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ABSTRACT

Background. Cavernous carotid aneurysms (CCA) are rare aneurysms with a relatively benign natural history. The association between CCA aneurysm and ipsilateral Internal carotid artery (ICA) thrombosis or occlusion has not been described previously. The management of patients with these dual lesions is a challenging problem.

Case description. In this report, we describe an 18-year-old man who presented with left abducent nerve palsy of 3 weeks duration, and imaging revealed left CCA with left ICA occlusion. The patient was managed conservatively with clinical and imaging follow-up. The patient recovered well with complete resolution of clinical symptoms and disappearance of left CCA.

Conclusions. The association of giant CCA and ICA occlusion on the same side is a rare phenomenon with no current consensus on the appropriate follow-up and management strategy. In this report, we described the first case of spontaneous complete disappearance of a giant CCA in the setting of ipsilateral ICA occlusion with complete resolution of symptoms at nine months of follow-up.

INTRODUCTION

Cavernous carotid aneurysms (CCA) are rare with a relatively benign natural history and a lower risk of morbidity and mortality compared to size-matched equivalent aneurysms at other locations [6]. When CCAs are asymptomatic or have mild tolerable symptoms, then conservative treatment can be justified with long-term clinic-radiological follow-up. Other forms of presentations may require a multidisciplinary team of neuroendovascular and micro-neurosurgeons with catheter-based therapy as the treatment of choice [10].

Although partial thrombosis of intracranial aneurysms is reported in

Keywords

cavernous carotid aneurysm,
internal carotid artery,
spontaneous resolution



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large and giant aneurysms, a complete thrombosis is a rare phenomenon [2]. An interesting association is reported between large CCA thrombosis and parent vessel occlusion with many suggested theories [2]. With extreme rarity, a decrease in the size of the thrombosed aneurysm represents a regression in the hemodynamic pressure and indicates a better prognosis [3]. Here we describe the first report on the spontaneous total disappearance of a giant CCA in the setting of ipsilateral internal carotid artery (ICA) occlusion with complete resolution of symptoms.

CASE SCENARIO

An 18-year-old male patient presented to us with left abducent nerve palsy and recurrent attacks of a severe headache of nonspecific localization and character. Apart from the palsy, the patient was neurologically intact and had no remarkable medical history. Brain magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) at presentation showed a giant (28mm x 24mm x 21mm) left cavernous ICA aneurysm with mass effect on the left temporal lobe with a striking shadow of the thrombosed aneurysm and absence of the left ICA proximal to the clinoidal segment on MRA (Figure 1 A-D). The patient couldn't afford the catheter angiography at that time.

We opted to treat him conservatively with regular clinico-radiological follow-up. A 3-month follow-up MRI showed a reduction in the size of the aneurysm, loss of flow void signal intensity, and his symptoms improved. At the 9-month follow-up, the patient was neurologically intact and did not experience headaches over the last three months. His brain MRI and MRA cerebral revealed a complete resolution of the cavernous aneurysm, no mass effect on the temporal lobe with the disappearance of the aneurysmal shadow, and the absence of the left ICA on MRA (Figure 1 E-H). A 9-month follow-up digital subtraction angiography (DSA) showed complete occlusion of the left ICA just distal to its origin with sufficient collateral circulation from the right ICA. No aneurysmal filling was detected (Figure 2). The patient remained asymptomatic at the last follow-up. No further investigations were planned, and annual clinical assessment follow-up visits were scheduled.

DISCUSSION

CCAs are rare and constitute approximately 2-9% of all intracranial aneurysms [5]. They are

predominantly asymptomatic, are discovered incidentally, and have a benign natural history with a low risk of rupture and complications. However, they may become symptomatic due to mass effects from an enlarged aneurysmal mass [6,14]. It is known that CCAs rarely cause rupture and subarachnoid hemorrhage (SAH) since the cavernous sinus is composed of thick double-dural membranes, which lay over the sphenoid body [16]. The optimal management strategy for CCA aneurysms remains controversial. Since CCAs are known to have a low propensity for a life-threatening sequela, conservative management with serial follow-up imaging seems to be a reasonable strategy. However, there are several compelling reasons to treat this entity, including a rupture with the formation of carotid-cavernous fistula, an increase in the size of the aneurysm on follow-up imaging, progressively worsening headache, and erosions of the sphenoid sinus. These signs and symptoms should be analyzed by a multidisciplinary team (neuro-endovascular and microsurgical) to choose the optimal treatment plan [1,6,14].

Aneurysms arising from the circle of Willis are likely to cause spontaneous SAH leading to a devastating neurological condition. On rare occasions, these aneurysms may undergo spontaneous thrombosis causing symptoms related to a local mass effect and or distal thromboembolism of the parent artery. Complete thrombosis of aneurysms occurs at a lower incidence in comparison to partial thrombosis (13-20% vs. 60%); generally, it tends to occur in large (>15mm) and giant (>25mm) aneurysms [2].

The association of ipsilateral ICA occlusion with giant CCA is rare, with only 18 reported cases in the literature [1]. Theories addressing the coexistence of these two pathologies and their progression is a matter of contentious debate due to a limited number of reported cases and the variability in the characteristics, progression, and management strategies. One explanation is that the large thrombosed CCA tends to compress on the ICA against the anterior clinoid process and causes stenosis of the parent artery [8,15]. Other proposed theories include direct stretch and compression of the parent artery by the giant aneurysm and proximal propagation of an intramural thrombus [12]. On the other hand, thrombosis of the parent artery can be initially formed in association with giant

CCA due to the presence of tough dural folds, bony structures along with the natural bend of the artery at this location, which facilitate parent artery deformity, blood flow stasis and eventually thrombosis. The resultant decreased flow into the aneurysm leads to intra-aneurysmal clot formation [2].

In our case, the treatment decision was challenging because the patient was young and had

an acute presentation with abducent palsy of 3 weeks duration. However, the absence of ICA ipsilateral to the giant CCA led us to choose conservative management with regular follow-up imaging in the absence of a defined management strategy for these lesions. Then the patient did well with complete resolution of symptoms at nine months follow-up.

Table 1. Characteristics and follow-up of patients with CCA aneurysm and ipsilateral ICA occlusion that were treated conservatively

Authors	Age-Year/ Sex	Side	Clinical presentation	Imaging at presentation (MRI and DSA)	Imaging at F/U (MRI and DSA)	Clinical outcome	Treatment
Gautier, 1986. (14)	65/F	left	CN III, IV, VI palsies	CCA: (23mm) Thrombosed ICA: occluded	-	Ophthalmoparesis persisted	The patient refused treatment
Sato et al. 1990 (15)	49/M	left	Headache, CN III, VI, V1 palsies	CCA: (3cm) partial thrombosed (MRI flow void) ICA: occluded	-	improved	
Kurokawa et al. 2001. (10)	50/F	Right	Headache, CN III, VI, and V1 palsies.	CCA: (25 mm) not thrombosed. ICA: Intact.	CCA: has heterogenous thrombus on MRI but has no flow in DSA. ICA: Occlusion of petrous segment. (Time from diagnosis: 2 years).	Only CN III palsy resolved.	The patient refused treatment.
Tsutsumi et al., et 2002 (11)	74/M	right	Cavernous sinus syndrome	CCA: complete thrombosis (no flow on DSA) ICA: occluded	-	NA	
Dehdashti et al. 2003 (4)	31/F	Right	Headache, CN III, and VI palsies.	CCA: (30 mm) has heterogenous thrombus on MRI but has no flow in DSA. ICA: Intact.	CCA: partial resolution (aneurysm decrease in size). ICA: Occlusion at the origin. (Time from diagnosis: 18 months).	Neurologically intact.	The patient refused treatment.
Perrini et al., et 2005(16)	47/M	Right	Headache, CN V1, V2 palsies.	CCA: (28mm), complete thrombosis ICA: mild stenosis	ICA: Occlusion at the origin. (Time from diagnosis: 6 weeks)	CN V palsy persist headache and diplopia resolved	Treated conservatively with anti-platelet
Vasconcellos et al., et 2009 (7)	47/F	Right	Headache, CN III, IV, V1, V2, VI palsies.	- CCA: giant	ICA: Occlusion (Time from diagnosis: 6 months)	CN V palsy persist headache and diplopia resolved	

	44/F	Left	Headache, CN III, IV, VI, V 1, V2 palsies.	CCA: giant	ICA: Occlusion (Time from diagnosis: 6 months)	CN VI palsy persist headache and diplopia resolved	
	65/F	Right	Headache, CN III, IV, VI, V1 palsies.	CCA: giant, partial thrombosis ICA: partial occlusion	ICA: Occlusion (Time from diagnosis: 5 years)	CN palsies persist, headache and diplopia resolved	
	84/F	Left	Headache, CN III, IV, VI, V 1, V2 palsies.	CCA: giant, partial thrombosis (heterogenous thrombus on MRI but has no flow in DSA). ICA: Occlusion at the origin.	-	CN III, VI palsies persist, CN IV, V partially improved, headache and diplopia resolved	
Sastri <i>al.</i> , et 2013 (9)	65/F	Left	CN III, IV, VI, V 1, V2 palsies.	CCA: giant, partial thrombosis. ICA: Occlusion at the origin.	-	Ophthalmoparesis improved, CN V persist	Treated conservatively on aspirin
	55/F	Left	Seizure, decrease vision, CN III, IV, VI, V1, V2 palsies.	CCA: (28mm) partial thrombosis ICA: Occlusion at the origin.	-	Ophthalmoparesis improved, Decreased Vision persist	Treated conservatively on aspirin
Das <i>al.</i> , et 2018(3)	45/M	Left	Headache, transient unconsciousness	CCA: (23mm) partial thrombosis ICA: Occlusion at the origin.	-	Neurologically intact	Treated conservatively on aspirin
Present cases 2022	18/M	Left	Headache, CN VI palsy.	CCA: (28 mm) has heterogenous thrombus on MRI but has no flow in DSA. ICA: Occlusion.	CCA: complete resolution (no aneurysm detected). ICA: Occlusion at the origin. (Time from diagnosis: 9 months).	Neurologically intact.	Treated conservatively on aspirin
Shaded rows represent the patients with complete follow-up imaging. ICA - Internal carotid artery, DSA - Digital subtraction angiogram, MRI - Magnetic resonance imaging, CCA - Cavernous carotid aneurysm.							

In our case, a total of fourteen patients with concurrent ICA occlusion and CCAs have been treated conservatively, highlighting the natural history of this unusual association [2, 3, 4, 8, 16, 11 - 13, 15] (Table 1). Unfortunately, follow-up imaging was reported only in 7 patients, and, more specifically, concurrent follow-up MRI and DSA were

available only in 3. Hence, only these 3 cases can be used to assess the actual progression of this disease with a high degree of certainty (Table 1-shaded rows) [3,8].

Lawton *et al.* proposed a new classification of thrombotic aneurysms based on anatomic relationships between the aneurysm, thrombus, and

lumen [9]. They defined the completely thrombosed aneurysm as one with the classic signals of thrombus on the brain, Computed tomography (CT) scan/MRI, and the DSA showing no filling of the lumen [9]. Of the abovementioned three cases, two cases had a complete thrombosis at presentation, and they end with varying degrees of aneurysmal regression on follow-up [3]. The other patient had a non-thrombosed aneurysm at the initial imaging, which progressed to complete thrombosis at the 2-year follow-up [8]. The association between the CCA-ICA occlusion, degree of thrombosis, and outcome cannot be inferred due to the limited number of cases and limited follow-up.

The timeline for the occurrence of the two pathologies (i.e., CCA and ICA occlusion) and which one is present at the presentation, and which one progress with the patient's symptoms is to be answered. For the fourteen cases that have been treated conservatively, eight patients had complete occlusion of the ICA at the presentation. The other six patients have either intact ICA at the initial

investigation (2/14) or partially stenosed ICA at the presentation (2/14), with the initial occlusion status of the remaining two patients not mentioned. All the six cases of initially not occluded ICA end with complete occlusion but with variable duration and extension of occlusion. Those cases with delayed occlusion of ICA in the association of ipsilateral CCA (6/14) had a reported time for the occlusion to occur to be from 6 weeks to 5 years with a mean of 18.9 months. The location of the occlusion is at the origin of the ICA from the common carotid artery, with only one patient having a localized occlusion of the petrous and cavernous ICA [2, 3, 4, 8, 16, 11-13, 15] (Table 1). Obviously, the limited number of cases produces this heterogeneity in the reported findings, and future studies are recommended to continue recruiting more patients and equally important that the patients should have a full radiological follow-up. This will have a profound impact on understanding this unique association, its related pathophysiology, and, most importantly, how to optimize treatment and prevention.

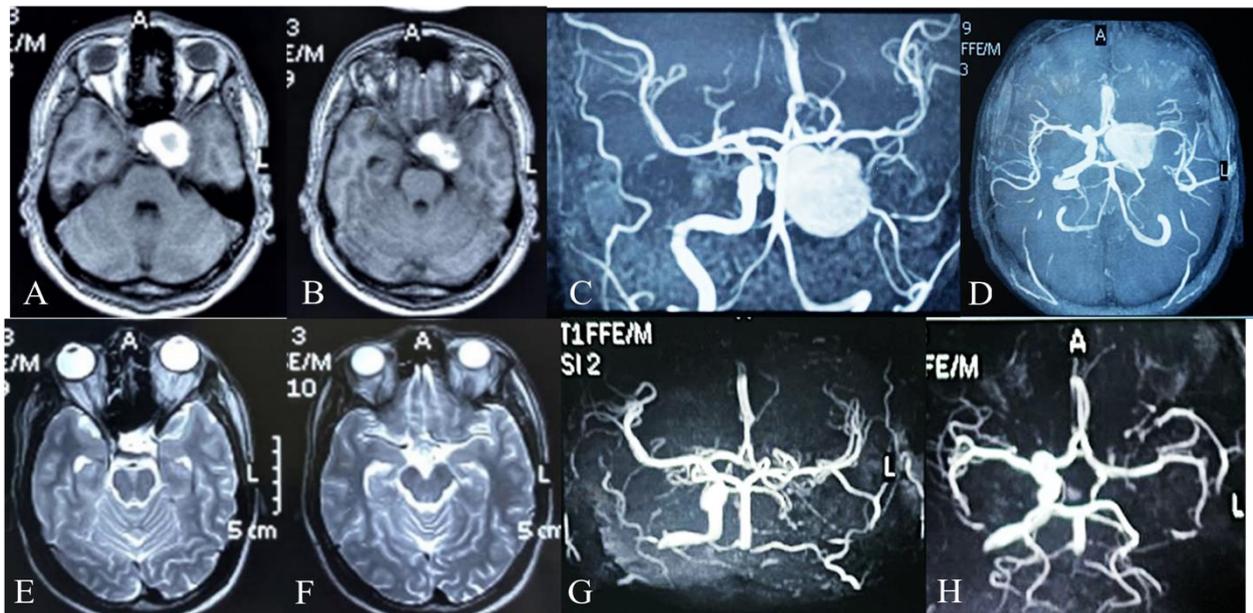


Figure 1. Brain MRI and MRA At presentation (A-D) showed a giant Left cavernous ICA aneurysm with mass effect on the left temporal lobe (A, B) with a striking shadow of the thrombosed aneurysm and absence of the left ICA proximal to the clinoidal segment in MRA (C, D). At 9-month follow-up (E-H) there is complete resolution of the aneurysm, no mass effect on the temporal lobe (E, F) with the disappearance of the aneurysmal shadow, and the same absence of the left ICA MRA (G, H).

CONCLUSION

The association of giant CCA and ICA occlusion on the same side is a rare phenomenon with no current consensus on the appropriate follow-up and management strategy. In this report, we described

the first case of spontaneous complete disappearance of a giant CCA in the setting of ipsilateral ICA occlusion with complete resolution of symptoms at nine months of follow-up.

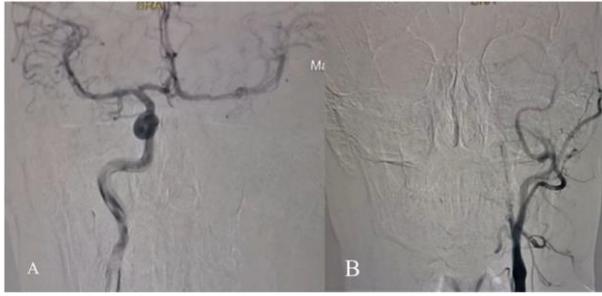


Figure 2. A 9-month follow-up DSA showed complete occlusion of the left ICA just distal to its origin with sufficient collateral circulation from the right ICA. No aneurysmal filling was detected in both (A) and (B).

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