Clip-wrapping of ruptured blood blister-like aneurysms of the internal carotid artery

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CLIP-WRAPPING OF RUPTURED BLOOD-BLISTERLIKE ANEURYSMS OF THE INTERNAL CAROTID ARTERY

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ABSTRACT (250 words)

OBJECTIVE:

To evaluate the safety and validity of microsurgical Gore-Tex clip-wrapping for ruptured blood-blisterlike aneurysms (BBAs) of the internal carotid artery (ICA).

METHODS:

Single-surgeon series. Retrospective study. Data analyzed were patient age, sex, Hunt and Hess grade (H&H), Fisher grade, time from rupture to hospitalization, aneurysm size and location, collateral capacity of the circle of Willis, time from hospitalization to aneurysm repair, aneurysm obliteration, complications, and modified Rankin scale (mRS) at follow-up.

RESULTS:

Six patients (2 males, 4 females) with ICA BBAs underwent clip-wrapping between 2011 and 2016. Median age was 50.0 years (range 31.7-54.0). H&H was grade 1 or 2 in 5 patients and grade 4 in 1 patient. All aneurysms were small (≤0.3 cm), without relation to vessel bifurcations, and located anteromedially, anterolaterally and superomedially on the ICA trunk. Right side was most frequent (66%). All aneurysms could be clip-wrapped without intraoperative ruptures and all parent arteries could be preserved in the acute phase. Clip-wrapping completely eliminated the BBAs in five patients, whereas one patient underwent additional stent-assisted coiling. There were no re-ruptures, delayed infarctions or deaths postoperatively. After a median follow-up of 57 months (range 20-90), outcome was favorable (mRS score 0-2) in all patients without radiographic signs of recurrences.

CONCLUSIONS:

Using the Gore-Tex clip-wrapping technique for ruptured ICA BBAs, the aneurysm could be treated without intraoperative ruptures or parent artery sacrifice in the acute phase. The outcomes were similar to other SAH patients and on follow-up, there were no aneurysm recurrences, indicating a durable long-term outcome.

Keywords: Aneurysm, blood blister-like aneurysm, internal carotid artery, subarachnoid hemorrhage

Abbreviations used in this paper: ICA = Internal Carotid Artery; BBA = Blood Blister-like Aneurysm

INTRODUCTION

Blood-blisterlike aneurysms (BBAs) are a rare subtype of intracranial pseudoaneurysmal lesions that account for 0.5–2% of ruptured intracranial aneurysms and result in abnormally high morbidity and mortality [1,4,7]. They represent approximately 0.9–6.5% of all internal carotid artery (ICA) aneurysms [2,7]. Unlike saccular or 'berry' aneurysms, which usually occur at branch points, BBAs are small and characterized by a broad-based bulge at non-branching sites on arteries [8]. Although they can occur almost anywhere, they most commonly arise from the superior or superomedial wall of the supraclinoid ICA [5,7,8]. The first symptom of almost all patients with BBAs is a severe headache; they are more frequently seen at a younger age [8], in females [9] and on the right side of the ICA [7] than the typical saccular ICA aneurysms. The BBAs lack the internal elastic lamina and media layers and are only covered with a thin fibrinous tissue and adventitia. They therefore frequently present with a subarachnoid hemorrhage (SAH) and are very prone to re-rupture [6, 7]. The combination of small size and SAH at presentation means they are easily missed on CT angiography and digital subtraction angiography (DSA) with 3D shaded surface remains the most accurate diagnostic method of BBAs [24], even though it is sometimes necessary to repeat it before being able to detect them [11].

BBAs are characterized by a dome shape and a poorly defined broad-based neck that limits the possibility of coiling [10, 12]. Different treatment options have been employed. Endovascular therapies with flow-diverting stents or stent-assisted coil embolization offer a lower risk of intraprocedural rupture complications, but have low aneurysm obliteration rates, especially in the acute phase [10, 13]. Microsurgery with direct clipping carries a high risk of rupture and subsequent ICA sacrifice [7, 10, 14], but offers superior aneurysm obliteration rates. However, regardless of the method used, Meling et al. demonstrated that ICA sacrifice in the acute stage of SAH inadvertently results in poor outcomes in patients not treated with combined revascularization surgery [7].

An alternative to direct microsurgical clipping is the clip-wrapping technique, wherein after exposing the ipsilateral C1 segment for proximal control, the

diseased ICA segment is dissected and the BBA is wrapped using various materials. It has the benefit of preserving the anterograde blood flow of the ICA as well as reinforcing the weakened wall of the aneurysm [3].

The aim of this single-center, retrospective study was to evaluate patients with ruptured BBAs of the ICA trunk treated with microsurgical Gore-Tex clip-wrapping and ICA preservation.

PATIENTS & METHODS

We performed a retrospective study of six subjects with ruptured ICA BBAs treated with microsurgical Gore-Tex clip-wrapping and ICA preservation by the senior author between January 2011 and June 2016.

The data analyzed was patient age, sex, time from symptoms to surgery, initial GCS, pre-op Hunt and Hess grade (H&H), modified Fisher grade, aneurysm size and location, pre-op angiography, modified Rankin scale (mRS) at follow-up, complications and follow-up length.

The patients were examined clinically and radiologically using cerebral computed tomography (CT) and CT angiography (CTA) shortly after arrival. All but one patient also underwent digital subtraction angiography (DSA).

Based on the preoperative CT findings, the patients were scored according to the Fisher scale [15], as well as the modified Fisher scale [16]. All individuals except one had SAH <1 mm (Fisher grade 2). Using the modified Fisher scale, five patients had thin or diffuse SAH as well as ICH and/or IVH (Modified Fisher grade 2).

The patients underwent routine postoperative clinical and neuro-radiological follow-up examinations. Outcome was assessed according to the modified Rankin Scale (mRS) and the patency of the parent arteries.

RESULTS

The clinical and radiological characteristics of the six patients with ICA BBAs treated with Gore-Tex clip-wrapping are listed in **Table 1**. The median age was 50.0 years (range 31.7 – 54.0 years). A female predominance was observed (2:1). The H&H grade pre-operatively was grade 1 or 2 in five patients and grade 4 in one patient.

All aneurysms were small (<0.3 cm) and unrelated to vessel bifurcations. The aneurysms were located anteromedially, one anterolaterally and three superomedially on the ICA trunk. BBA on the right side of the ICA was most frequent (66%). All patients underwent surgery in the acute stage.

The median time from onset of symptoms to surgery was of 1.0 day (range 0 – 14 days). Both CTA and DSA were negative for one patient and needed to be repeated every 4 days until day 14 before detection of the BBA. No signs of arteriosclerosis, vasospasm or dissection were present pre-operatively for any of the patients.

Aneurysm treatment

All patients underwent a standard frontotemporal craniotomy followed by sylvian fissure exposure. During the procedure, the ipsilateral cervical segment of the ICA is exposed in order to have direct proximal control of the intracranial ICA. Once the ICA BBA is exposed, a thin sheath of Gore-Tex (0.4 mm) is wrapped around the involved segment with the use of microforceps. An aneurysm clip is then applied to supplement the Gore-Tex wrapping in order to occlude and reinforce the fragile wall of the BBA with parent vessel preservation and without compromising the distal arterial blood flow. This particular material also provides relatively uniform distribution of forces around the aneurysm.

Outcome

The outcomes of the six patients are summarized in **Table 1**.

All aneurysms were clip-wrapped without intraoperative ruptures and all parent arteries were preserved in the acute phase. There were no re-ruptures, delayed infarctions or deaths postoperatively or during the course of the follow-up.

One patient had a small anterior choroidal artery (AChoA) infarction with a hemianopia and a hemiparesia that later improved. In one patient, the aneurysm continued to grow at 6 days following surgery and underwent a placement of a flow-diverter and coils. In our first patient, we opted for endovascular ICA sacrifice in a cold phase after a balloon test occlusion. In all other patients, the ICA was preserved and remained patent at follow-up.

After a median follow-up of 57 months (range 20-90), outcome was favorable (mRS score 0-2) in all six patients without radiographic signs of recurrences.

DISCUSSION

In this single-center retrospective study, we evaluated our experience with the use of the Gore-Tex clip-wrapping technique in six patients with BBAs of the ICA trunk. The poorly defined broad-based neck and the fragile wall makes the management of ICA BBAs challenging and no specific therapeutic option has shown an overall superiority compared to others for their treatment. Many different techniques, be it endovascular or surgical, have been employed. However, they all have advantages and drawbacks in the treatment of this high morbidity pathology.

Endovascular therapy

Endovascular therapy provides functional outcomes similar to surgery and provides a better safety profile in terms of perioperative and postoperative complications [6]. However, it still carries important re-repture and regrowth rates, which lead to repeat treatments [39], other risks such as converting to another treatment modality and incomplete aneurysm obliteration that need to be considered. These factors lead to a greater financial burden due to initial device cost but also retreatments and the need for regular angiographic controls [10].

The lack of a defined saccular component, as well as the small size characterizing most of BBAs, makes the effectiveness of coil embolization controversial in the management of ICA BBA [18]. The morbidity and mortality are also increased by the high risk of perforation and re-hemorrhage during the placement of the coils into the saccular component of ICA BBA [19]. Stent-assisted coiling (SAC) is associated with low occlusion rates both initially (range 20-46%) and at mid- to long-term follow-up (range 56-83%) [20]. It also carries a high retreatment rate of 27% (range 18-36%) [20]. Flow diversion devices (FDDs) have a very low initial occlusion rate (36%), but potential for higher mid- to long-term (91%) results than other techniques. However, FDDs have high rates of procedural complications (17%), early rebleedings (7%), morbidity (13%), and mortality (9%) [20, 21]. Recently, a new alternative may remedy these limitations as it has been shown that using two or more stents may greatly reduce the recurrence rate [39]. One of the major limitations of FDDs and endovascular therapy is the need for dual antiplatelet therapy in the acute phase of ruptured aneurysms.

Microsurgery

Direct clipping, surgical trapping (with or without high-flow bypass) and clip-wrapping with various materials are the main microsurgical options for ICA BBAs management. Shah et al. [17] showed in their recent meta-analysis that microsurgery provided higher total aneurysm occlusion rates than endovascular treatment both immediately (mean 88.9% vs. mean 63.9%; $p < 0.0019$) and at last follow-up (mean 88.4% vs. 75.9%; $p < 0.0001$). However, microsurgery does not seem as safe as endovascular approaches when complications following the procedure are compared (28.8% vs 3.2%, p = not significant) [17].

Direct clipping carries a risk as high as 30-50% of intraoperative rupture [25, 26, 27]. Successful direct clipping without intraoperative ruptures has been reported, but this strategy is not considered as a first-line treatment due to the overall high risk of complications [28, 29]. The only exception could be in ICA BBA of Bojanowski type I, which are characterized by only part of the wall being affected and without a visible sac on the DSA [30].

ICA sacrifice in the absence of cerebral revascularization such as in the acute stages of SAH, with raised intracranial pressure, hypercoagulopathy, and risk of vasospasm in collateral vessels like PCOM and ACOM has given extremely poor outcomes [7]. Even with the accurate evaluation of cortical cerebral blood flow demand by preoperative testing, such as the balloon occlusion test, ischemic complications can occur secondary to hemodynamic hypoperfusion or occlusion of the perforating artery, as well as delayed angiographic vasospasm [41]. Some authors have reported that trapping of ICA combined with high-flow extracranial-intracranial bypass (EC-IC bypass) could be one of the most definite treatments for ruptured BBAs [22, 23]. It is therefore important to perform scheduled EC-IC bypass and tolerance for ischemia before trapping of the ICA in patients with a ruptured BBA in the acute stage of SAH [40].

Gore-Tex Clip-wrapping

Before the introduction of Gore-Tex as an alternative technique for clip-wrapping, the use of cotton to aid clipping ("cotton-clipping" technique) was commonly used to manage intraoperative aneurysm neck rupture [31]. Lee et al. [32] reported their experience of using clip-wrapping with cellulose fabric (Bemsheet®) in 18 patients with ruptured ICA BBAs. However, the short follow-up period of 17 months was limited and didn't allow a full investigation of the potential common risks of rebleeding and regrowth.

Gore-Tex offers interesting intrinsic properties that make it an ideal wrapping material for aneurysms. By being an inert, biocompatible component, it does not cause tissue reaction or granuloma formation, and therefore allows the reinforcement of the arterial wall by encircling it without compromising the distal arterial blood flow. Even though it is made of a thin sheath, its biomechanical properties prevent any further dilation of the aneurysm, while at the same time uniformly distributing the capillary hydrostatic forces [42].

The combination of the clipping to secure the various materials (muscle fascia, cotton, cellulose fabric, or Gore-Tex) and wrapping around the entire circumference of the ICA when facing fragile BBAs has been observed to effectively reinforcing the vessel wall at the location of the aneurysm [33, 34]. However, subsequent rebleeding or regrowth may present as complications due to the difficulty in identifying the borders or a slight gap between the BBA's wall and the wrapping material. The ideal treatment would be a fine-tuned clipping to create just enough adherences between the wrapping material and the BBA [43].

In 2016, Safavi-Abbasi et al. [35] presented 30 patients (8 SAH cases) treated with the Gore-Tex clip-wrapping technique. In this BNI series, aneurysms recurred in only 2 of 30 patients after a mean follow-up of 3.5 years. However, as only 3 aneurysms were defined as BBAs, the rest being fusiform aneurysm. Thus, this study cannot be used to evaluate the efficacy of this technique for ICA BBAs.

Effect of Gore-Tex on Recurrences and Rebleeding rates

Johnston et al. [36] showed that the degree of aneurysm occlusion after the initial treatment was a strong predictor of the risk of subsequent rupture in patients presenting with SAH. The risk of rupture in aneurysms that were <70% occluded was 24.5% in the first year which was similar to re-rupture rates of untreated ruptured aneurysms [37]. Therefore justifying attempts to completely occlude aneurysms.

In our current study, among the six patients treated with Gore-Tex for their ICA BBAs, none presented with episodes of recurrences or re-bleeding of their BBA during the median follow-up of 50 months (range 20-77). So far all patients have shown favorable clinical outcomes, what remains to be determined is the long-term durability of Gore-Tex clip-wrapping. Our follow-up demonstrated satisfying results for the first few years, but has to be continued to be able to fully affirm the importance of Gore-Tex clip-wrapping in ICA BBAs treatment.

Combined options

In our series, we also employed combined microsurgical and endovascular techniques in 2 cases. It consisted of Gore-Tex clip-wrapping in the acute phase followed by delayed SAC or endovascular ICA sacrifice for definitive treatment. This combined approach of acute surgical stabilization followed by definitive endovascular reconstruction may reduce hemorrhagic complications while improving long-term treatment durability [38].

CONCLUSIONS

Blood-blister aneurysms remain a therapeutic challenge with potential high rate of morbidity and mortality. Different therapeutic options are available but there has not been showed a superiority of one over the other yet.

Microsurgery gives the highest obliteration rates both immediately and at long-term follow-up. However, it comes at the price of higher risk rate of intraoperative rupture with surgical exploration of ICA BBAs in the aim of a direct repair [6, 17]. Despite lower obliteration rates, endovascular therapy with its different types of technique offers a lower risk of intra and post-operative complications. The safety of this technique has made it the most often used technique in the treatment of BBAs.

In our clinical practice, we have adopted the clip-enforced Gore-Tex wrapping as our first-line surgical modality in treatment of ICA BBA s. It has shown promising results with complete obliteration rates in all our patients, paired with a low risk of complications. The specific properties of the Gore-Tex also yielded positive long-results.

LIST OF LEGENDS

Table 1: Patient and Radiological characteristics and Clinical Outcomes

COMPLIANCE WITH ETHICAL STANDARDS

Conflict of interests

The authors declare that they have no conflict of interest.

Ethical approved

Not applicable as no new patients were involved in this research.

Informed consent

Not applicable as no new patients were involved in this research.

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Table 1

Pt. No.	Age (years)/ Sex	Time from symptoms to surgery (days)	Initial GCS score	Pre-op H&H grade	BBA size (mm)	Modified Fischer scale	Pre-op angiography	ICA side & localisation	Post-op mRS	Follow-up (months)	Complications
1	31.7/F	1	14	1	3	2	Yes	Left/ Anterolateral	2	69.9	
2	51.1/F	1	15	2	3	2	Yes	Right/ Superomedial	1	90.1	
3	33.7/F	14	7	4	3	2	Yes	Right/ Superomedial	2	87.2	
4	54.0/M	0	15	2	2	2	Yes	Right/ Superomedial	2	44.9	Small AChoA infarction
5	53.8/F	0	15	1	2	0	No	Left/ Anteromedial	1	20.1	
6	49.0/M	1	13	2	3	2	Yes	Right/ Anteromedial	1	38.5	Continued aneurysm growth

Pt. No. = patient number; GCS = Glasgow Coma Scale; H&H = Hunt and Hess scale; ICA = Internal Carotid Artery