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Endovascular treatment for radiation-induced internal carotid artery pseudoaneurysm and usefulness of angiographic and nasal endoscopic confirmation

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ABSTRACT

Radiation induced carotid vasculopathy may present as steno-occlusive disease or less commonly as a pseudoaneurysm. Rupture of internal carotid artery (ICA) pseudoaneurysm can be a fatal complication in patients with nasopharyngeal carcinoma (NPC). Location of the pseudoaneurysm at the skull base makes surgical treatment very difficult. Endovascular therapy may be the treatment of choice. Preserving patency of the carotid artery is a desirable option, but incomplete occlusion of bleeding point of pseudoaneurysm may cause re-bleeding. Both angiographic and nasal endoscopy confirmation of occlusion may help to avoid re-bleeding. We describe a case of endovascular treatment for massive epistaxis due to rupture of radiation-induced ICA pseudoaneurysm in a patient with NPC, and would like to discuss significance of angiographic and nasal endoscopic confirmation.

1. Introduction

Saccular aneurysms of the petrous segment of internal carotid artery (ICA) are relatively uncommon vascular lesions. Most of the aneurysms are pseudoaneurysms that are related surgery such as carotid endarterectomy, trauma to the neck, mycotic infection, tumor invasion, chemotherapy, radiation, or iatrogenic injuries (block procedure, manual therapy, rehabilitation treatment …..) [1–3]. Presenting symptoms related to this aneurysm may include painless pulsating neck mass, hemorrhage, or embolic strokes [2,4]. Although less common, upper respiratory tract hemorrhage can be occurred. Sudden and massive epistaxis is usually fatal, and it is one of the most dreaded complications among patients with cancer of the head and neck region [5–8]. These situations have to require emergent diagnosis and treatment. Because of the location within the petrous bone, if not impossible, a direct surgical approach is difficult and it was reported that it carries a substantial risk with rates of stroke and death of up to 9% and with cranial nerve injury in as many as 23% [9]. With more advance of endovascular techniques in the field of neurosurgery, neurosurgeons are often consulted for such clinical presentations and their management from other departments. We describe a case of emergent endovascular treatment for massive epistaxis due to rupture of radiation-induced ICA pseudoaneurysm in a patient with NPC, and would like to discuss significance of angiographic and nasal endoscopic confirmation.

2. Case

A 63-year-old woman with significant past medical history of nasopharyngeal cancer (NPC) who was admitted to other (otolaryngology) department with massive epistaxis and syncope. She was treated in 2008 with surgery, radiation and chemotherapy. Few days ago before admission to otolaryngology department, she had nasal inflammatory symptom and epistaxis. But at that time epistaxis was not massive bleeding and stopped spontaneously. On admission day, the patient was hemodynamically unstable with massive active bleeding. Therefore immediate salvage procedure, compact nasopharynx packing and subsequent tracheostomy were performed by otolaryngologists. After that the patient was consulted to our department for evaluation and further treatment about massive epistaxis and syncope. There was no vascular abnormality such as pseudoaneurysm in petrous segment of the left ICA in enhanced neck CT (Fig. 1). Cerebral angiogram was performed to evaluate vascular distribution of nasal cavity area, and if possible, to embolize nasopharyngeal branches. In this process, anteroposterior and lateral cerebral angiograms confirmed the presence of a 13 mm saccular form pseudoaneurysm in just proximal to the mid petrous segment of the left ICA, which was not seen on CT a few days ago (Fig. 2). Immediately,

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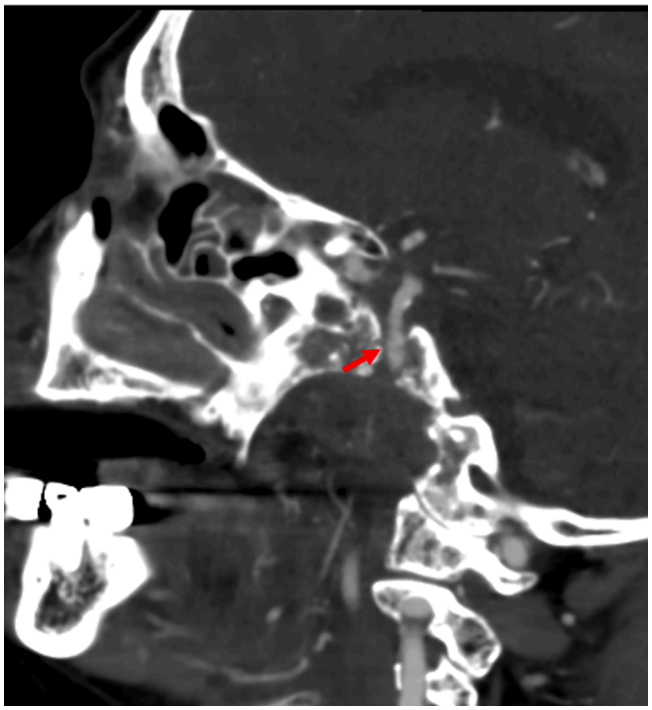

Fig. 1. There was no definite abnormality (no definite pseudoaneurysm) in enhanced neck CT in just proximal to the mid petrous segment of the left ICA (red arrow). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

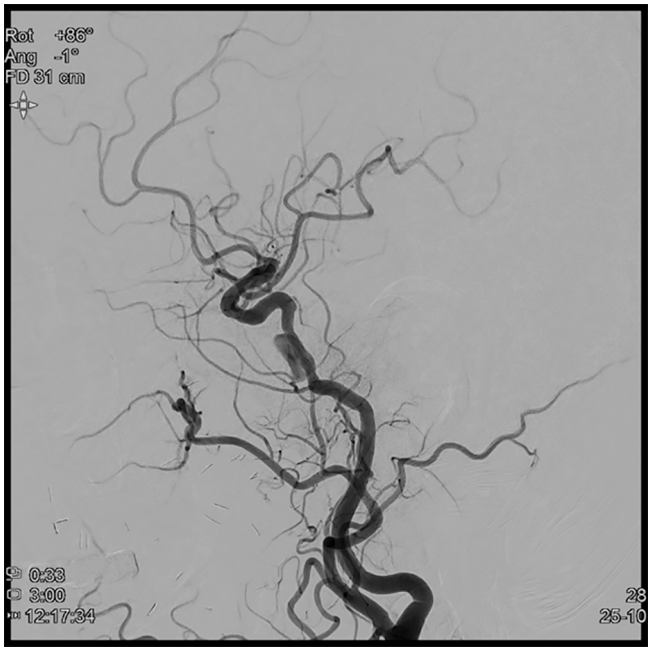

Fig. 2. Anteroposterior and lateral angiograms confirmed the presence of a 13 mm saccular form pseudoaneurysm that was not seen on CT a few days ago, just proximal to the mid petrous segment of the left ICA.

endovascular coiling was planned. During coiling, As the aneurysm was filled with more coils, diameter of the internal carotid artery became more narrower. Blood flow was more and more compromised. Under suspicious of dissecting pseudoaneurysm, stent (Neuroform Atlas, Stryker Neurovascular, Fremont, CA) was deployed in neck area immediately. Blood flow and lumen of ICA was marked improved after stent deployment. Aneurysm was completely occluded in final angiogram and the procedure was finished successfully (Fig. 3A–C). There was no more epistaxis after treatment. After procedure, the dual antiplatelet treatment began immediately. A few days later, nasal endoscopy was performed. The suspected point of bleeding was confirmed, but coil was not visible in this area (Fig. 4). 1 weeks later, follow up MRI revealed small flow signal in aneurysmal neck portion (Fig. 5). Because there was no active, massive bleeding, the patient was discharged without additional coiling, in expecting of occlusion by thrombus formation. Three weeks later, the patient had significant epistaxis (amount of soaking two hand towel). In repeated angiogram, more coil compaction and small blood leakage from previously small remained area of aneurysm neck were shown. Immediately, Additional coiling was done in remnant area and complete filing was achieved (Fig. 6A–C). This time, occlusion of leakage area by coil was confirmed in nasal endoscopy performed a few days later (Fig. 7A and B), and no more epistaxis was not occurred. There was no procedure related complication, and the patient had no neurological deficit and epistaxis on 3 months follow up periods.

3. Discussion

Radiotherapy of neck region can result in delayed development of a carotid artery pseudoaneurysm, which is one of the uncommon complications after radiation treatment of NPC. It typically occurs at the petrous and laceral segment of the ICA, and may present with life-threatening epistaxis. The mechanism of vascular damage caused by radiation is not fully understood, but it is known that it may be caused by the obliteration of vasa vasorum, premature atherosclerosis, and weakening and necrosis of the arterial walls. Ischemic injury combined with periadventital fibrosis may contribute to the rupture of irradiated large arteries and pseudoaneurysm formation. In addition, combined with high blood pressure of the vessel, it can result in the rupture of the arterial wall and even dissection with extravasation of blood [10–14]. As a second course of radiotherapy, the patient may have a higher risk of radiation damage to the surrounding tissue and be more prone to skull base osteoradionecrosis. As subclinical infection from osteonecrosis of the skull base is also implicated in the development of ICA pseudoaneurysm, there are increased chances of development of ICA pseudoaneurysm following a second course of radiation, which is also associated with increased chances of osteonecrosis of the skull base [12,15]. Carotid artery damage after cervical irradiation is not commonly occurred. In a 10-year study of patients who received cervical radiation for the tongue carcinoma, no cases of pseudoaneurysm or rupture were observed in 68 patients [16]. As similar to our patient, if it does occur, development of pseudoaneurysm may have many years after the conclusion of radiation therapy [17].

Until now, treatment of a pseudoaneurysm in the petrous segment in ICA includes either sacrifice or preservation of the ICA. Surgical management is technically challenging operations and have been associated with high morbidity and mortality rates [9,18]. The large study of carotid ligation in patients with head and neck cancer reported that complications including death or serious morbidity including coma, hemiplegia, or monoplegia occurred in 30.1% [19,20]. Sacrifice of the carotid artery would lead to severe cerebrovascular events, and therefore a balloon occlusion test should be performed before treatment. If the patient tolerate temporary balloon occlusion test, trapping or parent artery occlusion is an option. But, 5–22% of patients passing the occlusion test developed ischemic complications, including cerebral infarct [21,22]. If the patient cannot pass balloon occlusion test, extracranial-to-intracranial bypass surgery should be considered. Additional surgical options for carotid pseudoaneurysms include clipping the lesion, resection with end-to-end anastomosis, resection with interposition graft [2]. Resection with end-to-end anastomosis, Dacron, or saphenous vein graft is likely the safest open surgical procedure in regard to late thromboembolic morbidity and mortality but is associated with a high perioperative complication rate [15]. Preserving patency of

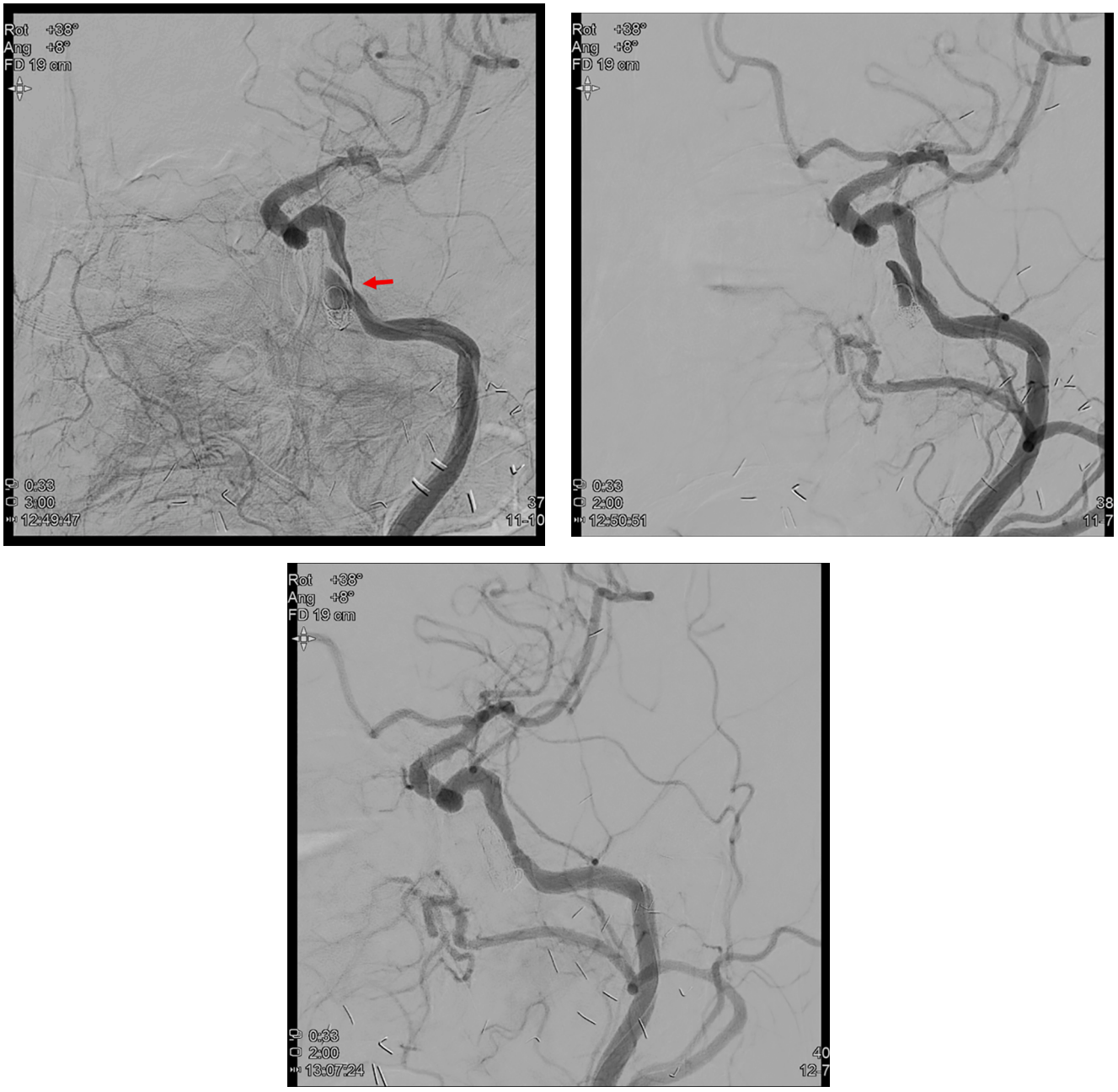

Fig. 3. A. The more coils was filled in aneurysm, the lumen of ICA was more narrowed (red arrow) and blood flow was compromised. B. In suspicious dissecting pseudoaneurysm, Stent was deployed in neck area. Blood flow and lumen of ICA was marked improved immediately. C. Aneurysm was completely occluded in final angiogram. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

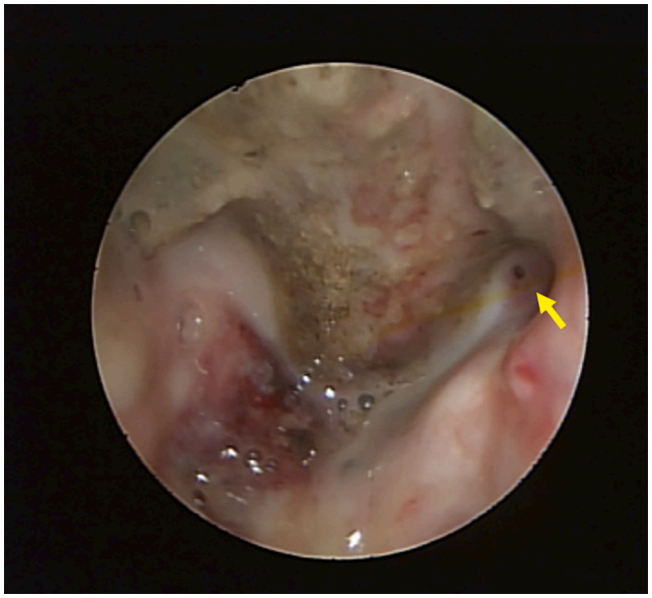

Fig. 4. A few days later, nasal endoscopy was performed. The suspected point of bleeding was confirmed (yellow arrow), but coil was not visible in this area. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

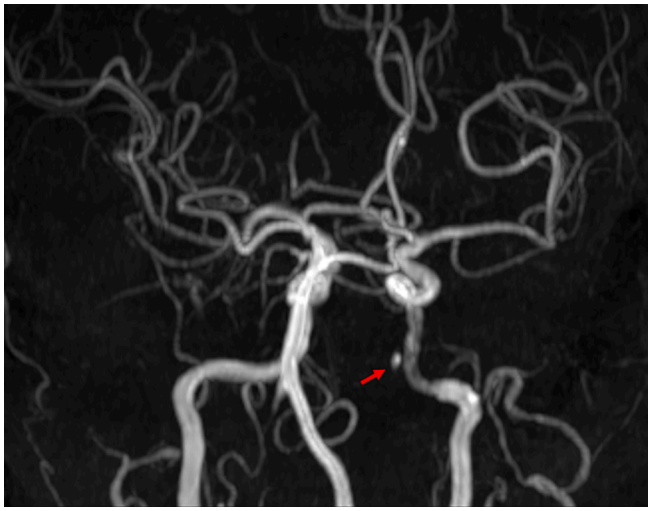

Fig. 5. One weeks later, follow up MRI revealed small flow signal in aneurysmal neck portion (red arrow). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

the carotid artery as far as possible is a more desirable goal. Some reports have revealed cerebral aneurysm formation or growth after permanent carotid occlusion [23,24]. Recently, endovascular treatment for the carotid artery pseudoaneurysms have been increased and performed in several center, and have demonstrated favorable outcomes. Endovascular treatment strategies include vessel sacrifice with detachable balloons or coils, coiling the lesion (with or without stent assistance), or placing a stent across the neck of the pseudoaneurysm. While the ideal goal of treatment is to occlude the aneurysm while preserving the parent vessel, sometimes, it is difficult to achieve this aim even with current endovascular techniques, which are the first modalities of treatment [2,3,18,25,26]. Pseudoaneurysm differs from other aneurysms because there is no true wall. Therefore Coil occlusion of pseudoaneurysms may have high recurrence rates as these lesions can easily expand in the absence of a true wall, and sometimes, the morphology of these lesions may not be suitable for coiling. The surrounding osteonecrotic bone cannot offer enough support to the artery and pseudoaneurysm. Rupture of a pseudoaneurysm connecting to nasal cavity as empty space is different from rupture of an intracranial aneurysm in a closed space. Hemostasis or effective occlusion is likely to be difficult. So, for complete hemostasis, it is important to completely occlude the area where the nasal cavity and the pseudoaneurysm are connected, and check whether or not it is blocked. Although occlusion of pseudoaneurysm was presented in angiogram, we think it is necessary to confirm with nasal endoscopy that leakage opening is completely occluded for avoiding subsequent bleeding after treatment.

Sacrifice of the carotid artery is the last option under such circumstances and can be performed when there is demonstration of adequate collateral circulation via the circle of Willis, as evaluated using the angiogram. There are some reports that an aneurysm was occluded by simply deploying a fenestrated stent across an the aneurysm neck which lead to make flow diversion and to create enough stasis for generation thrombosis within the aneurysm; however, this phenomenon is unreliable and not practical. When urgent hemostasis is required, covered stents (stent grafts) are often used. However, it has more stiffness and poor navigability compared to fenestrated stents, therefore it is still undergoing refinement in design [7,12,24].

In my case, like other intracranial aneurysms, it is thought that compaction or recur may occur later. Possibility of compaction or recur may be higher in this aneurysm compared to intracranial aneurysm. In emergency situation like massive epistaxis, we think it is important to reduce complications and quickly and effectively to complete emergency treatment. If BTO is performed in an emergency situation and ICA occlusion is performed only with only BTO result, the incidence of complications may be higher than our treatment. After resolving emergent situation, I think a number of evaluation and treatment options cab be available considering secondary treatment like ICA occlusion with bypass, flow diverter stent or covered stent in the future. To do so, I propose that it is necessary to confirm that the bleeding point is occlusion.

In Review of the literature, only a small number articles have reported development of ICA pseudoaneurysm after radiation for NPC, and they were case reports and a few small case series. In most studies, endovascular treatment remains the preferred modality of treatment. The study by Lam et al., [15,7] reported that successful temporary control of bleeding and airway are the two statistically significant factors, which predicted survival in this group of patients. Previous study showed that may patients had unfavorable outcome due to failure of bleeding control or complications of treatment. Even in patients who survived the treatment, approximately 16% suffered from severe neurological complications. The complications were mostly ischemic in nature arising due to stent occlusion and thromboembolism [5,12,14,24,15,27,28].

4. Conclusion

Nasal endoscopic localization of bleeding point of a massive upper airway hemorrhage may have limitation and be challenging lesion. If massive epistaxis is occurred in patients with PNC and radiation therapy, the possibility of carotid artery pseudoaneurysm should be considered. The endovascular embolization may be a good method to resolve the problems of ruptured pseudoaneurysm. This procedure is quick, safe, feasible, and less invasive for maintaining vessel patency while effectively occlusion the pseudoaneurysm. In emergent situation like massive epistaxis, we think it is necessary to check complete occlusion with angiographic and nasal endoscopic for avoiding rebleeding. However, longitudinal follow-up and larger series are necessary to evaluate the long-term efficacy of endovascular coiling. After resolving emergent situation, I think a number of evaluation and treatment options cab be available considering secondary treatment in the future with higher safety if necessary. This could be a safer treatment strategy

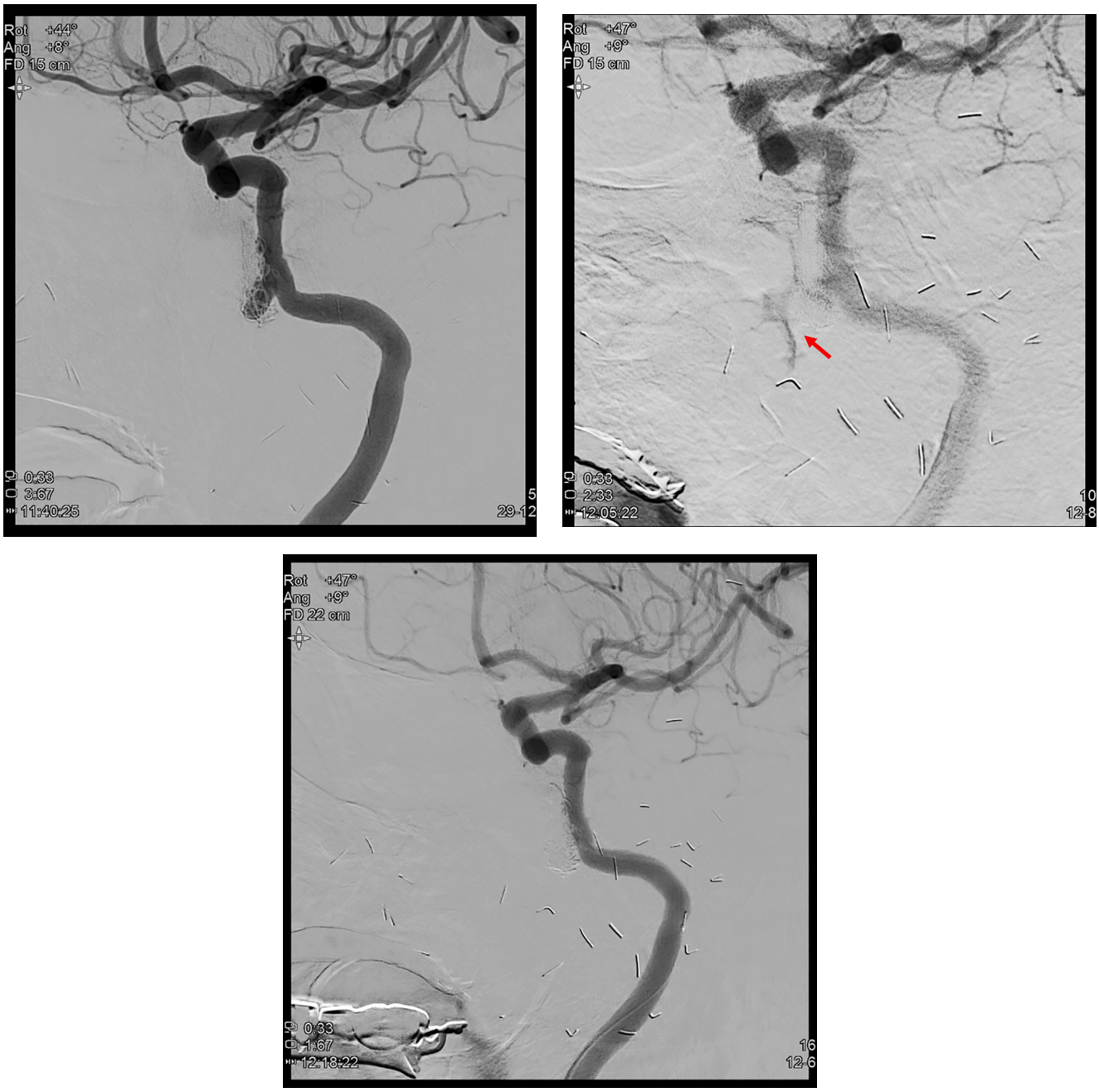

Fig. 6. A and B. In repeated angiogram, more compaction and small leakage (red arrow) from previously small remained area of aneurysm was shown C. Additional coiling was performed in remnant area and complete filing was achieved. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

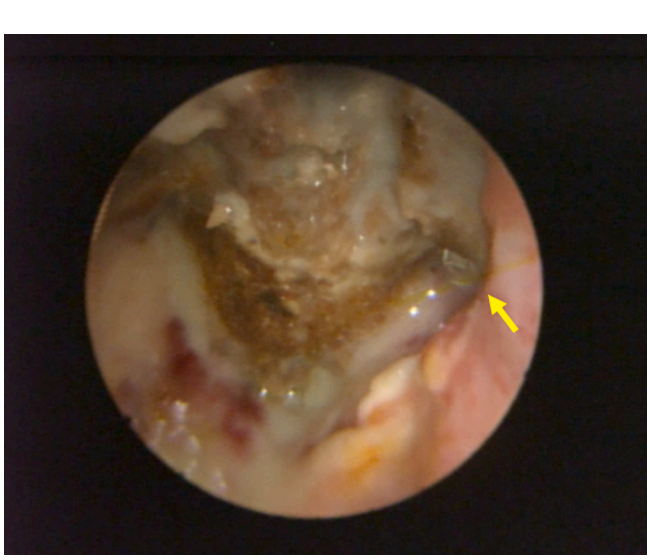

Fig 7. A and B. The leakage area occlusion by coil was confirmed in endoscope of nasal cavity after few days with no more epistaxis (yellow and white arrow). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

with less complication.

5. Support

Nil.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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