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Direct clipping of large persistent trigeminal artery aneurysm under deep hypothermia and circulatory arrest : A case report and review literature

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The aneurysms of persistent trigeminal artery (PTA) is rare, mostly treated by indirect surgical attack. Due to this relative inaccessible surgical approach, only four direct clippings of these aneurysms have been reported to date. The patient presented with painful ophthalmoplegia of the left eye. Cerebral angiography revealed a large aneurysm at the junction of C4 portion of the left internal carotid artery (ICA) and PTA.Due to its hemodynamic complexity with a possible inadequacy of blood supply to the left ICA. The surgical operation was performed the techniques of deep hypothermia, circulatory arrest with cardiopulmonary bypass and barbiturate treatment. In conclusion, PTA aneurysm is a rare and technically challenging lesions. Only symptomatic lesions should be treated. The careful evaluation of their anatomy and hemodynamic state with auxiliary techniques are the key to the success.

Key words: *Cerebral aneurysm, Circulatory arrest, Clipping , Hypothermia, Trigeminal artery.*

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รุ่งศักดิ์ ศิวานุวัฒน์, สุรชัย เคารพธรรม, ภูษิต ทรัพย์สมพล, วิชัย เบญจชลมาศ, ลาวัลย์ ตู้จินดา, อรนุช เกี่ยวข้อง. การผ่าตัดหนีบเส้นโลหิตแดง persistent trigeminal โป่งพอง ภายใต้ภาวะ deep hypothermia และ circulatory arrest : รายงานผู้ป่วย 1 รายและทบทวนวารสาร. จุฬาลงกรณ์เวชสาร 2544 ส.ค; 45(8): 685 - 95

เส้นโลหิตแดง persistent trigeminal โป่งพอง พบได้ยาก และยังรักษายาก เนื่องจากตำแหน่งทางกายวิภาค จากการทบทวนวารสารพบมีเพียง 4 รายเท่านั้นที่ได้รับการรักษาโดยการผ่าตัดหนีบเส้นโลหิตโป่งพองนี้ ได้รายงานผู้ป่วย 1 ราย อาการแสดงทางคลินิก คือ ปวดและไม่สามารถกลอกตาซ้ายได้ การตรวจทางรังสีวิทยาพบเส้นโลหิตโป่งพองขนาดใหญ่บริเวณรอยต่อของเส้นโลหิตแดง internal carotid ตำแหน่ง C4 กับเส้นโลหิตแดง persistent trigeminal ผู้ป่วยได้รับการผ่าตัดหนีบเส้นโลหิตโป่งพองด้วยเทคนิคพิเศษ ภายใต้ภาวะ deep hypothermia, circulatory arrest ร่วมกับ cardiopulmonary bypass และ barbiturate treatment หลังผ่าตัดไม่พบภาวะแทรกซ้อน ผู้รายงานเห็นว่า การศึกษาพยาธิกายวิภาค และการไหลเวียนโลหิต โดยละเอียดร่วมการใช้เทคนิคพิเศษเป็นปัจจัยสำคัญในการรักษาโรคนี้

A persistent trigeminal artery (PTA) is a rare connecting channel between the internal carotid artery and the basilar artery. It is frequently found in association with other intracranial lesions such as aneurysm, arteriovenous malformation, etc. Twenty – nine reported cases of PTA aneurysm have been reported. Thirteen of them had been surgically treated and but in only 4 cases that the aneurysm had been directly clipped. We present a case of large PTA aneurysm that was successfully treated by direct clipping under deep hypothermic-circulatory arrest and barbiturate treatment.

Case Report

A 48-year old right-handed man presented with progressive left periorbital pain and double vision over the past year. Examination showed ptosis and proptosis, and cranial nerve III, IV, VI paresis of the left eye and decreased pinprick sensation along the left trigeminal nerve distribution. A computerized tomography (CT) scan revealed a densely enhanced mass at the left cavernous sinus. (Fig.1 a, b) This lesion was also identified on the magnetic resonance imaging (MRI) scan as a signal void globular mass with a diameter of 20 mm. (Fig. 2 a, b) The digital subtraction cerebral angiography disclosed a large globular aneurysm 20 mm in diameter at the junction between C4 portion of left ICA and PTA. (Fig. 3 a,b) This PTA originating from the junction between C4 and C5 of left ICA, traversed dorsally and medially to join basilar artery segment between superior cerebellar artery (SCA) and anterior inferior cerebellar artery (AICA). The vertebral angiography clearly good posterior circulation, this aneurysm should be and adult-type PTA aneurysm. (Fig. 4) The detailed hemodynamic studies revealed no cross flow from the right ICA when the ICA was occluded. Alcock's test disclosed collateral flow from posterior circulation via PTA to ipsilateral ICA and anterior cerebral artery (ACA) but not to middle cerebral artery (MCA). Due to the progressive nature of the disease, surgical correction was clearly indicated.

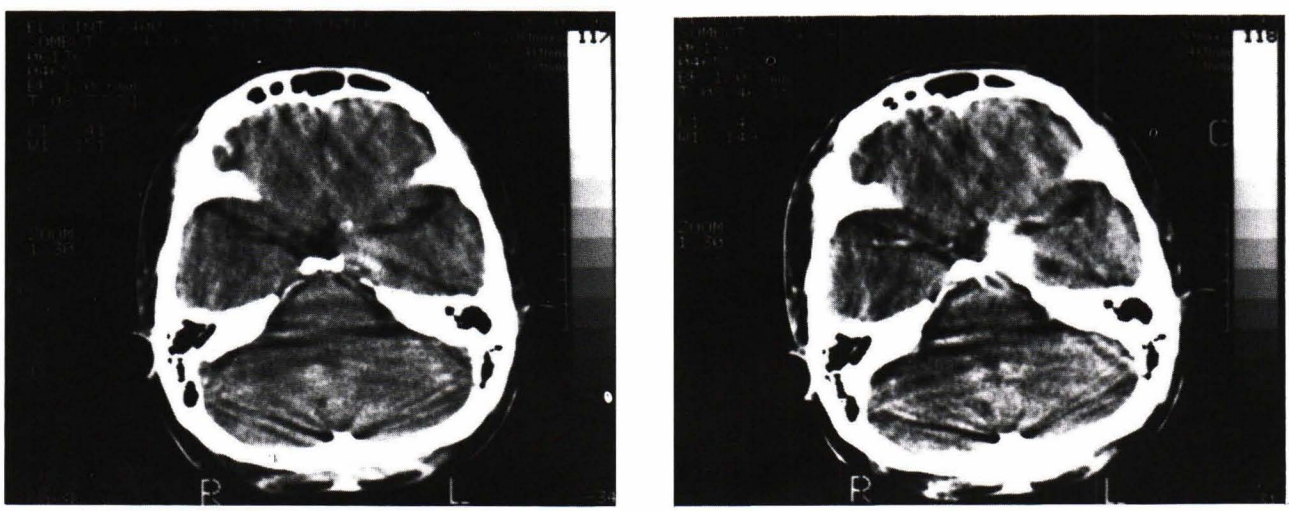

Figure 1. Axial computed tomographic scan (A) before and (B) after contrast, demonstrating a densely enhanced mass at the left cavernous sinus.

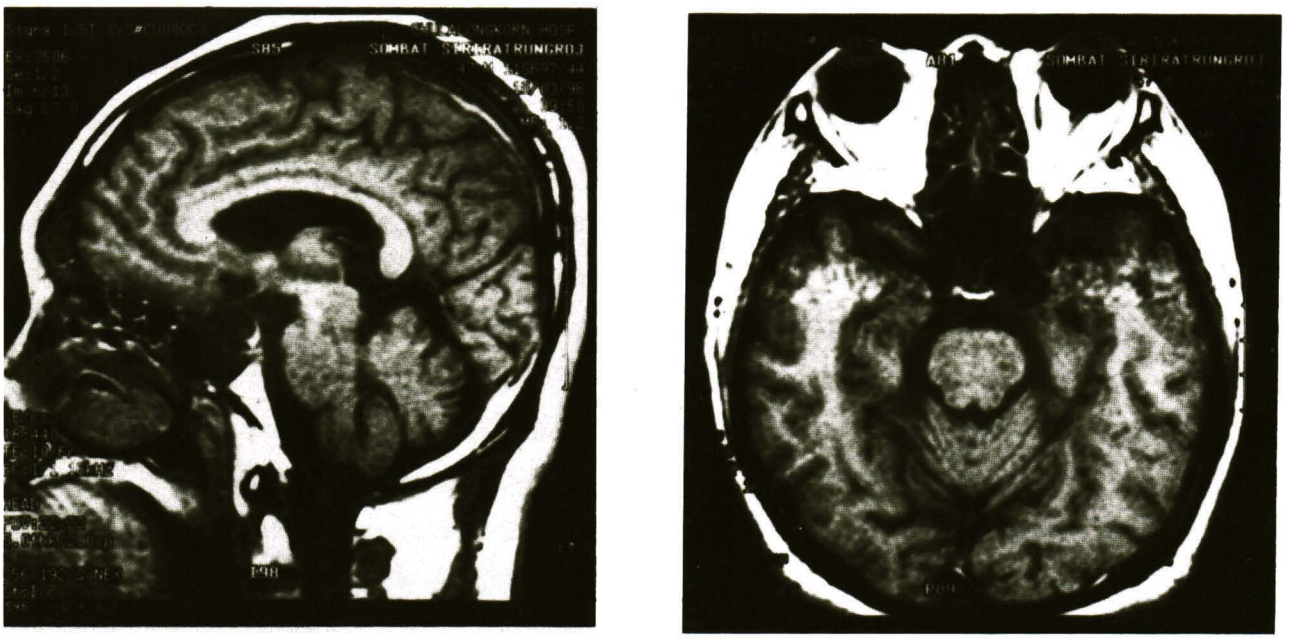

Figure 2. T1- weighted axial (A) and sagittal (B) MRI scans, showing a globular mass at the left cavernous sinus and signal void appearance.

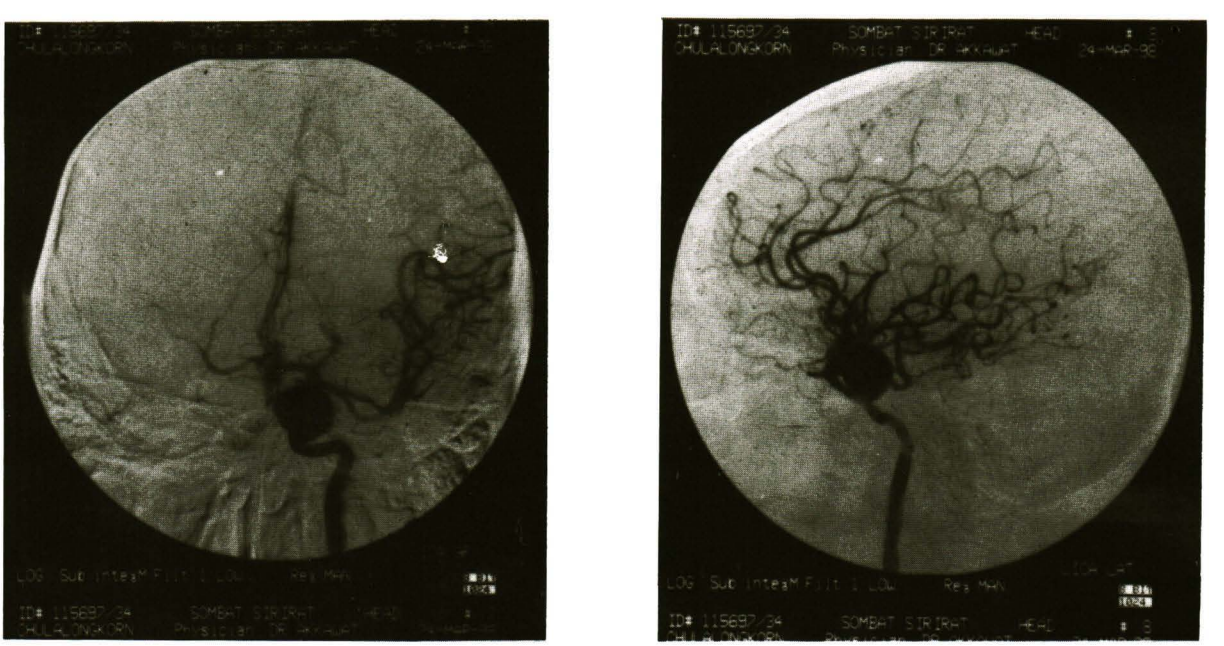

Figure 3. Left internal carotid artery angiogram, anteroposterior view (A), lateral view (B), demonstrating a large intracavernous aneurysm of the internal carotid artery associated with a persistent trigeminal artery. PTA arises between C4 and C5 to join the basilar artery.

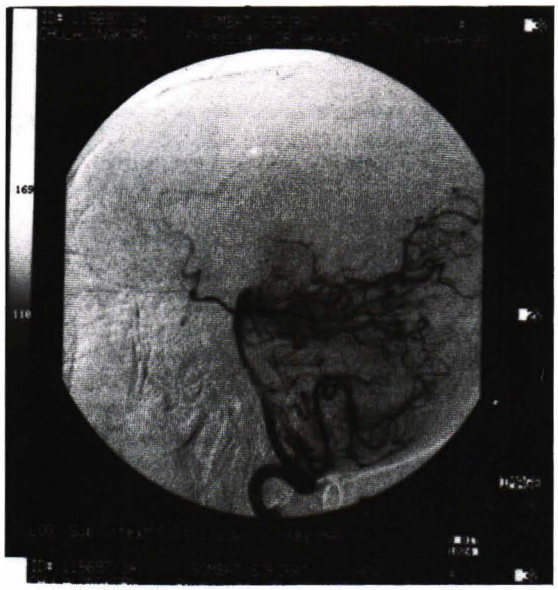

Figure 4. Left vertebral artery angiogram,lateral view, demonstrating the posterior circulation and adult type PTA.

Surgery was performed in 3 occasions.

1st operation. By left pterional craniotomy approach with proximal preparation of the left ICA at the neck. The anterior clinoid process was intradurally drilled off, then the carotid dural ring was opened and the cavernous sinus was entered through the medial triangle.The aneurysm neck was entirely obscured by its fundus and because of an inadequate collateral circulation to the left ICA, The operation was terminated.

2nd operation. Ten days after the 1st operation, the patient underwent surgery under deep hypothermia, circulatory arrest and barbiturate protection technique. [29,30] Using the former approach, the aneurysm was dissected as much as possible with meticulous hemostasis. The right femoral - femoral cardiopulmonary bypass was then begun after full heparinization was achieved. After core temperature of 15 degree was reached by process of cooling, thiopentone was loaded and infused to induce EEG burst suppression. When circulation was arrested, the aneurysm dome appeared softer, thus allowing further dissection possible. There was an inadvertent injury to the left oculomotor nerve during the process of identifying the neck of aneurysm. Attempt to obliterate the neck of aneurysm was limited due to along period of circulatory arrest (61 minutes). Four clips were applied to the aneurysm neck but complete obliteration could not be ascertained. The patient was rewarmed and cardiopulmonary bypass was stopped. He regained consciousness and was extubated at 7 and 38 hours postoperatively. His mentation was intact. He had no any increase in neurological deficits. Postoperative angiography revealed incomplete clipping of the aneurysm on its proximal side. (Fig 5 a,b)

3rd operation. Two additional clips were applied successfully under a mild hypothermia and barbiturate protection. The last angiography demonstrated complete clipping with preservation of PTA. (Fig. 6 a, b) The patient was discharged on the 9th postoperative day. His neurological deficits remained unchanged.

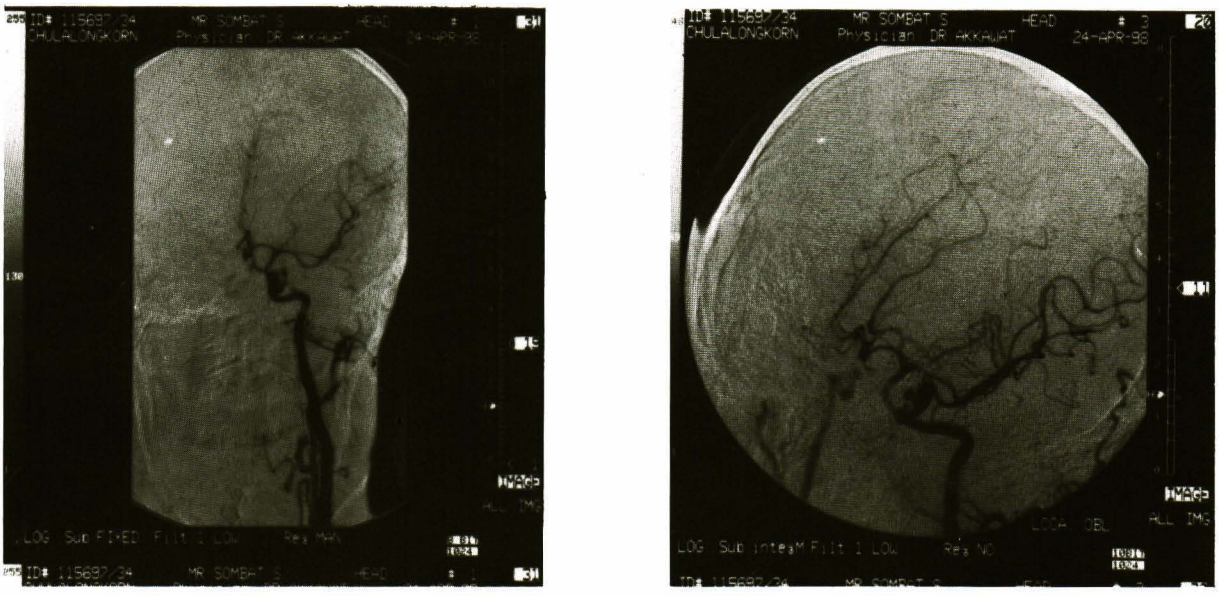

Figure 5. Left common carotid angiogram (post second operation), anteroposterior view(A), oblique view (B), demonstrating the residual aneurysm.

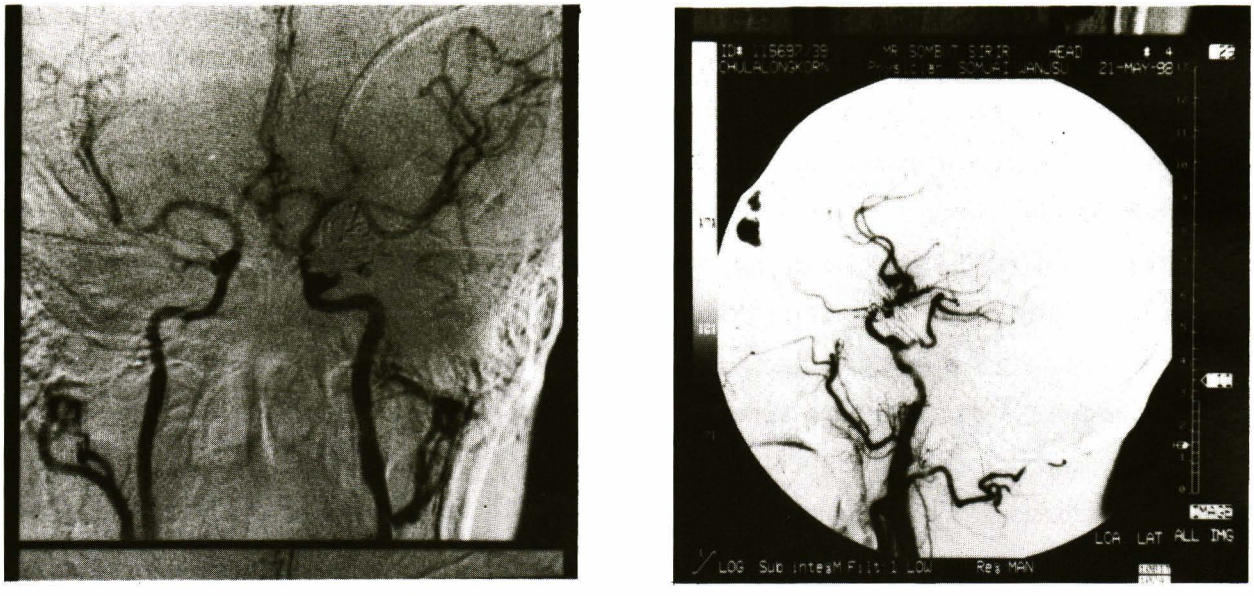

Figure 6. Left internal carotid angiogram (after third operation), demonstrating the complete clipping of the aneurysm and the preserved PTA.

Discussion

The persistent trigeminal artery (PTA) is a rare connecting channel between internal carotid artery and basilar artery. Richard Quain firstly described PTA first in 1844. [1] Paget, in 1948, systematized the embryological development of the cranial arteries in the human embryo. The anastomosis channels which provided blood supply to the hindbrain from primitive internal carotid artery was demonstrated. [2] The most important of them was the trigeminal artery. It develops since the 3 mm. stage of an embryo and usually regressed completely at the 45 mm. stage. It was demonstrated in 0.06 - 0.6 % of all cerebral angiograms, [3,4] Twenty five percent were associated with intracranial vascular anomalies such as aneurysm, arteriovenous malformation, agenesis or occlusion of ICA, moyamoya disease. [5-7]

The major sites of aneurysm associated with PTA were the circle of Willis or the PTA itself. Of the 29 PTA aneurysms reported in the literature, [3, 7, 8 - 30] 13 were located at ICA-PTA junction, 10 on the PTA trunk, 2 at the PTA-BA junction, 2 showed ICA-PTA dilatation. The location was not mentioned in another 2 cases.

There were 13 surgically treated cases of PTA aneurysm. (Table 1) Four cases underwent direct clipping, one was occluded by detachable balloon and another 8 cases had the proximal internal carotid artery occluded with or without bypass. [8, 11 - 13, 16, 18 - 20, 23 - 24, 27, 28, 30]

Linskey et al noted the relatively benign natural history of intracavernous ICA aneurysm, the majority of which do not require any therapeutic intervention. [31] Treatment should be considered the in those asymptomatic cases only when lesions extend into the subarachnoid space or arise at anterior genu of the carotid siphon or in patients presenting with symptomatic intracavernous aneurysms associated with subarachnoid hemorrhage, epitaxis, severe facial or orbital pain, progressive ophthalmopleia, visual loss or radiographic evidence of progressive enlargement. Alternatives as to the treatment of those selected patients include direct intracavernous clipping, proximal ligation of ICA (with or without bypass) or endovascular obliteration with microcoils or balloons. [32,33]

As for the PTA aneurysm, anatomically it is an intracavernous aneurysm and therefore, direct clipping is difficult because of the proximity in location and an intimate relation to the venous structure and to the cranial nerves in cavernous sinus. In inaccessible cases, occlusion of the proximal ICA by surgical ligation using Selverstone clamp or detachable balloon are all acceptable. Collateral circulation, however, must be assessed to determine whether proximal ICA ligation is a justified technique. The status of the collateral circulation also dictates the requirement of prophylactic EC-IC bypass. The type of PTA whether it is the adult type (posterior circulation is independent of PTA) or the fetal type (posterior circulation depends on PTA) must be carefully evaluated. In case of the fetal type, either preservation of PTA is necessary or an additional bypass from the anterior circulation to PCA is required.

Although surgically problematic difficult intracavernous aneurysms can be obliterated by endovascular techniques, the potential of recanalization, coil compaction, mass effect and long term outcome is still not well defined. Therefore, we chose not to perform an endovascular obliteration in this

Table 1. Surgical cases of PTA aneurysm.

No. Author (year)	Age	Sex	Location	Rupture	Sign and symptom	Surgery	Outcome
1. Davis(1956)	?	?	?	?	?	?	?
2. Bull(1969)	43	F	R.PTA	-	CSS (R. II,DV)	ICA Ligation	Died
3. Morrison(1974)	39	F	R.ICA-PTA	+	SAH ,R. V	AN Clipping	R.VI palsy
4. Enomoto(1977)	42	F	R.ICA-PTA	+	CCF,R.V,VI	CA Ligation	R.VI palsy
5. Kodama(1984)	75	F	R.ICA-PTA	+	CCF ,R.III,V	AN Clipping	Improved
6. Yamaki(1987)	55	F	R.ICA-PTA	-	R.III	ICA Ligation PTA Clipping STA - MCA	Improved
7. Higashida(1987)	69	F	L.ICA-PTA	-	CSS(L.III,VI)	AN Ball.Occl	Improved
8. Debrun(1988)	67	F	R.PTA	-	CSS(R.III,VI)	ICA Ball Occl [a] STA-MCA	Improved
9. Miyatake(1990)	57	F	L.ICA-PTA	-	CSS(L.VI)	ICA Ball Occl [b] (distal) STA-MCA,IC Ligation	Improved
10. Tokimura(1991)	44	F	R.ICA-PTA	+	SAH	AN Clipping	R III,IV paresis
11. Tsuboi(1992)	67	F	L.ICA-PTA	-	CSS(L.VI)	PTA Clipping STA-MCA,IC Ligation	Improved
12. Ahmad(1994)	51	F	R.ICA -PTA	-	CSS(R.V,VI)	PTA Clipping EC-MCA V.graft	Improved
13. Hayashi(1994)	47	F	R.ICA- PTA	+	SAH	AN Clipping	Improved
14. Present case	48	M	L.ICA-PTA	-	CSS(L.III,IV,V,VI)	AN Clipping	L.opthalmoplegia

Abbreviations: PTA = persistent trigeminal artery; DV = double vision; CC = common carotid artery; IC = internal carotid artery; EC = external carotid artery; STA = superficial temporal artery; MCA = middle cerebral artery; AN = aneurysm; CCF = carotid – cavernous fistula; CSS = cavernous sinus syndrome; IC ball. occl[a] , balloon occlusion of IC (distal to, at the level of and proximal to PTA); IC ball occl [b], balloon occlusion of IC (distal to PTA); L = left; R = right.

patient. Because of the poor collateral circulation, the timing of ICA occlusion followed by bypass potentially places the patient at risk of ischemia until the bypass is completed. Besides, long-term patency of bypass or graft operation has not been evaluated in our hospital. Direct surgical approach , therefore, was considered in this case.

There are auxiliary techniques in case of cavernous aneurysms where the direct approach is difficult. Parkinson[34] has combined his direct approach of the cavernous sinus with a simultaneous use of extracorporeal circulation and cardiac arrest. Recently, circulatory arrest has been introduced in the treatment of intracranial aneurysm. [35,36] The

advantages of this method are dry surgical field, easy to dissect and the aneurysms can be obliterated by direct clipping of their neck because it is facilitated by the absence of arterial pressure. The disadvantages include the necessity of a second surgical team and a second operative field, as well as of full heparinization, the limitation of cardiac arrest time, and arrhythmia due to hypothermia. Due to the fact that the aneurysm was inaccessible during the 1st operation, the direct surgical approach with deep hypothermia, circulatory arrest and barbiturate protection deemed appropriate.

In the treatment of PTA aneurysm, based on the complexity of its anatomy, it is difficult to clip directly. It is also important to know the hemodynamics of the collateral circulation and the type of PTA (adult or fetal type) before trapping procedure is performed. In case of inadequate collateral circulation, high flow vein graft or low flow STA - MCA anastomosis should be performed. There are many surgical techniques for the treatment of intracavernous aneurysms. Auxiliary techniques such as deep hypothermia and cardiac arrest, can be successfully applied once facing with such a difficult large cavernous aneurysm.

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