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Case Report

Acute hemorrhage in a colloid cyst of the third ventricle: A rare cause of sudden deterioration

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Abstract

Background: Acute neurological deterioration and death in a patient harboring a colloid cyst of the third ventricle remains a poorly understood phenomenon. Sudden neurological derangement caused by spontaneous bleeding within a colloid cyst is a rare and potentially fatal event, usually requiring immediate diagnosis and emergency surgical treatment.

Case Description: A 47-year-old male presented with acute right-sided hemiparesis and speech impediment, followed by rapid deterioration of consciousness. Neuroimaging studies showed a rounded mass at the roof of the anterior third ventricle, causing biventricular hydrocephalus along with a left-sided basal ganglia hematoma. The lesion showed scattered foci of a recent hemorrhage which extended into the left lateral ventricle. Surgical treatment involved emergency external ventricular drainage followed by the prompt elective total resection of the lesion via a transcallosal route. Pathological findings confirmed the diagnosis of a colloid cyst with focal areas of vascular congestion and blood extravasation within its wall.

Conclusions: Spontaneous bleeding into a colloid cyst of the third ventricle may cause acute obstructive hydrocephalus and intracranial hypertension due to rapid enlargement of the lesion. This event may account for the sudden neurological deterioration and/or death observed in a previously asymptomatic patient. The diagnosis of hemorrhagic phenomena within a colloid cyst represents a challenge due to the variable signal usually displayed by these lesions on computed tomography (CT) and magnetic resonance imaging (MRI). Emergency ventricular drainage followed by elective tumoral removal constitutes a valid and safe treatment strategy.

Key Words: Basal ganglia hematoma, colloid cyst, spontaneous intracranial hemorrhage, third ventricle, sudden death

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INTRODUCTION

Colloid cysts are benign neoplasms that develop in the

anterior part of the third ventricle roof. [6] They constitute a mere 1% of all intracranial tumors and comprise approximately 15–20% of intraventricular neoplasms, [13]

with an estimated incidence of three people per million per year. [2] The majority of patients affected by colloid cvsts remain asymptomatic. [3] Symptomatic patients are usually diagnosed in their mid-adulthood presenting a wide range of neurological disturbances, from subtle intermittent headaches to coma or even sudden death.^[23] Such high variability in the clinical picture may depend on the adaptation of cerebrospinal fluid (CSF) dynamics in relation to the changes in size and position of the lesion.[23] Nevertheless, the particular mechanisms determining a severe neurological deterioration are still poorly understood. Acute bleeding within a colloid cyst of the third ventricle represents a rare event causing a sudden increase of the cyst volume that may lead to acute obstructive hydrocephalus and rapid neurological derangement. This life-threatening phenomenon has only been reported in nine instances in the scientific literature to date. We report on a patient suffering sudden impairment of consciousness due to acute hemorrhaging within a colloid cyst of the third ventricle, who was successfully treated at our institution. As the analysis of clinical and radiological features of third ventricle colloid cysts involving acute bleeding may contribute to the understanding of the mechnisms leading to a rapid neurological deterioration or sudden death in these patients, we reviewed the historical cohort of welldescribed cases associating this specific event in the literature.

CASE REPORT

A 47-year-old man, with a history of primary systemic hypertension and anxiety disorder with agoraphobia, was admitted in our department with acute-onset speech impediment and loss of strength in the right side of his body, accompanied by headache, vomiting, and deterioration of his level of consciousness. A Glasgow Coma Scale (GCS) score of 8 [E2, V2, M4 (E: Eye response, V: Verbal response; M: Motor response)] and a right-sided hemiplegia were noted during his initial assessment in the emergency room. His arterial blood pressure was found to be 210-140 mmHg. Acute obstructive hydrocephalus due to a rounded hyperdense mass within the third ventricle was revealed on the emergency brain computed tomography (CT), in addition to a moderate size acute hematoma in the left basal ganglia [Figure 1a and b]. The immediate placement of an external ventricular drainage in the frontal horn of the right ventricle allowed controlled evacuation of blood-stained CSF and alleviation of intracranial hypertension (initial value of 32 mmHg). After clinically stabilizing the patient, a presumed diagnosis of a colloid cyst within the third ventricle was made by magnetic resonance imaging (MRI) [Figure 1c]. The tumoral lesion was completely removed 2 days later through a right far lateral interhemispheric transcallosal approach.[13] Pathological analysis of the lesion confirmed the diagnosis of a colloid cyst with vascular congestion and foci of acute bleeding within its wall [Figure 2]. The patient developed postoperative bifrontal pneumocephalus that spontaneously resolved itself over a period of several days. Once the CSF pathways' permeability was restored, the ventricular drain was removed with favorable evolution. Follow-up MRI study confirmed total resection of the

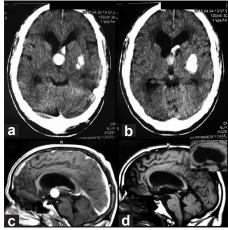


Figure 1:(a, b) Emergency brain CT displaying a left-sided putaminal hematoma of 9 cm3 and a rounded hyperdense lesion of 2.5 cm in its major diameter, located at the anterior third ventricle, which causes acute biventricular hydrocephalus. There are signs of intraventricular bleeding in contact with the outer border of the lesion at the left foramen of Monro. (c) Brain TI-weighted Gadolinium-enhanced MRI. This parasagittal slice shows a homogeneous hyperintense rounded lesion in the anterosuperior part of the third ventricle. (d) Postoperative mid-sagittal slice of a TI-weighted MRI showing no signs of residual tumor at the third ventricular chamber. The inset in the upper right corner corresponds to a right parasagittal slice displaying the small callosal section performed during the surgical approach

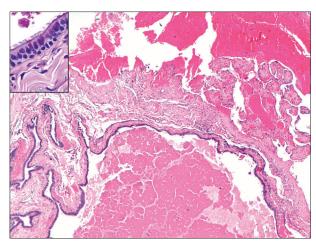


Figure 2: Colloid cyst showing a lining of columnar epithelium and acellular debris in the cavity, with signs of vascular congestion and recent hemorrhage within its wall (H and E, ×100). The high power inset shows ciliated columnar cells and mucin-containing cells (PAS-diastase, ×630)

lesion [Figure 1d]. Right-sided hemiparesis (3/5) with impaired propioception and a mixed type sensitive and motor disphasia persisted at the latest follow-up assessment 1 year after surgery. Neuropsychological assessment revealed bradipsychia, anosognosia, as well as attention, concentration, and working memory significant deficits.

DISCUSSION

Third ventricle colloid cysts are benign neoplasms which originate from ectopic endodermal elements migrated into the velum interpositum during central nervous system development. [6] These lesions are composed of a mixed array of epithelial and goblet cells, the latter secreting proteinaceous material that accumulates progressively within the cyst.[17] The appearance of clinical symptoms in patients afflicted by colloid cysts is considered by most authors to be the result of a CSF dynamics failure to adjust to changes in the volume and/ or position of the colloid cyst.[22] Rapid expansion of a colloid cyst may produce clinical symptoms associated with increased intracranial pressure, while a gradual growth of the lesion usually permits the intracranial pressure to remain within normal levels for longer periods, in spite of the progressive enlargement of the cerebral ventricles. In the latter situation, patients may remain asymptomatic or present with symptoms resembling normal pressure hydrocephalus.[23] Colloid cysts have also been observed to induce marked neuropsychological disorders in patients in whom hydrocephalus was absent. These emotional, behavioral, and memory disturbances are attributed to direct compression or insufficient blood supply to specific diencephalic nuclei and/or pathways functionally related to major limbic circuits. [5,16]

The decision-making process in colloid cyst patients poses a complex task due to the scarcity of scientific information regarding the disease's history and the increasing number of colloid cysts found incidentally on neuroimaging studies. In addition, the accurate incidence and mechanisms involved in sudden death of colloid cyst patients remain poorly understood. [6,7,24] It is known that sudden neurological decline associated with colloid cysts is rare among wholly asymptomatic patients, these lesions being identified as the cause of sudden death in only 0.001-0.009% of autopsies. [9,22] Prompt surgical treatment has traditionally been recommended for patients presenting clinical or radiological progression, with the goal of minimizing the risk of sudden clinical deterioration.^[22] Several surgical modalities are currently used to treat colloid cysts, including microsurgical and endoscopic techniques. [4,8,21,25,27] The former provide a significantly higher percentage of total tumoral removal while the latter are associated with slighty lower complication rates.^[4] Observation and serial neuroimaging are considered a

valid, safe option for the majority of small-size colloid cysts (less than 1 cm) diagnosed incidentally in patients with a normal ventricular system.^[14]

The frequent finding of ventricular enlargement and brain herniation in autopsy specimens displaying a colloid cyst of the third ventricle has led most authors to postulate that hydrocephalus may play a major role in unexpected deterioration. The rapidity at which hydrocephalus develops, more than the degree of ventricular dilation, is the critical factor related to sudden death in these patients.^[5,24] Nevertheless, neither the cyst size nor the degree of ventricular dilation may prove particularly useful variables to predict the risk of sudden neurological deterioration or death. [6] Determining the accurate cause of death in autopsy specimens lacking macroscopic signs of hydrocephalus or brain herniation represents the most intringuing task. In these cases, sudden death has been atributted either to venous infarction or to a rapid compression of the third ventricle walls, producing a reflex irritation of the hypothalamic cardiovascular regulatory centers.^[24]

In 1927, Marchand and Chatagnon reported the first case of a hemorrhagic lesion within the third ventricle in a 57-year-old woman complaining of anxiety with agoraphobia.^[19] Although no accurate or definite histological diagnosis of such a hemorrhagic lesion was provided, this report highlighted the mechanism of sudden spontaneous bleeding within third ventricle lesions as a cause of sudden death. Acute intra-cyst bleeding should therefore be considered an alternative mechanism for sudden deterioration in patients with a third ventricle colloid cyst. This phenomenon has only been reported nine times to date [Table 1]. Thus, considering the overall incidence of colloid cysts, tumoral hemorrhaging should be regarded as an exceptional cause of sudden deterioration in previously asymptomatic patients. Among the nine such patients suffering from sudden impairment of conscioussness, hydrocephalus was a constant finding on neuroradiological studies or during autopsy. [3,10,11,15,18,20,26,29] In these patients, a rapid cyst growth associated with acute intratumoral bleeding could have either caused acute hydrocephalus or worsened a halted hydrocephalus.

It is not clear which factors can facilitate colloid cyst bleeding, nevertheless arterial hypertension^[18] and coagulation disturbances^[12] may play a role. Our patient is, to our knowledge, the first in whom the colloid cyst bleeding coexisted with a basal ganglia hematoma. Our patient suffered from arterial hypertension, thus both basal ganglia hematoma and tumoral bleeding may have been precipitated by a hypertensive crisis. Alternatively, systemic hypertension may be secondary to intracranial hypertension associated with obstructive hydrocephalus in this patient.^[1,6]

Table 1: Cases of hemorrhagic colloid cyst reported in the scientific literature

Case	Author	Age/ sex	Predisposing factor	Previous symptoms	Sudden deterioration	Size (mm)	Hydrocepha- lus	Neuroimaging	CSF shunt/ surgery	Outcome
1	Zeitlin <i>et</i> <i>al.</i> ^[29] 1937	42/F	No	Present	Present	50	Present	No	No/No	Exitus
2	Shuangshoti et al. [26] 1965	32/F	No	Present	Present	28	Present	VG	TS/No	Exitus
3	Kriz ^[15] 1967	15/F	No	Absent	Present	NA	Present	No	No/No	Exitus
4	Malik <i>et</i> <i>al.</i> ^[18] 1980	63/M	Arterial hypertension	Present	Present	20	Present	PEG CT: Hyperdense	TS/No	Exitus
5	Mathiesen et al.[20] 1997	48/M	No	Present	Present	17	Present	MRI: High T1 signal	EVD/STA	Excelent
6	Hadar <i>et</i> <i>al.</i> ^[12] 1999	60/M	Leukemia with thrombocytopenia	Absent	Absent	10	Absent	MRI: High T1 signal	No/No	Exitus (other cause)
7	Beems <i>et al.</i> ^[3] 2006	35/F	No	Present	Present	30	Present	CT: Hyperdense; MRI: High T1 signal	EVD/EPR	Severe neurological sequelae
8	Farooq <i>et al</i> . ^[10] 2008	9/F	No	Present	Present	20	Present	MRI: High T1 signal	EVD/MTR	Excelent
9	Godano <i>et</i> <i>al.</i> ^[11] 2010	29/F	No	Absent	Present	20	Present	MRI: High T1 signal	EVD/ETR	Vegetative state
10	Present case	47/M	Arterial hypertension	Present	Present	25	Present	CT: Hyperdense; MRI: High T1, low T2 signal	EVD/MTR	Dysphasia, hemiparesis, memory disturbance

F: Female, M: Male, CT: Computed tomography, MRI: Magnetic resonance imaging, HCP: Hydrocephalus, EVD: external ventricular drainage; VG: Ventriculography, PEG: Pneumoencephalography, TS: Torkildsen shunt, STA: Stereotactic aspiration, EPR: Endoscopic partial resection, ETR: Endoscopic total resection, MTR: Microsurgical total resection, NA: Not available

Colloid cysts can present wide signal variability on CT and MRI scans due to changes over time in the proportion of their cholesterol, protein, and water contents. [2,28] All hemorrhagic colloid cysts described to date displayed high density/intensity signals in CT/T1-weighted MR scans and a low intensity signal in T2-weighted MRI sequences. As hyperdense/hyperintense CT/MRI signals may mask the presence of bleeding within colloid cysts, preoperative confirmation of such bleeding remains a challenge. [3,10,12,15,18,26,29] In our patient, evidence of ventricular hemorrhage adjacent to the cyst wall on the initial CT allowed us to diagnose tumoral bleeding [Figure 1], which was confirmed later in the biopsy.

All patients treated with emergency external ventricular drainage followed by surgical resection of the mass survived [Table 1]. [3,10,11,20] Two patients, despite being shunted before the hemorrhagic event, also would eventually suffer neurological deterioration and death. [18,26] Acute shunt obstruction related to cyst bleeding was believed to be the cause of the fatal outcome in both instances. This information suggests that the primary objective of treatment must be the rapid alleviation of

hydrocephalus by external ventricular drainage. This procedure must be followed by prompt resection of the tumor in order to prevent new hemorrhagic events which could produce shunt obstruction and/or damage to vital structures located near the third ventricle walls. [11] Surgical strategy – endoscopic, microsurgical or stereotactic aspiration – employed for removal of hemorrhagic colloid cysts did not seem to influence the final outcome [Table 1].

CONCLUSION

Spontaneous, acute bleeding of a third ventricle colloid cyst is a rare, potentially fatal phenomenon due to the rapid development of obstructive hydrocephalus or to the activation of a preexisting arrested hydrocephalus. Factors involved in the bleeding in these benign lesions remain unknown, although hypertensive crises may very well play a role. Detection of bleeding within a colloid cyst is complicated due to wide variations in the CT/MRI scans with these lesions. Surgical management of these patients must include emergency restoration of CSF hydrodynamics using a shunting procedure, followed by prompt surgical removal of the hemorrhagic cyst.

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