CASE-BASED UPDATE

Iatrogenic intracranial aneurysms in childhood: case-based update

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Abstract

Purpose Iatrogenic aneurysms are very rare in children. Characteristic clinical manifestations are variable and asymptomatic course is possible especially for fusiform dilatation of internal carotid artery. Even though radiological diagnosis is easy, the management of iatrogenic intracranial aneurysm is still a subject for discussion.

Methods Fusiform dilatations of internal carotid artery were diagnosed on three pediatric patients during follow-up imaging after primary surgery for suprasellar–parasellar tumor. All patients were asymptomatic. Conservative treatment was proposed because the lesion did not show any progression in subsequent examinations. Patients are stable under conservative treatment.

Conclusions Iatrogenic aneurysm may have an unusual presentation and their therapy still remains unclear. Fusiform dilatation of internal carotid artery rarely causes symptoms and there is no published paper of subarachnoid bleeding. Treatment would be difficult, since the main arterial branches arise from the dilated carotid segment. Conservative treatment is a choice only if aneurysm has no progression or in case of spontaneous healing. Intervention should be performed only in case of progression or if the aneurysm becomes symptomatic.

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Introduction

Intracranial aneurysms are extremely rare in the pediatric population in comparison with adults. Only 1–5 % of all intracranial aneurysms are shown by the pediatric population [7, 26]. Intracranial aneurysms affect more frequently male children than females (1.3–2.8:1) [5, 7, 26]. Infections and trauma are most common risk factors for childhood intracranial aneurysms [5, 7, 26, 28]. Approximately, 33 % of childhood aneurysms are traumatic [5, 16, 28]. Prompt treatment by surgery or endovascular techniques should be done because traumatic aneurysms may gradually expand and rupture in a few weeks or months [3, 24, 26]. Although urgent treatment is necessary to prevent intracranial hemorrhage, especially pseudoaneurysms are usually difficult to treat without sacrificing the parent artery [3, 24].

Iatrogenic aneurysms take a small part among traumatic aneurysms. To our knowledge, 37 cases of iatrogenic intracranial aneurysm in children have been reported to develop after surgical interventions, such as tumor removal [2, 5, 6, 10, 16, 21–23], endoscopic procedures [8, 14, 18, 26], ventricular puncture or shunting procedure [9, 19, 20, 27], aspiration of subdural hematoma [17], aspiration of abscess [12], myringotomy [25], temporary clip occlusion [28], and decompressive craniectomy for closed head injury [28]. The clinical data of those cases are displayed in Table 1. The best way to manage iatrogenic intracranial aneurysm is still a subject for discussion, although it was like that for posttraumatic aneurysms. Most authors are in favor of a prompt surgical or endoscopic treatment [3, 24, 26]. Fusiform aneurysmal dilatation of the internal carotid artery (FDCA) is another vascular complication that can occur following radical surgery for sellar-parasellar region tumors [2, 10, 16,

| Table 1 | | Cases of iatrogenic intracranial aneurysms in childhood (r | acranial aneur | rysm: | s in childhood (review of literature) | ature) | | | | | |
|---------|------|--|----------------|-------|---------------------------------------|----------------------------|-------------------------|---------------------------|--------------------------------|------------------------|--------------------------|
| No. | Year | Author | Age S | Sex | Sex Primary surgery | Rx | Site | Clinic presentation | Treatment | Course | Interval to diagnosis |
| 1 | 1966 | Overton and Calvin | 9 months N | М | Aspiration of subdural hematoma | No | Cortical artery | Asymptomatic | Excision | NA | 8 weeks |
| 2 | 1974 | Lassman et al. | 16 years N | Σ | Aspiration of abscess | No | MCA | NA | Conservative | NA | 10 weeks |
| 3 | 1976 | Scharfettter et al. | 3 years N | М | Ventricular puncture | No | Angular gyrus artery | NA | Clip - excision | N/A | 2 weeks |
| 4 | 1985 | Trammer et al. | 4 years F | ц | Myringotomy | No | ICA | NA | Clip - occlusion | NA | 5 days |
| S | 1987 | Gutierrez et al. | 15 F | Ľ۲ | Tumor removal | NA | PICA | NA | Clipping | NA | 4 years |
| 9 | 1991 | Sutton et al. | 9 years | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 16 months |
| 7 | 1991 | Sutton et al. | 6 years N | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 5 months |
| 8 | 1991 | Sutton et al. | 12 years F | ш | Tumor removal | Yes | ICA | Asymptomatic | Clipping | Death because of tumor | 15 months |
| 6 | 1991 | Sutton et al. | 4 years 1 | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 4 months |
| 10 | 1991 | Sutton et al. | 19 years N | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 17 months |
| 11 | 1991 | Sutton et al. | | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 7 months |
| 12 | 1991 | Sutton et al. | | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 13 months |
| 13 | 1991 | Sutton et al. | | NA | Tumor removal | Yes | ICA | Asymptomatic | Conservative | Constant | 13 months |
| 14 | 1991 | Sutton et al. | | NA | Tumor removal | Yes | ICA | Asymptomatic | Conservative | Constant | 8 months |
| 15 | 1994 | Ventureyra and | | ц | Decompressive craniectomy | No | ACA | NA | Clip - excision | Death | 7 weeks |
| 16 | 1994 | Higgins Ventureyra and | 5 years N | М | Temporary clip occlusion | No | MCA | NA | Clip - excision | NA | 44 months |
| 17 | 1994 | Sutton | 10 years | NA | Tumor removal | Yes | ICA | Headache | Clipping | No further aneurysm | 10 years |
| 18 | 1994 | Sutton | | NA | Tumor removal | Yes | ICA | Asymptomatic | Conservative | Constant | 8 years |
| 19 | 1995 | Lakhanpal et al. | | М | Tumor removal | No | ICA | Visual impairment | Surgical | Constant | NA |
| 20 | 1995 | Lakhanpal et al. | 12 N | М | Tumor removal | Yes | ICA | NA | reconstruction Conservative | Constant | 16 months |
| 21 | 1997 | McLaughlin et al. | 3 years F | ĹŢ. | Endoscopic Procedure | No | Basilar artery | NA | Clip ligation | Complete obliteration | 1 month |
| 22 | 1998 | Bendszus et al. | | NA | Tumor removal | No^{a} | ICA | Asymptomatic | Conservative | Progressive | 15 months |
| 23 | 1998 | Bendszus et al. | 6 years N | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 15 months |
| 24 | 1998 | Bendszus et al. | 10 years N | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 15 months |
| 25 | 1998 | Bendszus et al. | 8 years N | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Progressive | 15 months |
| 26 | 1998 | Bendszus et al. | 14 years N | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 15 months |
| 27 | 1998 | Bendszus et al. | 10 years N | NA | Tumor removal | No | ICA | Asymptomatic | Conservative | Regressive | 2 days |
| 28 | 1998 | Bendszus et al. | 11 years N | NA | Tumor removal | No^{a} | ICA | Asymptomatic | Conservative | Progressive | 15 months |
| 29 | 1999 | Shirane et al. | 4 months F | ц | Shunting procedure | No | ICA | Generalized | Surgical | No further aneurysm | 3 weeks |
| 30 | 2001 | 2001 Horowitz et al | 3 vears N | Σ | Endosconic Procedure | Ŋ | Basilar artery | seizures A symptomatic | reconstruction Endovascular | Complete obliteration | 74 h |
| 2 | 1007 | | | | | | finn much | Augusta duri fer r | trapping | | 1 |

| No. | NO. ICAI AULIOI | Age | Sex | Sex Primary surgery | 2 | | | | | diagnosis |
|-----|--------------------------|------------|-----|-------------------------|----|----------------|------------------------------|------------------------------------|-----------------------|-----------|
| 31 | 31 2002 Tirakotai et al. | 10 years F | ц | Tumor removal | No | No ICA | Headache | Clipping | Complete obliteration | 1 year |
| 32 | 2006 Jenkinson et al. | 15 years | ш | Shunting procedure | No | No MCA | Loss of | Resection | NA | 8 weeks |
| 33 | 2006 Tubbs et al. | 10 years M | М | Shunting procedure | No | No ACA | consciousness Monoparesis | Surgical trapping | NA | 1 week |
| 34 | 2007 Dunn et al. | 6 years | Σ | Tumor removal | No | ACA | Asymptomatic | STA graft | NA | 3.5 years |
| 35 | 2008 Rezende et al. | 4 years | ц | Endoscopic Procedure | No | Basilar artery | Headache | interposition Coil embolization | Complete obliteration | 24 h |
| 36 | 2011 Trivelato et al. | 7 years | Σ | Endoscopic Procedure | No | ICA | Asymptomatic | Coil embolization | Complete obliteration | 3 months |
| 37 | 2012 Ogilvy et al. | 5 years | Μ | Tumor removal | No | ICA | Asymptomatic | Stent-assisted | Complete obliteration | 3 months |
| 38 | Personal Case 1 | 6 years | Х | Tumor removal | No | ICA | Asymptomatic | colling Conservative | Constant | 1 year |
| 39 | Personal Case 2 | 11 years | Ц | Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 1 year |
| 40 | Personal Case 3 | | Μ | 8 years M Tumor removal | No | ICA | Asymptomatic | Conservative | Constant | 2 years |

 Table 2
 Classification of traumatic aneurysms

| Mechanism of injury [9, 28] | Histopathological structure [4, 5, 9, 24] |
|--|--|
| TICA following closed head injury | True aneurysms |
| TICA following missile injury | False aneurysms (pseudoaneurysm) |
| TICA following penetrating head injury | Mixed aneurysms |
| TICA following iatrogenic injury | Dissecting aneurysms |

21–23]. The management of this vascular complication and its pathogenesis are still controversial [23].

Classification and pathology

Lasjaunias et al. classified childhood aneurysms in four groups: (1) dissecting aneurysms, (2) infectious aneurysms, (3) traumatic aneurysms, and (4) classic saccular aneurysms [11]. Group 1 and group 3 can be evaluated together. Traumatic intracranial aneurysms (TICA) can be classified according to their mechanism of injury or histopathological structure (Table 2).

FDCA is another entity. Focal arterial disruption while dissecting the tumor from arterial wall may lead to injury of the vasa vasorum [2, 22, 23]. This arterial wall weakening is thought to cause fusiform dilatation (Fig. 1). However, all wall layers are intact in FDCA; if the tumor itself invades the adventitia of internal carotid artery

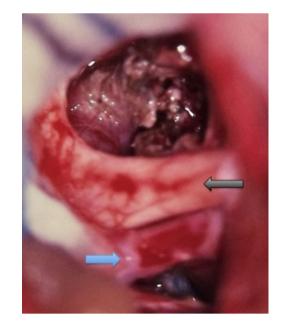


Fig. 1 Perioperative microscopic view of case 3. Superficial external wall injury of carotid artery (*blue arrow*) was noticed during excision of adamantinomatous craniopharyngioma via right pterional approach. Optic nerve (*black arrow*) was intact

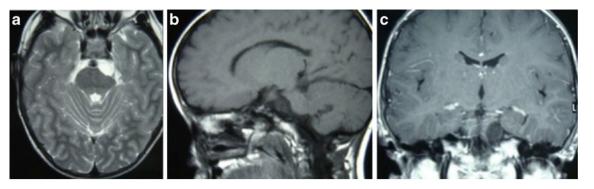


Fig. 2 Case 1, initial presentation of patient. Left cerebellopontine angle (PCA) mass which is slightly hyperintense to CSF in axial T2-weighted imaging (\mathbf{a}), slightly hyperintense to CSF in sagittal T1-weighted imaging (\mathbf{b}), and shows no enhancing in coronal T1 contrasted imaging (\mathbf{c})

(ICA), it may cause disruption during surgery [10, 21]. Also, another mechanism which injury of the sympathetic plexus of the ICA was postulated to lead to neurogenic vasoparalysis and, thus, arterial dilatation may occur [2, 23]. Radiation therapy might develop FDCA [23].

Clinical presentation

Two thirds of pediatric patients harboring traumatic aneurysms will experience a symptomatic aneurysmal hemorrhage [5, 28]. However, asymptomatic patients were also reported whose iatrogenic aneurysm was diagnosed during follow-up [5, 26]. Clinic presentation varies from simple headache to loss of consciousness and even sudden worsening due to acute bleeding is possible [9]. Dysphasia, hemiparesis, visual impairment, and seizure have been reported associated with iatrogenic intracranial aneurysms in childhood [9, 20]. After aneurysm rupture, mortality rate is more than 30 % [5, 14, 26–28].

Diagnostic imaging and evaluation

Although digital subtraction angiography (DSA) is still the gold standard technique, the role of computed tomography angiography (CTA) and magnetic resonance angiography (MRA) is still under evaluation even though some authors propose them as alternative to invasive techniques. A few reports have presented that CTA and MRA can diagnose aneurysm larger than 5 mm with an accuracy of 90 % [29, 30].

Doppler ultrasound detects anterior cerebral artery (ACA) and middle cerebral artery (MCA) aneurysms larger than 5 mm with sensitivity of about 0.82 and 0.79, respectively. A significantly lower accuracy was reported for aneurysms of the cavernous and terminal internal carotid arteries and posterior communicating artery [31].

Both true and false aneurysm may be fusiform or saccular [9]. Most common localization for TICA is the ACA (38 %) [5, 28]. Thirty-seven children with iatrogenic aneurysms were reported in the literature [2–4, 6, 8–10, 12, 14, 16–23, 25–28]. The acquired malformation involved the

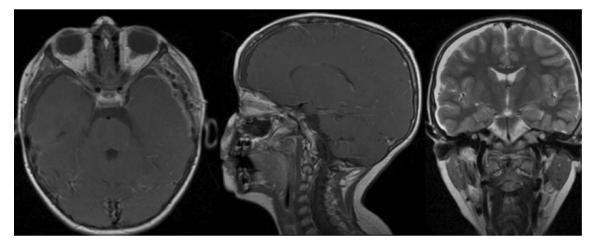


Fig. 3 Case 1, early postoperative MRI shows no residual tumor

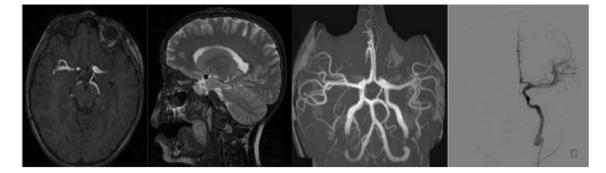


Fig. 4 Case 1, first year follow-up. Fusiform dilatation at supraclinoid segment of the left internal carotid artery was seen in MRA and then DSA

ICA in 25 cases. Additionally, a few iatrogenic aneurysms were reported to involve the ACA, the MCA, the posterior inferior cerebellar artery, and the basilar tip (Table 1).

There is no particular indication universally shared on how to search for iatrogenic aneurysms in children who had undergone intracranial surgery. However, Tirakotai et al. advice pediatric neurosurgeons to consider FDCA within 2 years after surgery for craniopharyngioma or midline gliomas [23].

Management and treatment

Conservative treatment

Although TICA try to expand gradually and rupture in a few weeks or months, spontaneous regression was also reported [3, 15, 24]. Nevertheless, even after spontaneous healing, angiographic follow-up should be continued because of possible risk for re-expansion.

There is no mention in literature about the management protocol for iatrogenic aneurysm in childhood. In general, conservative treatment is a choice only if the aneurysm does not progress or in case of spontaneous healing. Dunn et al. followed pericallosal artery aneurysm for 6 months which was initially interpreted as residual tumor [5]. The patient underwent surgery because of progression.

In case of conservative treatment, the patients should undergo annual follow-up magnetic resonance (MR) imaging studies to evaluate aneurysmal growth and stability [7]. Sutton et al. first reported in 1991 and then updated in 1994 nine (15.7 %) supraclinoid carotid artery aneurysms among 57 operated on children for craniopharyngiomas between 1982 and 1993 [21, 22]. Only one patient underwent aneurysm clipping while operating for regrowth of tumor and died because of tumor recurrence. Eight of the nine patients remain alive at a mean of 6.6 years after diagnosis. Similarly, in 1998, Bendszus et al. reported FDCA in 7 children among 62 (11.3 %) operated cases because of suprasellar– parasellar tumor [2]. Three children had progression and one child had regression in 6 months follow-up. However, all of them were conservatively followed (Table 1).

FDCA rarely causes symptoms and there is no published paper of subarachnoid bleeding (Table 1). In any case, treatment of FDCA would be difficult, since the posterior communicating artery and anterior choroidal arteries (and in some cases the middle cerebral arteries) arise from the dilated carotid segment [21, 22]. Therefore, according to the conservative option, intervention should be considered only in case of progression or if the aneurysm becomes symptomatic [2, 16, 21, 23].

Surgery

Although successful surgical interventions such as clipping, trapping, wrapping, resection, primary reconstruction, or arterial graft interposition have been described [3, 4, 6, 9, 12, 14, 17, 19, 20, 23, 25, 27, 28], especially pseudoaneurysms are not always suitable for surgical occlusion because of absence of a true neck or an aneurysm wall [18, 26]. They may be difficult to treat without sacrificing parent vessels [1, 3, 16, 24]. Surgery for dissecting aneurysm may also be

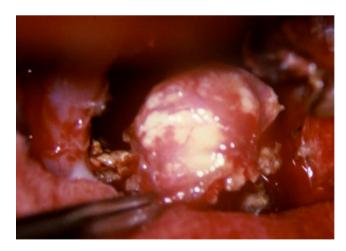
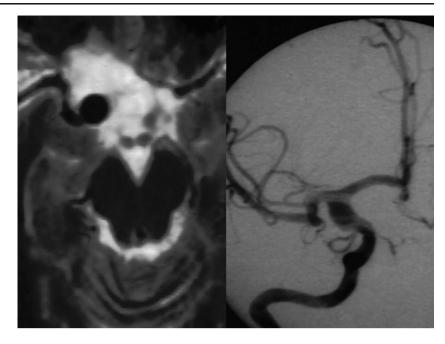


Fig. 5 Case 2, perioperative microscopic view of tumor showing the intimate relation of the calcified part of the tumor with the wall of the ICA

Fig. 6 Case 2, fusiform dilatation at supraclinoid segment of the right internal carotid artery was seen in MRA (*left*) and DSA (*right*)



difficult especially for the ones in vertebrobasilar system due to the risk of incidental damage to the vertebral perforating arteries [13].

Endovascular treatment

Endovascular techniques which is virtually less invasive than open microsurgery widely demonstrated to be associated with successful results. First, Horowitz et al. described endovascular treatment of a 30-month-old child with a traumatic basilar artery aneurysm after endoscopic third ventriculostomy [8]. The false aneurysm was managed with endovascular trapping without morbidity. However, endovascular trapping may be risky especially when the collateral blood circulation is not sufficient [8, 18]. Afterward, few selective endovascular coiling was reported for childhood iatrogenic aneurysm [18, 26]. Selective coiling should be done cautiously because traumatic aneurysms can be pseudoaneurysms. Fibrocollagenous capsule laceration may lead to rebleeding [1, 18, 24, 26].

Ogilvy et al. described stent-assisted coiling to FDCA which was operated for craniopharyngioma first [16]. The aneurysm was first diagnosed 3 months after surgery; stent-assisted coiling was performed 5 months later because of radiological progression. This technique suggests better vessel preservation [16].

Prognosis

There is no large series to determine prognosis of iatrogenic aneurysms but approximately two thirds of patients with traumatic aneurysm experience symptomatic hemorrhage, with an associated mortality rate of 30–41 % [5, 26, 28]. Surgical or endovascular obliteration of aneurysm reduces the overall risk of mortality [5, 26–28].

Conclusion

Iatrogenic intracranial aneurysms are very rare in childhood and hard to be recognized. These lesions may insidiously progress making their management a real challenge. Conservative treatment is a choice only if aneurysm has no progression or in case of spontaneous healing. Fusiform aneurysms should also be followed especially when involving the main arterial branches. Follow-up observation should be continued regarding the risk of expansion. Endovascular techniques are

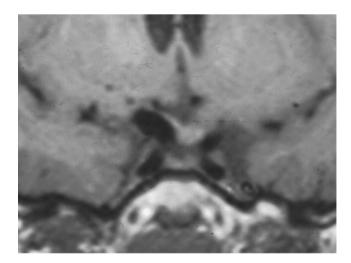


Fig. 7 Case 2, follow-up MRA showed no progression in fusiform dilatation of right internal carotid artery

effective therapeutic options when the aneurysms are considered as unclippable. Like surgical treatment, lethal complications may occur after endovascular treatment due to the fragile fibrocollagenous wall of aneurysm or lack of collateral arterial branches.

In spite of the good outcomes of iatrogenic aneurysms reported by the only two papers dealing with clinical series [2, 21, 22], most of the anecdotal reported cases have been treated surgically. This attitude may be influenced by the evolutive character of congenital aneurysm and the progression of the lesion often observed in cases following traumatic vascular injuries associated with head injuries.

Exemplary cases

Case 1

A 6-year-old boy, with a 3-month history of headache and transient dysarthria and progressing left hemiparesis, was admitted to Pediatric Neurosurgery Department of Rome Catholic University in 2008. His neurological condition improved with steroid administration. No history of minor or major cranial injuries was reported. Neuroimaging (MRI) revealed an extra-axial fatty mass at the left cerebellopontine angle extending to the prepontine cistern (Fig. 2). Auditory brain stem response and motor-evoked potentials were normal. He underwent surgery, and the lesion was gross totally removed by left pterional approach. After the operation, he experienced partial transitory deficit of the left third cranial nerve. Brain MR on the fifth postoperative day revealed no residual tumor (Fig. 3). Histopathological examination revealed an epidermoid cyst. A 6-mm-diameter fusiform aneurysm at terminal internal carotid artery with extension to M1 was diagnosed after 1 year of follow-up (Fig. 4). He was asymptomatic. Conservative treatment was proposed because of absent progression. After 4 years follow-up, he presents normal psychomotor development and his hemiparesis and third nerve palsy have totally recovered. Fusiform aneurysm is stable under conservative treatment.

Case 2

A 11-year-old girl, with a 5-month history of anorexia, was admitted to Pediatric Neurosurgery Department of Rome Catholic University in 1988 because of severe weight loss. Neurological examination was normal. No history of minor or major cranial injuries was reported. Neuroimaging (CT scan) revealed 2.5-cm-diameter sellar–suprasellar and solid-cystic mass with gross calcifications. This mass had prechiasmatic extension. She underwent surgery, and the lesion was gross totally removed by right pterional approach. External wall damage on the right ICA was seen at the end of the procedure without, however, no aneurysmatic dilatation (Fig. 5). After the operation, she experienced insipid diabetes, nystagmus, and hemianopsia. Histopathological examination diagnosed adamantinomatous craniopharyngioma and complete recovery of her postoperative neurologic deficits. Patient was discharged with hormone replacement therapy because of hypopituitarism. A fusiform aneurysm at supraclinoid segment of the right internal carotid artery was diagnosed during follow-up (Fig. 6). The lesion was not association in any sign or symptom. Conservative treatment was proposed and the lesion did not show any progression in subsequent examinations (Fig. 7). At last follow-up examination, 24 years after first surgery, no changes on size or shape of the aneurysm were noticed on the MR control. She works as a nurse under hormone replacement therapy.

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