Nationwide surveillance of IC anterior (or dorsal) wall aneurysm: with special reference to its dissecting nature

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Summary

Two hundred and twenty-one cases of IC dorsal aneurysm (ICDA) with subarachnoid hemorrhage (SAH) from 365 cases in the nationwide surveillance of ICDA (NSICDA) data bank were studied with special reference to the dissecting type. Dissection of the internal carotid artery (ICA) was confirmed in 50 out of 221 SAH cases. In 193 surgically treated cases, 40 were of the certified dissecting type. Including those with clinical features which strongly suggests the existence of dissecting changes in the ICA wall, 97 cases (55.6% of operated) were thought to be a dissecting type. Incidence of intraoperative bleeding is significantly higher and surgical outcome is significantly worse in the dissecting type than in the non-dissecting type. Treatment options for this peculiar and formidable aneurysm (An) are described.

Keywords: IC dorsal aneurysm; arterial dissection; nationwide surveillance.

Introduction

Aneurysms arising from the superior (or anterior) wall of the C1-C2 portion of the ICA have been known as an ICDA [6, 11–13] or, in Japan, IC anterior wall aneurysm [9, 13]. The term "blood blister-like aneurysm" has been widely used for the non-saccular and wide-based type as well [1, 2, 4, 7, 13, 14, 16–18]. ICDA has also been well known as a strange and dangerous aneurysm that easily ruptures at the neck during surgery [1, 3, 5, 7, 8, 10–14, 17, 18]. In addition, it often regrows and reruptures even after seemingly successful clipping [1, 3, 6, 8, 9, 11–15]. The reason why this peculiar An shows these strange clinical manifestations has not yet been fully clarified. In this report, we will show the data obtained from the nationwide surveillance on the ICDA in Japan with special reference to its dissecting nature and present some illustrative cases from our own series.

Materials and methods

NSICDA was conducted in 2004 by retrograde registration of cases with ICDA treated in the period between 2001 and 2003 in hospitals approved by board committee of Japan Neurosurgical Society. ICDA was defined simply as an aneurysm on the superior (or anterior) wall of C1-C2 portion of the ICA without any branch artery around the neck. Four hundred and ninety-nine out of 1237 board-approved institutes (40.3%) responded to the surveillance. Eventually 365 cases with ICDA, of which 144 were incidentally found as an unruptured An, and 221 (comprising 60.5% of the total) found with SAH, were registered from 181 hospitals. In this report, 221 SAH cases were studied in detail especially with regard to its dissecting nature. Neurological grade on admission to hospital was assessed by Hunt-Kosnik's grading, and outcome at discharge by Glasgow Outcome Scale (GOS). Statistical analysis was made by Student's *t*-test, Chi-square test or Mann–Whitney *U*-test.

Angiographical characteristics of ICDA

ICDA arises exclusively from the superior (or anterior) wall of the C1-C2 portion of the ICA without any branching artery and often shows unusual and peculiar features on angiogram. It may look as a saucer-like bulging without any neck portion (Fig. 1A), a broad based and double contoured bulging (Fig. 1B) or a quite irregularly ragged shaped dome like a bunch of grapes (Fig. 1C). Abnormal narrowing of the ICA, proximal or distal to the dome, may frequently be associated with the ICDA (Fig. 1B). These peculiar figures of ICDA strongly suggest that it is different from the ordinary aneurysm with which neurosurgeons are facing in their daily practice, and the possibility is implied that many of these unique An might be dissecting in nature.

Cases with dissection in NSICDA

Fifty-six cases, comprising 15.5% of the 361 total cases, were registered to NSICDA as clinically confirmed dissecting aneurysm. Restricted to 221 cases manifested with SAH, 50 (22.5%) were dissecting. Postoperative mortality rate of dissecting aneurysm is 25% and that of the non-dissecting type is 17.8%; hence the overall surgical outcome of

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Fig. 1. Abnormal angiographic figures of ICDA: ICDA appears as various abnormal figures on angiogram. A) a saucer-like bulging without any neck portion, B) a broad based and double contoured bulging with abnormal narrowing of the ICA proximal (*or distal*) to the dome, C) an irregularly ragged shaped dome like a bunch of grape



Fig. 2. Surgical outcome of dissecting and non-dessecting type ICDA: A) In 221 ruptured ICDA, 193 cases were surgically treated, of which 40 were reported to be a certified dissecting type. Surgical outcome of dissecting type is significantly (P < 0.05) worse than that of non-dissecting type. B) Adding 30 cases of non-dissecting type, in which intraoperative neck tear took place, to the dissecting group, surgical outcome is more significantly (P < 0.01) worse in the group than the group of remainders

the first type is significantly worse (P < 0.05) than that of the latter one (Fig. 2A). There were 30 cases out of 153 operated non-dissecting aneurysms in which serious intraoperative bleeding occurred in a way that the aneurysmal neck tore off from the parent ICA wall instead of bleeding from the dome. Since this kind of wall disruption (direct neck tear) is quite unusual in normal berry-type An, this type may well be regarded as dissecting in nature. Including cases with intraoperative neck tear definitely into the dissecting group, the postoperative mortality rate becomes 31.4%, and overall outcome is essentially the same as that of pure dissecting group (Fig. 2B).

Illustrative case

Case 1

A 45-year-old housewife was admitted with severe headache which had suddenly occurred 2 h earlier. She was awake and showed no neurological abnormality other than headache. Brain CT scan on admission revealed SAH restricted within the right side of the basal and ambient cisterns (Fig. 3A). Cerebral angiogram on the day of admission demonstrated no An on the right side, while ICDA was present on the left C2 portion (Fig. 3B and C). Since no An was found on the right side where the blood clot was exclusively present, early surgical exposure was abandoned. The patient suddenly became semicomatose (grade 4) on day 5, and immediate angiogram revealed a newly developed round bulging on the superior wall of the right C2 portion (Fig. 3D). She died of brain damage caused by the rebleeding, and permission for autopsy was given by her family.

Histopathological finding of the right ICA disclosed dissection of the wall at the ruptured site (Fig. 4A), and dissection of the left ICA where unruptured ICDA was present (Fig. 4B).

Case 2

A 55-year-old female was transferred from a local hospital where she suffered the second SAH and had become semicomatose. Brain CT scan



Fig. 3. Case 1; 45-year-old housewife with bilateral ICDA: A) Brain CT scan on admission revealed SAH restricted within the right side of the basal and ambient cisterns. B) Right carotid angiogram on the day of admission demonstrated no aneurysm. C) An aneurysmal bulging, which was definable as ICDA, was present on the left C2 portion (*arrow*). D) Right carotid angiogram taken immediately after the rebleeding on the day 5 revealed a newly developed ICDA in the C2 portion (*arrow*). See the text for the details





demonstrated SAH with perifocal edema around the right Sylvian fissure and intraventricular hemorrhage (Fig. 5A). An angiogram taken at the previous hospital disclosed a fusiform bulging of the superior wall of the right C1–C2 (Fig. 5B). She died 5 days after admission, and autopsy revealed a definite dissection at the bulging site (Fig. 5C).

Discussion

ICDA is quite a peculiar type of An exhibiting unique features on angiogram as demonstrated in illustrative

cases and shows unusual clinical manifestations such as preoperative rapid growth of the dome, intraoperative neck tear, postoperative rebleeding or regrowth of dome, or postoperative ICA narrowing or obstruction [1, 3, 5, 7, 8, 10–15, 17, 18]. What sort of pathogenetic condition could produce these abnormal clinical characteristics of ICDA? In 1993 the authors suggested that many of the ICDAs might be dissecting in nature and aneurysmal bulging be a pseudo-An like fragile lump without any



Fig. 5. Case 2; 55-year-old female with right ICDA: A) Brain CT scan on admission demonstrated SAH and intraventricular hemorrhage with perifocal edema around the right Sylvian fissure. B) Right carotid angiogram taken at the previous hospital disclosed a fusiform bulging of the superior wall of the right C1–C2 portion (*arrow*). C) Histology of the right ICA revealed a definite dissection at the bulging site (*arrow*). See the text for details. Elastica-Van Gieson stain; $\times 10$ magnification

definite histological evidence [10]. Soon after that proposal, we happened to have an autopsy case in which the ICA wall dissection was histologically confirmed (illustrative cases 1 and 2) [11]. Numerous reports have been published thereafter describing the dissecting change of the ICA wall found in cases with ICDA [1, 2, 6–8, 10, 11, 15]. Many of the ICDAs can be regarded as usual berry type An because the wall and the neck of the dome seem to be as firm and thick as in the ordinary An and to which clipping can be done safely. On the other hand, many of the ICDAs have undoubtedly an abnormally thin and fragile wall, where the clipping procedure may possibly cause disastrous neck tear leading very often to fatal outcome. The NSICDA study undertaken in Japan confirmed the inference that a dangerous and abnormally shaped ICDA is dissecting in nature since more than 20% of the registered SAH cases were reported to be definitely dissecting. Probably the true incidence of dissecting An is by far beyond this range because there are many that present abnormal clinical features like postoperative re-rupture or regrowth of the dome, extremely thinned wall of the neck, or intraoperative neck tear besides those registered as dissecting type. Including these abnormal type An into the dissecting group, at least 97 out of 193 operated SAH cases (50.3%) are suspected to be of the dissecting type. Incidence of intraoperative bleeding was significantly more frequent (P < 0.01) in the suspected dissecting group (55.6%) than in the remaining 96 cases (17.7%).

The overall surgical outcome was also significantly worse (P < 0.01) in the suspected dissecting group than in remaining cases (Fig. 6). Mortality of the former group is 28.9% while that of the latter is 17.7%.

It is well known that clipping is not safe and reliable treatment for arterial dissection. Since we can confirm that many ICDAs are dissecting in nature, we have to warn that any attempt for simple clipping must be avoided if the facing ICDA shows any clinical or angiographic signs different from ordinary An. In an analysis of NSICDA data bank which we have published elsewhere [14], the postoperative outcome of early surgery within 96 h after the onset of SAH is significantly worse than that of delayed one (P < 0.02; data not shown in this report). At the chronic stage, healing processes of the dissecting site may set in and make the fragile tissue much firmer and tight, and this will allow the neck to be clipped in the ordinary way in most cases.

Judging from the data described here, we recommend the following option for the treatment of ICDA suspected to be of the dissecting type.

- 1. In case of early surgery, sufficient preparations for dangerous hemorrhage during the surgical procedure is required including securing the cervical ICA before opening the dura, drilling off the anterior skull base in order to secure wide surgical space and the proximal C2 portion, and to be ready for emergency EC-IC bypass.
- 2. A high-flow bypass and trapping of the ICA instead of attempting to obliterate the dome by clipping is probably the best option for early surgery at the present.
- 3. If self-expandable stent is available, stenting the C1-C2 portion with or without additional coil packing to the dome might be the optimal treatment [4, 16].
- 4. If patient's condition permits without rebleeding at the acute stage, delayed surgery should be considered.

Conclusions

Precise analysis of the NSICDA data base disclosed a high incidence of dissecting type An in ICDA manifested by SAH, of which surgical outcome is significantly worse than those without dissecting features. As the outcome of early surgery is significantly worse than that of delayed one, an attempt of simple clipping at acute stage without sufficient preparations is to be avoided in order to cope effectively with hazardous intraoperative bleeding.

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