the first screening, remains negative.

During the follow-up (42 months since the removal of myxoma, 32 months since detection of cerebral involvement) the clinical

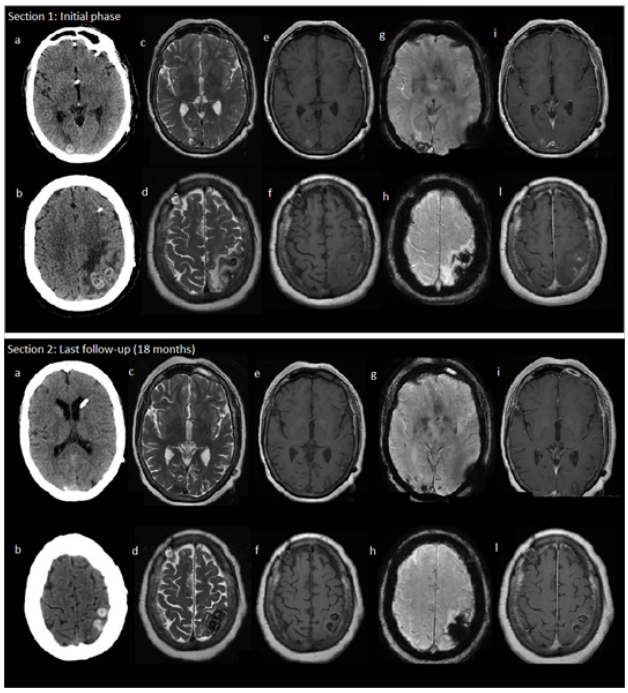

Figure 1: Section 1 shows neuroimaging findings in the initial phase: the brain CT scan shows haemorragic metastatic lesions in the right occipital and left parietal regions (a-b); the MRI shows haemorragic metastatic confluent lesions surrounded by vasogenic edema (c-f) with haemorragic components in subacute or chronic phases (g-h); Metastases were mildly and patchy enhancing gadolinium (i-l). Section 2 shows neuroimaging findings at the last follow up (18 months later - 28 months after complete resection of myxoma): we show a reduction of nodular lesions, in size, vasogenic oedema, contrast enhancement (a-b, c-d, i-l); any new haemorragic lesions was detected, while blood components were detectable as haemosiderine (g-h) Mixed with calcifications (a-b).

situation remained stable. Only a brief epileptic seizure similar to the one described above occurred, and Carbamazepine was started while Clobazam stopped. The series of brain MRIs -performed at 2, 5, 12, 18, months - showed a favorable evolution of the neuroradiological picture with a reduction of nodular lesions, decreasing in both vasogenic edema and enhancement (Figure 1, section 2 a-b, c-d, i-l). No more acute bleeding was detectable but persistent hemosiderin components both in the parenchymal and subarachnoid space were observed (section 2 g-h). CT scans progressively showed increased hyperdensity in some of the cortico-subcortical left parietal lesions consistent with calcifications (section 2, a-b).

Discussion

After a complete curative treatment with local resection, myxomas can rarely cause metastatic disease to the brain [1-5]. In medical literature, including the present case, there only 35 cases of myxoma cerebral metastases (Table 1), that are described with variable outcomes [1-33], including fatal cases [5-11]. Although a standard management of patients with cerebral myxoma metastases has not been established, the most reported treatment is surgery, with or without adjuvant radiotherapy or chemotherapy [2,5,11-16].

Our report shows a patient who has not been treated with brain surgery or adjuvant therapy that experienced a benign course of disease at prolonged follow-up. Over time, our patient had no substantial changes in the neurological status, while the MRI evidence of reduction of metastases size, blood components and vasogenic edema. In addition, we observed the evolution of some of the hemorrhagic lesions into calcifications, supporting the idea of a change of disease from an evolving to an inactive stage.

The natural history of brain metastases of myxoma is extremely variable, as it may be stable over months, or manifesting with

Author	Year of report	Age	Sex	Interval to recurrence (months) since myxoma diagnosis	Other recurrent sites	Surgery for brain lesions	Radiotherapy	Chemotherapy	Outcome follow-up (months)
Our case	2020	72	Female	10	No	No	No	No	AWD42
Ghodasara et al. [16]	2020	63	Female	12	No	No	No	No	-
Rajeshwari et al. [11]	2020	56	Female	-6	-	No	No	No	DOD6
Rajeshwari et al. [11]	2020	17	Male	18	-	Yes	Yes	No	-
Wan et al. [18]	2020	39	Female	7	No	Yes	No	No	AWD18
Roque et al. [13]	2020	48	Female	7	No	No	Yes	No	AWD18
Maas JA et al. [12]	2019	62	Male	12	Fingertips	Yes	No	No	NED48
Asranna et al. [19]	2017	57	Female	12	No	-	-	-	-
Rose et al. Case 15									
2016	44	Male	5	No	No	Yes	No	DOD17	
Rose et al. Case 2 [5]	2016	52	Female	0	No	No	No	Yes	AWD6
Raza and Kamal [1]	2012	47	Female	4	No	Yes	No	No	-
Wolf et al. Case 1 [20]	2008	60	Male	-	No	Yes	No	No	-

Wolf et al. Case 2 [20]	2008	65	Female	-	No	No	No	No	-
Moiyadi et al. [2]	2007	35	Male	48	No	Yes	Yes	No	AWD6
Altundag et al. [14]	2005	41	Female	15	No	Yes	Yes	No	AWD 63
Acikel et al. [21]	2004	58	Female	0	No	No	No	No	-
Hirudayaraj et al. [22]	2004	50	Female	-1	No	Yes	No	No	-
Hou et al. [6]	2001	37	Female	10	Bone	No	No	No	DOD 12
Bernet et al. [15]	1998	31	Male	2	Muscle, lung	Yes	Yes	Yes	NED 120
Scarpelli [23]	1997	64	-	144	No	Yes	No	No	-
Samaratunga et al. [24]	1994	60	Female	-7	No	Yes	No	No	NED 21
Kanda et al. [25]	1994	70	Male	-7	No	Yes	No	No	NED 9
Wada et al. [26]	1993	70	Male	-	No	Yes	No	No	-
Todo et al. [7]	1992	32	Female	10	Jejenum	No	No	No	DOD 10
Chozick et al. [27]	1992	61	Female	-	No	Yes	No	No	-
Kotani et al. [8]	1991	48	Male	3	Soft tissue, aorta	Yes	No	No	DOD 53
Ng and Poon [3]	1990	54	Male	6	No	Yes	No	No	AWD 18
De Morais et al. [9]	1988	73	Male	0	Kidney, pancrea, stomach	No	No	No	DOD 1
Kadota et al. [28]	1987	44	-	3	Skin	Yes	No	No	-
Bazin et al. [29]	1987	56	-	48	No	Yes	No	No	-
Morimoto [30]	1986	44	Female	-	Skin	Yes	No	No	-
Markel et al. [31]	1986	18	Female	30	Bone	No	No	No	AWD 39
Seo et al. [32]	1980	36	Female	96	Bone	Yes	No	No	AWD 120
Budzilovich et al. [10]	1979	52	-	0	No	No	No	No	DOD 1
Rankin and DeSousa [33]	1978	44	Female	96	No	Yes	No	No	AWD120

Table 1: Reported cases in literature of cardiac myxoma metastasizing to the brain whit relative treatment and outcomes.

Abbreviations: NED: No Evidence of Disease; DOD: Dead of Disease; AWD: Alive With Disease. Modified from Rose at al., Moiyadi et al., and Altundag et al.

neurological deficits in a brief period of time with fatal outcome [2,5,12,14]. In the series of 27 cases collected by Maas and collegues [12], eight patients did not undergo brain surgery; among these, only two were still alive after a follow up of 63 and 39 months respectively, five died within 8 months on average, one was lost to follow up. The few heterogeneous cases reported so far does not allow to indicate possible factors for disease progression, although the worst prognosis has been observed in patients who had other recurrent sites of disease in addition to the brain, thus underlining a more aggressive disease in these patients [5,14].

Although the mechanisms by which myxoma may have malignant potential have yet to be elucidated, the presence of robust inflammatory and vasculitic changes associated with metastatic lesions suggest a strong focal inflammatory reaction induced by myxoma [14,17]. In our case, the good response to corticosteroids suggests a main role of inflammation produced by myxoma, more than a malignant local invasiveness of the tumor. However, this finding must be confirmed -or not- in additional cases.

In conclusion, a "wait and see" strategy with a conservative approach may be considered in patients with myxoma brain metastases, as no strong evidence exists to treat these patients with surgery, radio- or chemotherapy. Anti-inflammatory treatments, monitoring clinical and neuroradiological pictures and seizure controls are appropriate options.

Multicenter studies with uniform multidisciplinary clinical and radiological criteria at the follow-up are needed to better characterized the natural history of brain metastases management in myxoma.

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