Efficacy of Clip-Wrapping in Treatment of Complex Pediatric Aneurysms

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Abstract

Purpose: Pediatric aneurysms (PAs) are distinct from their adult counterparts with respect to typical location, aneurysm type, and known predisposing risk factors. Many strategies have been employed to treat PAs, but, although it has been used frequently in adults, clip-wrapping in pediatric patients has only been reported once. We present a series of pediatric patients that underwent clip-wrapping and discuss this strategy as an effective means of treating unclippable PAs.

Methods: Pediatric patients with clip-wrapped aneurysms over a 5-year period were retrospectively identified. Clinical presentation, surgical management, and clinical and radiological outcome of the patients were evaluated.

Results: Five pediatric patients with aneurysms were treated with clip-wrapping during the specified period. Three had traumatic pseudoaneurysms, with two subarachnoid hemorrhages from aneurysm rupture. One patient presented with mycotic pseudoaneurysm rupture causing a large intraparenchymal and subarachnoid hemorrhage. Another patient had a dissecting complex saccular lenticulostriate aneurysm with 4 perforating vessels arising from the dome. Four patients had good clinical results, with Glasgow Outcome Scale (GOS) scores of 5 after at least 1-year follow-up (mean 24.2); one patient had a GOS score of 5 at discharge but no additional follow-up. Post-operative neuroimaging demonstrated vessel patency after clip-wrapping with no recurrent hemorrhages or increase in aneurysm size; however, one had progressive occlusion of the

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artery in a delayed fashion and had a small clinical ischemic event from which she fully recovered.

Conclusions: Clip-wrapping appears to be an effective underutilized technique for treatment of pediatric complex aneurysms that cannot be treated with conventional methods.

Keywords: Complex aneurysms; pediatric; clip-wrapping

Running Title: Clip-wrapping pediatric aneurysms

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Introduction

Pediatric aneurysms (PAs) comprise between 0.7 and 5% of all reported aneurysms [5-8, 11-14]. They are distinct from their adult counterparts in multiple ways. For example, a male predominance (as large as 11:1) is typically reported in the pediatric population, but is not seen in the adult population [8]. In addition, there are three times as many posterior circulation aneurysms and a preponderance of carotid terminus aneurysms in children [8, 11, 14]. The presentation of aneurysms in children also varies dramatically from that in adults. Whereas adults generally have poor Hunt-Hess scores on admission, the Hunt-Hess grade in the vast majority of pediatric patients with aneurysmal subarachnoid hemorrhage (SAH) is 1–3, while poor Hunt-Hess scores of 4 or 5 usually occur in as few as 15% of patients (although the highest reported incidence was high at 42%) [7, 9, 11, 14]. Approximately one third of patients with PAs have comorbidities such as trauma, cerebral ischemia, and hematologic, immunologic, vasculitic, and rheumatological disorders, whereas comorbidities are not common in adults [5].

Since the first report on PAs in 1939, there have been well over 1000 aneurysms reported in over 30 different series and approximately 100 case reports [5, 7, 8, 13]. A myriad of strategies have been employed to treat PAs, including standard clip ligation, endovascular methods, trapping with and without bypass, bypass alone, wrapping, and proximal occlusion [5]. Although good outcomes are reported in >90% of patients in large series where standard clip ligation is usually employed, patients with unclippable PAs comprise a group that warrant extra consideration [8, 11, 13]. These unclippable PAs are “difficult or impossible to repair primarily” [5] given that there is often circumferential wall weakening or involvement of the parent vessel with the abnormal

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neck. Endovascular treatment has been performed much less than microsurgery in PA management, and the series describing patients who underwent endovascular coiling are small [7, 9, 14]. Given the paucity of long-term follow-up with endovascular treatments in PA patients, clip-wrapping is an important tool already shown to be effective in adults in treating fusiform aneurysms, pseudoaneurysms, and other aneurysms that cannot be treated with conventional methods because of the location of perforating vessels or the shape of the aneurysm [3, 4]. Clip-wrapping has been shown to be efficacious in protecting against long-term growth or hemorrhage [3].

In our review of the more than 1100 cases of PA in the literature, only a handful of patients in one study underwent clip-wrapping as a treatment for their unclippable PA [5]. We describe five pediatric patients that underwent clip-wrapping for treatment of their aneurysms and examine their outcomes. We also discuss clip-wrapping as an effective means of treating unclippable PAs that may not be amenable to endovascular treatments.

Methods

The University of Utah maintains a comprehensive database of consecutive adult and pediatric neurosurgical cases. The database was queried to generate our patient sample. Patients younger than 16 years of age who had cerebral aneurysms treated at the University of Utah or Primary Children Medical Center from January 1, 1999, to December 31, 2011, were included.

A retrospective review of each patient who was treated with clip-wrapping was conducted to identify demographics, clinical outcome, and follow-up data. Radiological

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data were acquired from direct review of the relevant imaging. All chart reviews and data acquisition were in compliance with the regulations determined by the University of Utah Institutional Review Board and with the Health Insurance Portability and Accountability Act (HIPAA) of 1996.

Results

We identified 13 pediatric patients who had surgical treatment for cerebral aneurysms. Three patients were treated with standard clipping, two with clipping and vessel reconstruction, one with clipping and bypass, one with clipping and vessel trapping, and one with aneurysm excision and vessel sacrifice. No patients were treated with endovascular techniques. Five patients were treated with clip-wrapping of their fusiform aneurysms or pseudoaneurysms during the specified period (Table 1). All five clip-wrapping cases were treated by the senior author (WTC).

Of the five patients treated with clip-wrapping, three had traumatic pseudoaneurysms, with two subarachnoid hemorrhages from aneurysm rupture. One patient presented with a large intraparenchymal hemorrhage (IPH) with extensive SAH from mycotic pseudoaneurysm rupture. The fifth patient suffered from headaches and presented with a dissecting pseudoaneurysm with 4 perforating vessels arising from the dome [1]. All of the patients had successful clip-wrapping and good clinical results, although patient 5 had a basal ganglia stroke after clip-wrapping. Each had a Glasgow Outcome Scale (GOS) score of 5 at discharge except for the patient who suffered the stroke, who had a GOS score of 4 at discharge. One patient moved out of state after discharge and was lost to follow-up, but the others all had GOS scores of 5 at 1-year

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follow-up. Postoperative neuroimaging demonstrated vessel patency after clip-wrapping with no recurrent hemorrhages or increase in aneurysm size during follow-up except for one patient that had delayed middle cerebral artery occlusion at 18 months after surgery. Fortunately, she had developed extensive collateral arterial supply and recovered fully from her ischemic event. Average follow-up, with the exception of the patient that moved, was 30.25 months, with CT angiography demonstrating no recurrences.

Illustrative Cases

Case 1

This 3-year-old boy with spina bifida and shunted hydrocephalus fell down the stairs and had a shunt fracture along with some traumatic SAH (Figure 1A). He underwent shunt revision and was discharged home at his neurologic baseline. One month later, he presented with a four-day history of nausea and vomiting and was found to have interhemispheric SAH and a right frontal IPH (Figure 1B,C). CT angiograms demonstrated a 6.5 × 5.5-mm callosomarginal artery aneurysm, which appeared to be somewhat saccular (Figure 1D,E).

The patient underwent a bifrontal craniotomy for interhemispheric approach for clip-wrapping of his ruptured callosomarginal aneurysm. Intraoperatively, the aneurysm was noted to have no focal neck and was actually a dissecting pseudoaneurysm, with the neck incorporating a 1- to 2-cm segment of the arterial wall. A clip was placed on the top of the dilated vessel dome, the affected segment was wrapped, and a permanent clip was placed to hold the muslin wrap. The patient did well after surgery and was discharged

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home at his neurologic baseline. Postoperative imaging showed stable aneurysm size (Figure 1F).

Case 2

An 11-year-old boy with a 3-week history of a strep throat infection presented with a large left frontoparietal IPH with extensive SAH in the basal cisterns (Figure 2A,B) after being found unconscious. His initial Glasgow Coma Scale score was 9, and he was hemiparetic on his right side. The patient's CT angiogram and cerebral angiograms revealed two small left middle cerebral artery (M2) fusiform mycotic pseudoaneurysms (Figure 2C). The patient required decompressive craniectomy with external ventricular drain placement because of decreasing mental status. Subsequent serial cerebral angiograms demonstrated the mycotic pseudoaneurysms were enlarging despite treatment with appropriate broad-spectrum antibiotics at day 12 of admission (Figure 2C–E).

The patient was taken back to the operating room for definitive treatment of these enlarging mycotic aneurysms. Intraoperatively, two fusiform pseudoaneurysms were seen located 1 cm from each other on the left superior M2 branch (Figure 2E). As there were perforating vessels arising from the vessel and given the patient's dominant hemisphere, sacrifice of the artery through trapping and distal revascularization with bypass was considered but not chosen to avoid risk of neurological deficit from sacrifice of the vessel between the aneurysms and its perforating vessels. These aneurysms were individually clip-wrapped with muslin as noted in Figures 2E and 3. The patient was discharged for

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rehabilitative care. At 18-month follow-up, he had no motor or speech deficit, and his aneurysms were almost completely gone (Figure 2F).

Discussion

Pediatric Intracranial Aneurysms

Aneurysms in children differ from their adult counterparts with respect to their typical location, aneurysm type, patient demographics, known predisposing risk factors, Hunt-Hess grade after rupture, and clinical outcome [5-8, 11-15]. Perhaps most concerning about the presentation of PAs is their propensity for hemorrhage, rehemorrhage, and recurrence. Re-hemorrhage rates of treated PAs have been reported to be >50% in some series as they are more likely to re-rupture than aneurysms in adults [5]. Another difference in PAs is highlighted by one small series of ruptured PAs, in which 7/12 aneurysms were <7 mm in size and located in the anterior circulation, whereas, in adults, aneurysms of this size and location have a 0% rupture rate at 5-year follow-up [14, 17]. According to two recent large studies [5], treated PAs have an extremely high recurrence rate around 8.5% at an average of 4.2 years after presentation whereas the adult recurrence rate is 1.5%. It is concerning that recurrence rates in PAs treated endovascularly have been reported to be as high as 20–40% in some series [9, 13]. Although some recent series by have supported employing endovascular management techniques for PAs, other studies have found microsurgery more effective because of lower recurrence rates, with long-term average follow-up as high as 25 years [7, 9, 13]. Studies showing long-term follow-up after endovascular treatment are needed since the life expectancy of pediatric patients with cerebral aneurysms is often greater than 50

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years [13]. The largest series of PAs, covering 73 years and 114 pediatric patients with 135 aneurysms treated almost exclusively by microsurgery, was recently published with an average follow-up of 25 years; the results of this study supported microsurgery as an effective treatment with good long-term results [9]. Some of the reports that show similar recurrence rates between microsurgical and endovascular treatments lack long-term follow-up, which is especially important given the average recurrence occurs at around 4 years [13]. Fortunately, multiple studies have demonstrated that good outcomes are achieved >90% of the time for all PAs, and this is much higher than has been demonstrated in adult aneurysms, which usually have lower GOS scores [8, 11, 13]. Furthermore, giant aneurysms and fusiform aneurysms are very common in children but are less common in adults [5]. Fusiform and dissecting aneurysms comprised 60% of PAs but make up just 0.1% of adult aneurysms [5].

Interestingly, fusiform PAs have higher rates of recurrence than other PAs, and other studies have shown that to be true even with direct clip ligation [13]. Fusiform aneurysms have been a long-standing challenge for vascular neurosurgeons. Despite the outcomes achieved with the use of clip-wrapping in unclippable adult aneurysms, clip-wrapping has not been discussed in the PA literature and its use has only been reported in a handful of patients in one PA study [13]. A good example of clip-wrapping being underutilized is a recent report of 4 complex fusiform middle cerebral artery PAs that were treated with vessel ligation or trapping with extracranial–intracranial bypass [13]. Furthermore, recent evidence shows that parent artery occlusion, sometimes used with aneurysms that are too complex for standard clip ligation, predisposes patients to a higher risk of recurrence of their aneurysm, highlighting why clip-wrapping can be an important

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part of the treatment armamentarium for PAs [13]. Another example of relative underutilization of clip-wrapping is the aforementioned largest study of PAs just published, which included 7 different surgical and 1 endovascular techniques while clip-wrapping was never used for any of the 135 treated aneurysms [9]. There are instances in which clip-wrapping may be a better option than vessel sacrifice and revascularization; one example is shown Case 2, in which clip wrapping enabled preservation of the intervening perforating vessels between the mycotic aneurysms on the same vessel. Given our experience with clip-wrapping PAs, we suggest this may be a vital and effective alternative in treating these complex aneurysms.

Clip-Wrapping Technique

Our five patients had pseudoaneurysms/dissecting aneurysms treated with clip-wrapping as illustrated in Figure 3. Four of the patients had GOS scores of 5 at discharge. In the four patients available for follow-up, all were GOS 5 (at one year); another patient returned to an out-of-state home at discharge. Complications included a slowly accumulating extra-axial hematoma that had to be evacuated two days after initial surgery in a two-month-old infant (patient 4) and a basal ganglia stroke that occurred after clip-wrapping of the dissecting pseudoaneurysm in patient 5 that incorporated perforating vessels. Intraoperative neuromonitoring in that case showed a brief decrease and loss in motor evoked potentials for 20 minutes, which returned to baseline intraoperatively with retrograde flow in the two larger perforating vessels. Postoperatively, the patient had a basal ganglia stroke and was GOS 4 at discharge, but

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GOS 5 at follow-up despite having a delayed middle cerebral artery occlusion 18 months after surgery with an ischemic event from which she recovered fully.

This clip-wrap technique provides the advantage of wrapping of the vessel to induce fibrosis and strengthen the wall over time, but also offers immediate protection from rebleeding by reinforcing the rupture site with a snug wrap (with muslin or woven synthetic fabric) that is secured by an aneurysm clip. Indeed, this technique has been used over an extended period by the senior author (WTC) for unclippable aneurysms in adults for native vessel repair and avoidance of parent vessel sacrifice with or without bypass. This experience has been described previously in application for treating pseudoaneurysms in adult patients by the author [2] and others [4], and the senior author has noted exceptional control and no rehemorrhage (unpublished results). This is in contrast to merely wrapping with muslin or woven synthetic patch, which may not provide immediate protection from rerupture. One study of 10-year follow-up in adult patients with wrapped aneurysms showed early and late rebleeding rates of 8.5 and 1.5% [16].

Although this series doubles the number of previously reported cases of clip-wrapping, it is a small sample. Nevertheless, it highlights that this method of treatment could be used successfully more often. In contrast to adult aneurysms, PAs tend to recur at a much higher rate and do so several years after presentation [13]. Furthermore, extremely rare complications may occur in a delayed fashion, such as muslin-induced vasculopathic stenosis [10] and delayed vessel occlusion as occurred in one of our patients [1]. This demonstrates the vital need for long-term follow-up with imaging in these, and all PA, patients.

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Conclusion

Many treatment strategies have been described to treat PAs. Although clip-wrapping of adult aneurysms has been well-described, use of this technique has almost never been reported with pediatric patients. Our 5 pediatric patients had good outcomes when their fusiform aneurysms/pseudoaneurysms were treated with clip-wrapping. Clip-wrapping can be employed as a treatment method in patients with complex aneurysms that cannot be treated with conventional methods. Clip-wrapping is a tool for treatment of PAs in which sacrifice of the parent vessel is to be avoided. The experience noted here demonstrates good long-term control with minimal complications, indicating that clip-wrapping should be added to the armamentarium for the management of complex and fusiform pediatric intracranial aneurysms.

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Figure 1. (A) Non-contrast sagittal head CT study demonstrating interhemispheric SAH sustained after the patient fell down a flight of stairs. (B,C) Non-contrast axial head CT scans one month later showing a right frontal intraparenchymal hemorrhage (B) and interhemispheric SAH (C). Axial (D) and sagittal (E) CT angiograms demonstrating callosomarginal aneurysm. (F) Postoperative angiogram showing good vessel patency and stable aneurysm size after clip-wrapping.

Figure 2. (A, B) Non-contrast axial head CTs demonstrating a large left frontoparietal IPH with extensive SAH in the basal cisterns. (C) Cerebral angiogram demonstrating one small left M2 pseudoaneurysm (arrow). (D) Three-dimensional reconstruction showing the two pseudoaneurysms on the left M2 segment (arrows). (E) Cerebral angiogram obtained 12 days after panels C and D, showing enlargement of the previous left M2 distal pseudoaneurysm and a more proximal L M2 pseudoaneurysm that developed 1 cm away (arrows). Given the two infectious aneurysms on the same artery with obvious intervening perforating vessels in between, a decision was made to not perform a vessel sacrifice and distal revascularization with superficial temporal artery to middle cerebral artery bypass. Instead, the aneurysms were individually clip-wrapped to preserve the parent artery and the important intervening perforating arteries. (F) Cerebral angiogram 18 months after surgery showing the two clip-wrapped pseudoaneurysms have almost completely healed (arrows).

Figure 3. Artist's rendition of clip-wrapping of complex PA. (A) Muslin wrap placed around the weakened portion of the dissected pseudoaneurysm. (B) Muslin wrap placed

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around the entire portion of the involved fusiform aneurysm. (C) Clip-wrap technique applied to vessel with snug wrap (muslin or woven synthetic fabric) secured by an aneurysm clip to strengthen and secure the weakened vessel.

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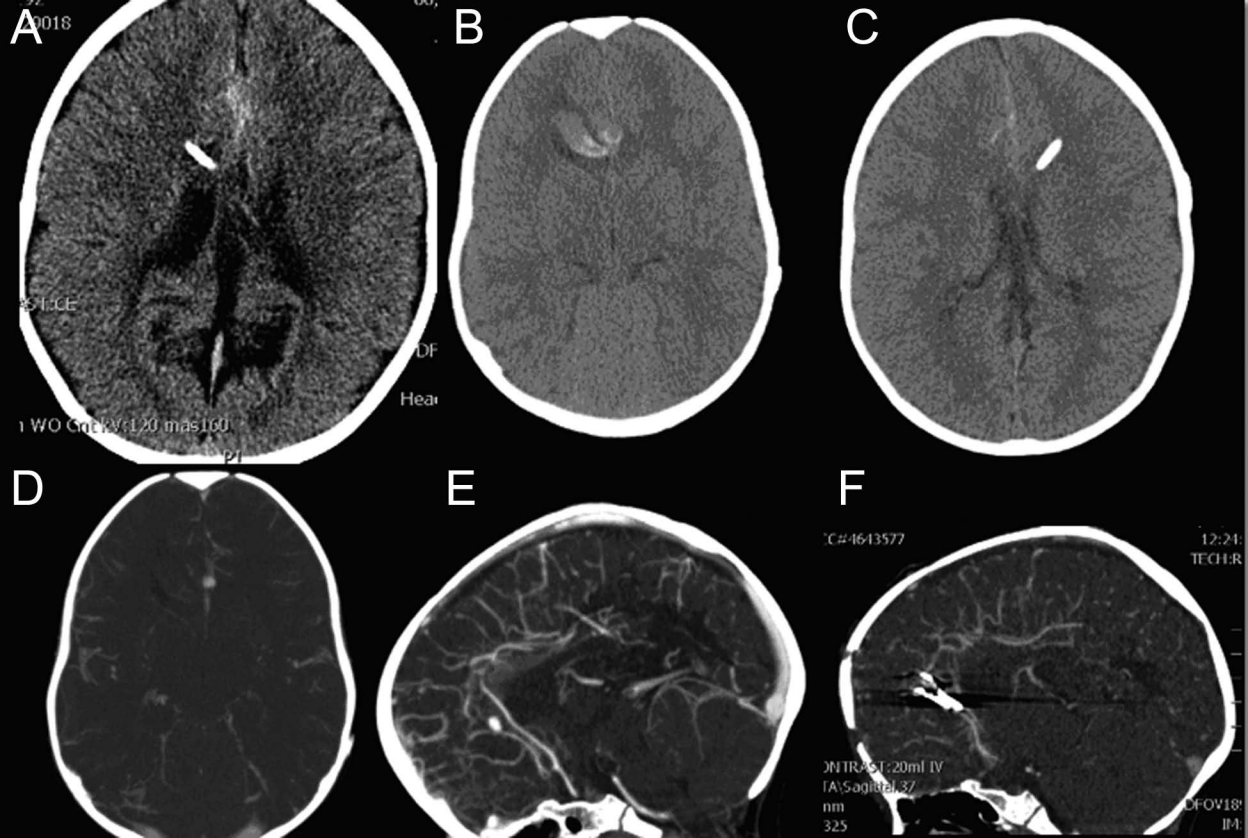
Table 1: Aneurysm characteristics, treatment, and outcome in pediatric patients who underwent clip-wrapping

Case	Age (yr) / Sex	Aneurysm location/ Size (mm)	Aneurysm type	Presentation	GCS score	Treatment(s)	Clinical or angiographic vasospasm?/ Treated?	Compli-cations	Total follow-up (months)	GOS score at discharge/ follow-up
1	3/M	Pericallosal (6.5x5.5)	Fusiform pseudoaneurysm	Rupture; nausea/emesis 1 month after fall down stairs	14	Clip-wrap	Mild/no	None	12	5/5
2	11/ M	Two left M2 (1x1, 1.5x1.5)	Fusiform pseudoaneurysm (mycotic)	Rupture; large IPH and extensive SAH	9	1. Hemi-craniectomy 2. Clip-wrap	Yes/yes	None	18	5/5
3	4/M	Right vertebrobasilar (2.3x1.4)	Dissected blister aneurysm	Follow-up imaging abnormality after blunt trauma 1 year earlier	15	Clip-wrap	No/No	None	55	5/5
4	0.2/ F	Right anterior choroidal	Fusiform pseudoaneurysm	Extensive SAH after MVA	3	Clip-wrap and repair of distal carotid tear	Yes/no	Extra-axial hematoma evacuation	Lost at discharge (patient	5/ n/a

		(2.1x2.0)						postoperative day 2	from out of state)	
5	9/F	Left MCA (8x4)	Dissected pseudoaneurysm	Complex migraine headache	15	Clip-wrap involving 4 perforating vessels from thick part of pseudoaneurysm	No/No	Basal ganglia stroke; Delayed MCA occlusion	42	4/5

M2, M2 segment of middle cerebral artery; MCA, middle cerebral artery; IPH, intraparenchymal hemorrhage; SAH, subarachnoid hemorrhage; MVA, motor vehicle accident; GCS, Glasgow Coma Scale; GOS, Glasgow Outcome Scale; n/a, not applicable and/or not available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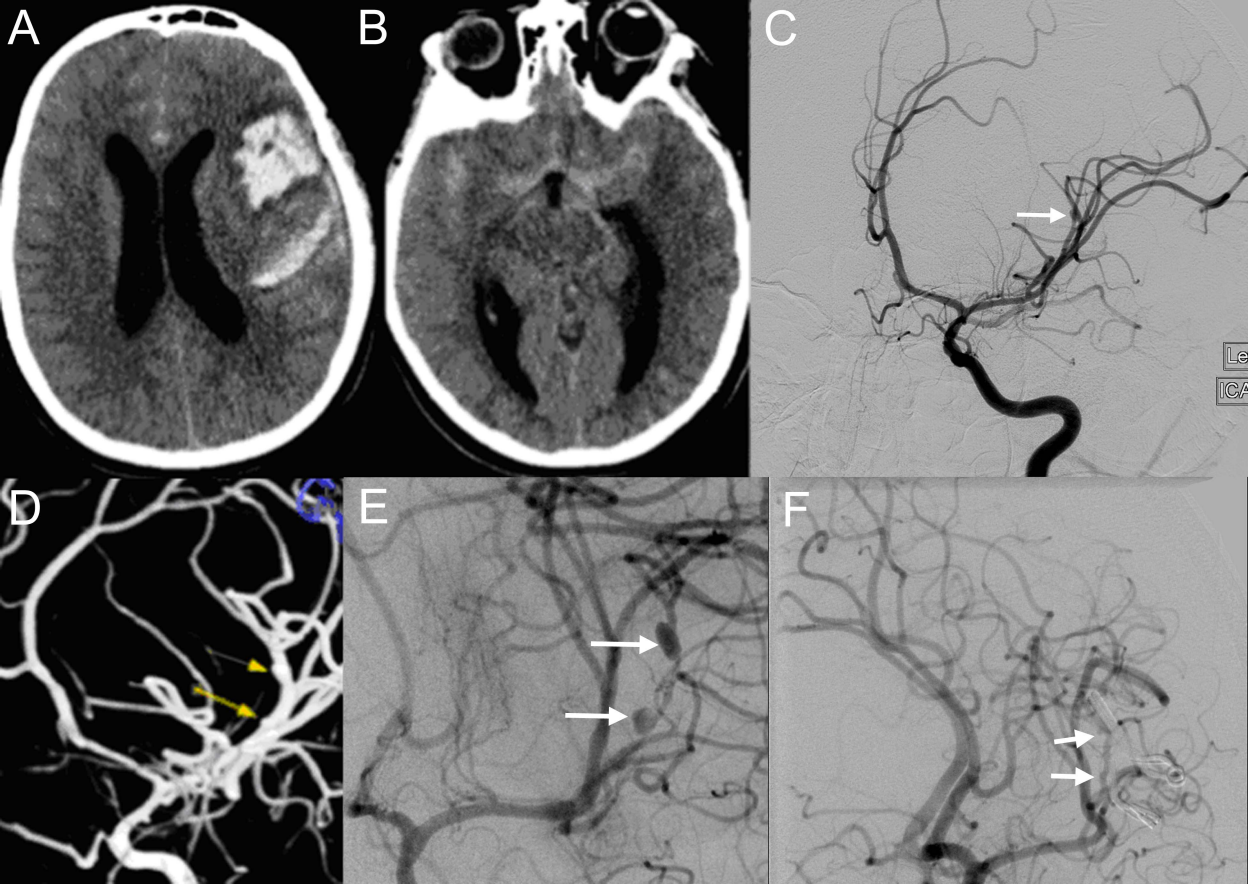
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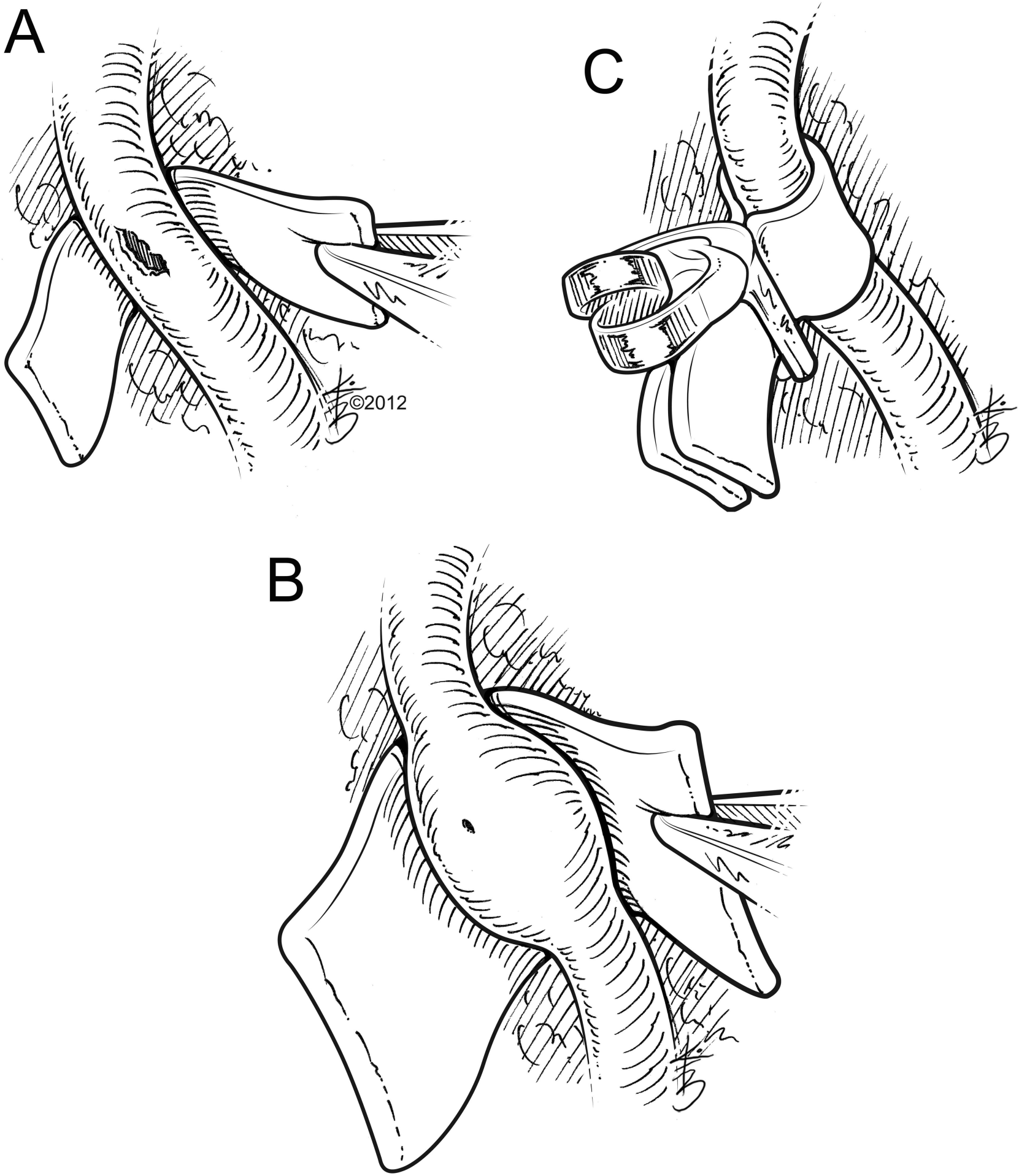
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