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The complementary multimodal treatment of recalcitrant cerebral aneurysms. Two centres experience

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Abstract

Background: The main treatment of cerebral aneurysms is the direct surgical clipping or endovascular coil embolization. However, some cerebral aneurysms that we reviewed in the literature are still not susceptible to a single treatment approach. These aneurysms can be referred to as complex aneurysms.

Objective: We aim to report these aneurysms and share our clinical experience with their treatment and diagnosis.

Methods: All cases of cerebral aneurysms treated in New York University and in Mansoura University from 2010-2021 were retrospectively reviewed.

Results: 18 patients with 21 cerebral aneurysms were treated by combined surgical and endovascular modalities. Aneurysms associated with arteriovenous malformations (AVMs) in 3 patients, associated with vasospasm in 7 patients, and 3 patients had double aneurysms. A total of 18 patients with aneurysms were treated with combined endovascular and microsurgical therapy. Early angiogram (< 1 week) reveled; complete obliteration of 19 aneurysms (90%) out of total 21 aneurysms, residual filling was observed in 2 aneurysms (10%). Late radiological follow up (> 3 months- 2 years) reveled; a stable residual filling in one and the other case underwent retreatment.

Conclusions: The recalcitrant or complex cerebral aneurysms can be better referred to as diseases rather than lesions as many clinical and anatomical factors make their treatment difficult. Endovascular and microsurgery could be complementary to each other and create a multimodal approach for treating them.

Keywords
complex aneurysms, cerebral, multimodal, complementary treatment

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Background

Direct surgical clipping or endovascular coil embolization is the most common treatment for cerebral aneurysms. (4) Despite significant advancements in microsurgical techniques and equipment, as well as endovascular devices in a parallel direction, some brain aneurysms that we evaluated in the literature are still not susceptible to a single treatment approach. These aneurysms can be referred to as recalcitrant or complex aneurysms. (6,8,10-12,21)

We aim to report these challenging aneurysms and share our clinical experience with their treatment and diagnosis.

Methodology

After IRB approval for both New York University and Mansoura University, all cases of cerebral aneurysms treated in New York University and in Mansoura University from 2010-2021 were retrospectively reviewed.

Inclusion criteria

Patients proved radiological (CT or MR Angiogram or digital subtraction angiography) to harbor cerebral aneurysms that was treated by both endovascular and open surgery with any of the following criteria:

1. Ruptured or unruptured
2. Single or multiple
3. Giant and/or widely necked aneurysms
4. With or without AVMs
5. With or without vasospasm
6. Compressing or involving origin of adjacent arteries.
7. Aneurysms were primary managed with surgery or endovascular embolization and showed residual or recurrence on follow-up.

Exclusion criteria

Aneurysms that were treated by one modality only (endovascular or open surgery).

Patient demographics, aneurysm characteristics, procedural details, clinical outcome (Glasgow outcome scale) and radiological follow-up were analyzed. The radiological follow-up was achieved using digital subtraction angiography (DSA) and/or magnetic resonance angiography (MRA) and/or CT scan angiography (CTA).

Results

18 patients harboring cerebral aneurysms were found to be treated by combined surgical and endovascular modalities.

Patient demographics

There were 11 females and 7 males. The mean age was 58 years (age range, 40–70 year).

Characteristics of aneurysms

In total, there were 21 aneurysms in 18 patients. Three patients had aneurysms linked with arteriovenous malformations (AVMs), seven patients had vasospasm, and three patients had double aneurysms (Figure1).

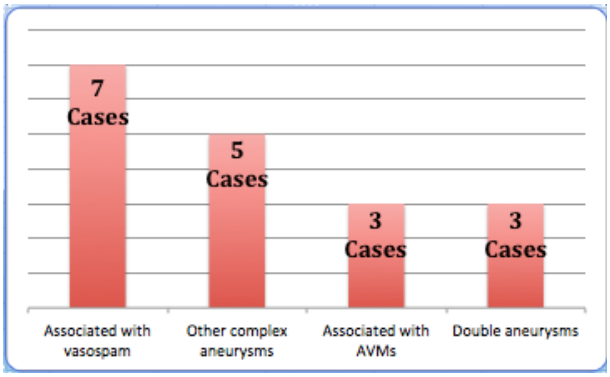

Figure 1. Diagram illustrating the included aneurysm cases.

Aneurysmal rupture and/or bleeding from associated AVMs were the commonest presentation, in 14 patients. The admission Hunt and Hess (HH) grade distribution of this patient group was as follows: Grade I, II 9 patients; and Grade III or more, 5 patients. The other 4 patients with unruptured aneurysms were as follow: incidentally discovered 1 patient during screening for familial aneurysm; 1 patient presented with third cranial palsy; 1 patient presented with agitation and confusion and another patient with aneurysm associated AVM was presented by convulsions.

The mean aneurysm size was 8.5 millimeters (mm). There were 16 small aneurysms (up to 10 mm diameter), and 5 large aneurysms (15–28 mm in diameter). 9 aneurysms had a wide neck (>3mm in width), 6 were saccular in shape and 4 were lobulated. The most common locations of the aneurysms were the posterior communicating artery (PComma) and middle cerebral artery (MCA) and for the AVMs associated with the aneurysms were as follows: 2 cerebellar and 1 temporal. (Table 1)

Table 1. Aneurysmal location

Parent artery	Number (n)
PComm	5
MCA	4
AComm	3
ICA –bifurcation-	3
ACA	2
PICA-AICA complex	1
SCA	1
Anterior choridal	1
Ophthalmic	1

PComm, posterior communicating; AComm, anterior communicating; PICA-AICA, posterior inferior cerebellar artery anterior inferior cerebellar artery; SCA, Superior cerebellar artery.

Treatment techniques

A total of 18 patients with aneurysms were treated with combined endovascular and open surgery. 1 case with residual aneurysmal neck following surgical clipping underwent coil embolization. 3 cases with recurrent aneurysm following pervious coil embolization underwent surgical clipping. 3 cases of double aneurysms underwent coil embolization and surgical clipping. 4 cases of AVM flow related aneurysms underwent partial embolization of the nidus and coil embolization of aneurysm followed by surgical resection. 7 cases developed cerebral vasospasm following surgical clipping underwent balloon and pharmaceutical angioplasty. To facilitate surgical clipping: preoperative balloon occlusion test (BTO) and intraoperative DSA was applied in 3 and 7 cases receptively. (Figure 2 and 3)

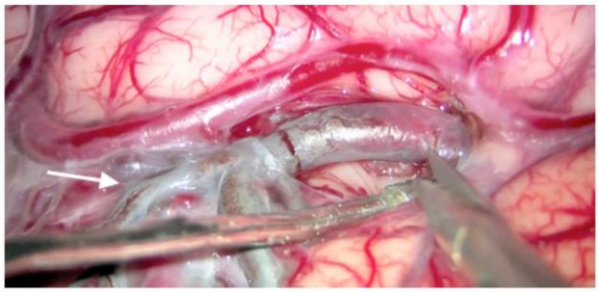

Figure 2. An intraoperative image showing complementary surgical resection of AVM after pervious endovascular onyx embolization of the AVM associated with an aneurysm. A white arrow points to onyx in the nidus.

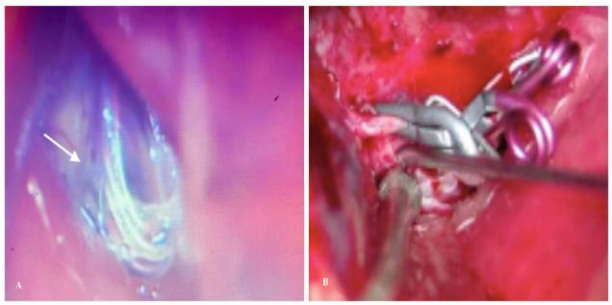

Figure 3. (**A**): An intraoperative image showing a pervious coiled aneurysm that developed significant recurrence (white arrow points to the coil in the aneurysmal sac). (**B**): An intraoperative image showing clipping of the same aneurysm in figure 3A with two clips after removal of the coils.

Outcome

The average radiological and clinical follow-up intervals were 13 and 16 months, respectively. All patients had an early (one week) postoperative radiological follow-up. An early angiography revealed full obliteration of 19 aneurysms (90%) out of a total of 21 aneurysms, with residual filling found in 2 aneurysms (10%), which were followed up on. Late radiological follow up (> 3months- 2 years) was done for the patients on follow-up visits. A stable residual filling in one case and the other underwent retreatment (clipping with intraoperative DSA guidance). (Table 2)

Table 2. Summary of the clinical outcome

Patients	Clinical outcome prior to discharge (< 1 month) (N)*			Late outcome (>3 months)
	Neuro logically intact	Same neurol ogical deficit post-manag ement	New neurologi cal deficit post-manage ment	
Individual Aneurysms	17	0	1**	No significant change
Aneurysms associated with AVMs	2	0	1***	Complete improvement

Neurological status factors: Glasgow coma score, motor, cranial nerve and cognitive functions; other psychological and intellectual assessment were not included.

* Number of patients.

** 1 patient developed monocular blindness post ophthalmic artery aneurysmal clipping postoperative angiogram revealed patent ophthalmic and orbital branches though ophthalmology consult revealed no optic disc abnormalities, but diagnosis was posterior ischemic neuropathy with poor prognosis.

*** 1 patient developed right lower limb weakness grade 4+ and positive Romberg, patient had avulsed PICA during AVM embolization and sacrifice the vertebral artery was done, later developed massive cerebellar infarction later on, however gradual improvement was not noticed during outpatient clinic visits in 6 months.

Discussion

Complex and recalcitrant aneurysms

Many researches had attempted to categorize recalcitrant aneurysms in some way. Hacin-Bey et al., as a combination of anatomical aneurysm variables and clinical factors describe complex aneurysm characteristics. These characteristics are summarized in a table in his study. (8) (See Table 3)

Table 3. Hacin-Bey et al. table of features that define recalcitrant or complex aneurysms (7).

Aneurysm anatomy (best assessed by 3D aneurysm reconstruction from DSA or CTA data)	Clinical features (detailed clinical risk stratification important)
Size: large or giant, too small for a clip or coil	Clinical grade at presentation: HH_3.
Shape: fusiform, serpentine, pseudoaneurysm, dissecting aneurysm.	Timing: vasospasm at the time of presentation.
Content: filled with thrombus, calcified wall, dysplastic vessel wall.	Medical comorbidities: cardiovascular, pulmonary, renal or endocrine comorbidity.
Neck: difficult surgical access, broad, calcified, involving perforator vessels, and other branching vessels.	Advanced age.
Perianeurysmal environmental: aneurysm embedded ineloquent brain tissue, bone, edema, scar from previous surgery.	
3D, three dimensional: DSA, digital subtraction angiography: CTA, computed tomography angiography; HH, Hunt and Hess. Used Table from with permission from Wolters Kluwer Health with modifications.	

The ISAT (International Subarachnoid Aneurysm Trial) requires subjective agreement that an aneurysm might be treated by endovascular or open surgery (5). Many aneurysms, however, did not fit the requirements, such as: 1- patients with life-threatening intracerebral or subdural hematomas; 2- incompatible neck-to-dome ratios; 3-parent artery or branch artery incorporation into the dome; 4-fusiform aneurysms; 5-thrombotic aneurysms; 6-giants; 7-blisters; 8- pseudo/traumatic aneurysms; 9-those with mass effect; and 10-those that had failed repeated endovascular treatment (16,20).

Complementary multimodal therapeutic approach

Choudhri et al described the Stanford neurosurgical experience with the combination of endovascular and open surgery in 67 cases. The aneurysms in all of the individuals in the study were completely obliterated, with no mortality. (2) The combination method was used to treat a total of 96 aneurysms, according to Lawton et al. The aneurysms were enormous or giant in size in 43% of the cases, and fusiform or dolichoectatic in 34%. In 91 aneurysms, the angiographic obliteration was complete (95%) (15).

Chen et al. and Cockroft et al., both had reported the treatment of recalcitrant cerebral aneurysms by surgical reconstruction of aneurysm neck followed by endovascular coiling on a planned concept. Cockroft et al. also reported in his series the initial coil embolization of ruptured basilar tip aneurysm to reduce the risk of rebleeding followed by permanent surgical clipping (1,3).

The radiological and clinical out come of this multimodal treatment in these reported studies and our study was favorable. These good results of combing both surgical and endovascular techniques encouraged the evolution of a new treatment modality, which is the hybrid cerebrovascular surgery.

Hybrid cerebrovascular surgery era

In several subspecialties, hybrid surgery is regarded as a cutting-edge technique. The term "hybrid" refers to a combination of standard surgical and endovascular methods. Actually, it's a multimodal technique that may be done in one session or over a period of time (scheduled). The use of hybrid surgery in the treatment of cardiovascular disorders has ushered in a new era in disease management.(18,19)

In the same direction for the cerebrovascular diseases, hybrid operative theatres have been innovated in the neurosurgical institutes in last few years. Between November 2003 and August 2011, Muryma et al. and Kurtia et al. published two case series of patients with intractable complicated cerebrovascular lesions who were treated with a combination strategy (endovascular and surgical). (14,17) Other case reports were documented in the literature too. (5,7,9,13) A furthermore studies are expected be conducted on a larger scale concerning the hybrid cerebrovascular surgery in the nearby future.

Conclusion

The recalcitrant or complex cerebral aneurysms can be better referred to as diseases rather than lesions; of which many clinical factors in add to the anatomical one define their complexity and make their treatment difficult. Endovascular and open surgery could be complementary to each other and create a multimodal or combined approach for treating these aneurysms, and that concept has

evolved to what known nowadays as the hybrid cerebrovascular surgery.

Contributorship statement

Mohamed Deniwar, Mohamed Kassem and Ashraf Ezz Eldin designed the study. Mohamed deniwar, Mohamed Kassem, Ashraf Ezz Eldin and Jafar J. Jafar, participated in data extraction, analysis, writing and drafting of the manuscript, Jafar J. Jafar critically revised the manuscript and all authors approved the final version.

IRB approval

After IRB approval for both New York University (i14-01394) and Mansoura University (R.21.03.1240), all cases of cerebral aneurysms treated in New York University and in Mansoura University from 2010-2021 were retrospectively reviewed.

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