CASE REPORT

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To wait for a spontaneous recovery of the third cranial nerve palsy occurring after the coiling of a PComA aneurysm or to implement surgical treatment? – A case report.

Da li treba čekati spontani oporavak slabosti trećeg kranijalnog nerva nastale nakon koilinga PComA aneurizme ili sprovesti operativno lečenje?

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Abstract

Introduction. In the last two decades a method of endovascular embolization has been imposed as a method of choice in the treatment of unruptured intracranial aneurysms. Therefore, the problem of treating posterior communicating artery (PComA) aneurysms presenting with the third cranial nerve (TCN) palsy has become even more complex. The case of a patient reported in the paper itself has presented a dilemma of whether to wait for spontaneous resolution of ophthalmoplegia developed after the coiling of a PComA aneurysm or whether to implement an early surgical treatment. **Case report**. An unruptured saccular aneurysm, directed inferolaterally in the right internal carotid artery (ICA) segment in the position of the PcomA origin, was diagnosed in a 58-year-old male patient. The aneurysm was measuring 9 mm in diameter while the neck was measuring 5 mm. The day before the planned embolization, the patient developed ipsilateral ophthalmoparesis, whereas the first day after the endovascular procedure was completed, the patient developed right-sided complete ophthalmoplegia. Ten weeks after the endovascular embolization our team decided to perform a microsurgical treatment including aneurysm clipping and coil extraction. Eighteen months after the surgery, the patient made a full recovery of the functions of *musculus* (*m) levator palpabrae*, *m. rectus medialis* and pupillary function, with a partial recovery of the functions of *m. obliqus inferior*, *m. rectus inferior* and *m. rectus superior*. **Conclusion**. According to medical research and literature, the partial recovery of the TCN palsy is expected to happen in the first few weeks after embolization. Despite the completion of endovascular treatment progression of ophthalmoparesis to ophthalmoplegia without any simptoms of clinical improvement after 10 weeks is considered to be an indicator of long-standing TCN compression, which can lead to irreversible nerve damage. Despite the increase in the use of an endovascular embolization method in the treatment of PComA aneurysms preceeded by the TCN palsy, neurosurgical treatment is believed to have been necessary. Still, there is one question left to be answered - did we react too late in this particular case?

Key words:
intracranial aneursym; oculomotor nerve; embolization, therapeutic; neurosurgical procedures; ophthalmoplegia; recovery of function.

Apstrakt

Uvod. U poslednje dve decenije metoda endovaskularne embolizacije nametnula se kao metoda izbora u lečenju nerupturiranih intrakranijalnih aneurizmi. Stoga je problem lečenja aneurizmi u regiji zadnje komunikativne arterije (PComA), udružene sa slabošću trećeg kranijalnog nerva (TKN), postao još kompleksniji. Slučaj bolesnika prikazanog u ovom radu stvorio je dilemu da li treba čekati spontani oporavak oftalmoplegije koja je nastala nakon embolizacije aneurizme na PComA ili je potrebno sprovesti rano operativno lečenje? **Prikaz bolesnika**. Kod bolesnika muškog pola životne dobi od 58 godina dijagnostikovana je nerupturirana inferolateralno orijentisana bilobarna aneurizma na desnoj unutrašnjoj karotidnoj arteriji – *arteria carotis interna* (ACI) u regiji ishodišta PComA, dijametra 9 mm i širine vrata 6 mm. Dan pre planirane embolizacije kod bolesnika se javila ipsilateralna oftalmopareza, a prvog dana nakon endovaskularne procedure došlo je do razvoja kompletne desnostrane oftalmoplegije. Nakon 10 nedelja perzistentne oftalmoplegije doneli smo odluku da se sprovede operativno lečenje u vidu

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klipsovanja aneurizme i ekstrakcije koilova. Nakon 18 meseci od operacije došlo je do potpunog oporavka funkcije *musculus* (m.) *levator palpabrae*, *m. rectus medialis* i pupilarne funkcije, sa parcijalnim oporavkom funkcije *m.obliqus inferior*, *m. rectus inferior* i *m.rectus superior*. **Zaključak**. Prema literaturi, očekivano vreme, barem delimičnog oporavka TKN nakon embolizacije je tokom prvih nekoliko nedelja. Progresija oftalmopareze u ofalmoplegiju uprkos sprovedenom endovaskularnom lečenju, a bez kliničkog poboljšanja nakon 10 nedelja od sprovođenja istog, shvaćena je kao indikator izražene kompresije TKN koja bi mogla dovesti do ireverzibilnog oštećenja nerva. Uprkos sve većoj učestalosti metode endovaskularne embolizacije u lečenju aneurizmi u regiji PComA koje su praćene preoperativnom slabošću TKN, smatramo da je operativno lečenje bilo neophodno. Ostaje pitanje da li je naša reakcija u ovom slučaju zakasnila?

Ključne reči:
aneurizma, intrakranijalna; n. oculomotorius, embolizacija, terapijska; neurohirurške procedure; oftalmoplegija; funkcija, povratak.

Introduction

The question of an effective treatment alternative of unrupted intracranial aneurysms poses a particular problem encountered by neurosurgeons, especially nowadays when the detection rate of aneurysms during routine neurodiagnostic examinations has been increased [1]. Particular neurosurgical approaches have always been required for the treatment of posterior communicating artery (PComA) aneurysms. These aneurysms reported to occur in 13–30% of all cases cause the third cranial nerve (TCN) palsy [2]. Therefore, even before the era of 'high definition' neuroimaging, aneurysms was detected prior to the occurence of spontaneous subarachnoid hemorrhage due to aneurysmal rupture. The clinical presentation of unruptured PComA aneurysms includes symptoms associated not only with the TCN palsy, but also with retrobulbar pain [3]. Considering the wide use of endovascular coil embolization to treat unruptured intracranial aneurysms [4], particularly in the last decade, the problem regarding optimum management of unruptured PComA aneurysm became even more complex.

Possible mehanisms for the occurrence of the TCN palsy caused by a PComA aneurysm are the following: aneurysm induced mass effect and consequent compression of neighboring nerve, pulsatile effects of aneurysm and the combination of the latter two mechanisms. Coiling of PComA aneurysms was considered to affect the compression effect on the TCN, and aneurysms in this region were believed to require surgical treatment. However, in the past few years there was an increasing number of reports speaking in favour of complete symptomatic recovery or partial relief of the TCN after coil embolization [5–7]. Also, they refered to the fact that no significant differences were recognized in the clinical outcome observed while following treatment compared with surgical clipping aneurysms [8–10].

The patient's case presented in our paper has initiated a dilemma of whether to wait for a spontaneous recovery of the TCN palsy which progression is observed immediately after endovascular coiling of an unruptured PComA aneurysm. Once more, the reported case put an emphasis upon the complexity of pathology manifested within the case itself along with the emergence of taking a patient-centred approach which should be tailored to each individual. Other possible complications relating to the management of a PComA aneurysm have also been discussed within the paper itself.

Case report

A 58-year-old male patient was admitted to the outpatient clinic of the Department of Neurology because of repeated episodes of right-sided headaches followed by ipsilateral retrobulbar pain. The computed tomography (CT) scan of endocranium revealed no evidence of intracranial hemorrhage or any other pathological lesions. During the further course of examination, magnetic resonance (MR) angiography was performed, complete with the digital subtraction angiography (DSA) study. With 3D DSA of the cerebral vessels, an unruptured aneurysm was recognized (Figure 1A). It revealed a bilobed saccular aneurysm of the right internal cartoid artery (ICA) proximal to the PComA origin, directed inferolaterally. A diameter of the aneurysm sac was 9 mm, with a proximal neck diameter of 6 mm and

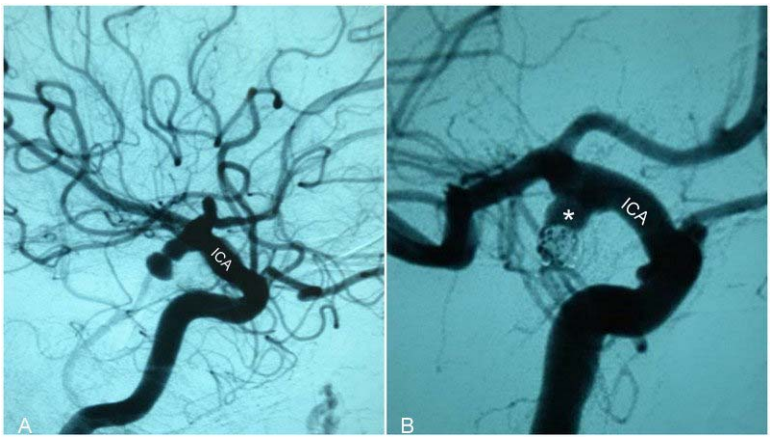

Fig. 1 – Preoperative cerebral digital subtraction angiography (DSA) shows: A) bilobed saccular aneurysm of posterolateral wall of the internal carotid artery (ICA), communicating segment, aneurysmal sac was measuring 9 mm in diameter while the neck was measuring 6 mm in diameter, with a fundus which was directed inferolaterally; B) DSA immediately after coil endovascular embolization shows that approximately 75% of the total aneurysm sac volume was filled (arrow), with a neck residue (asterisk).

a dome-to-neck ratio of 1.5. Endovascular coil embolization was recommended as an initial treatment. It was also planned along additional placement of a stent for the finally recovery of aneurysm.

The day before the scheduled endovascular embolization the patient developed a TCN palsy causing ptosis which was accompanied by lateral deviation in the right eyeball. Although it was followed by ophthalmoparesis, the procedure of endovascular embolization (EE) was administered in order to relieve compression of TCN by relieving pulsating effect of the aneurysm.

The EE was performed and after treating the intracranial wide-necked aneurysm, approximately 75% of the total aneurysm sac volume was filled, which was revealed by the immediate postprocedural DSA images (Figure 1B). However, in the immediate postprocedural course the patient developed right-sided ophthalmoplegia, including drooping of the upper eyelid (ptosis), pupilary dilation (mydriasis) and paralysis of the bulbomotor muscles innervated by TCN.

Eight weeks after procedure there was no regression of neurological deficit. Taking into consideration that it was not possible to completely fill the aneurysm and that in the postprocedural course the patient exhibited the progression to complete ophthalmoplegia, our clinical team decided to perform neurosurgical operative treatment 10 weeks after the endovascular embolization.

After the right-sided pterional craniotomy, due to the level of magnification increased by an operating microscope it was enabled to present the right *nervus (n) opticus* and right ICA in the first place. In a microdissection procedure a wide-necked aneurysm was first identified on the posterolateral wall of the ICA proximal to the PComA origin, with a fundus which was directed inferolaterally, causing compression of the TCN (Figure 2A). *N. oculomotorius* presented a flatter surface, while the fundus caused its slightly elevated prominence and convex outer surface. The two clips were placed across the neck of the aneurysm – a slightly curved clip, measuring 9 mm and the straight one, measuring 8.3 mm (Figure 2B). Then, microdissection of the aneurysm fundus was carried out. Using the microscissors, the fundus was cut sharply and the placed coils were gradually removed (Figure 2C and 2D). The TCN compression was relieved completely, but nerve was bent and arch-shaped and it was made thinner.

In the early postoperative period after surgery the patient awoke with right ophthalmoplegia. Immediate postoperative angiography showed the absence of aneurysm rest (Figure 3).

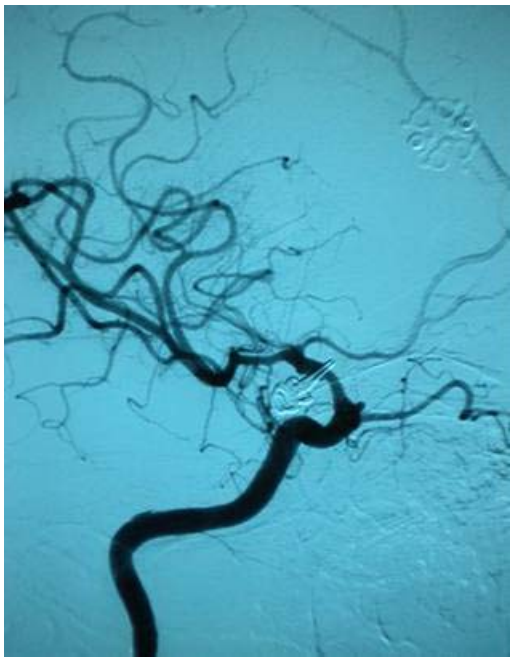

Fig. 3 – Immediate postoperative cerebral digital subtraction angiography shows complete obliteration of the aneurysm.

The postoperative course was uneventful. Examination performed two months after surgery revealed regression rate of neurological deficit. The patient had a full recovery, with resolution of the eyelid ptosis first, which was followed by the recovery of external ocular movement and then improvement in pupillary function. Follow-up data concerning the patient's neurological state were collected during a six-month period. Eighteen months after the surgery the patient had a full

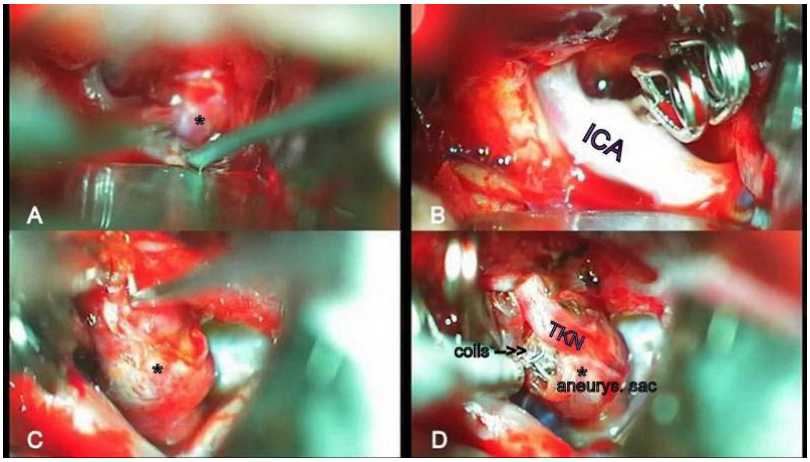

Fig. 2 – Intraoperative images under magnification (× 8) of an operating microscope show: A) microdissection of an aneurysmal sac (asterisk); B) by placing two clips at aneurysmal neck which is at posterolateral wall of internal carotid artery (ICA) followed by the (C) aneurysmal sac opening and (D) removing of coils from aneurysmal sac with third cranial nerve (TKN) decompression.

recovery of the functions of *m. levator palpebrae*, *m. rectus medialis* and pupil function. It also demonstrated that the patient had a partial recovery of the functions of *m. obliquus inferior*, *m. rectus inferior* and *m. rectus superior*.

Discussion

Although it can be stated that the adequate management of unruptured intracranial aneurysms is still one of the most controversial topics [11], unruptured PComA aneurysms, presenting with TCN palsy, require urgent treatment to maximize the potential of functional recovery and prevent subarachnoid hemorrhage [12]. A surgical procedure of aneurysm clipping used to be a method applied in a standard treatment of TCN palsy caused by an expanding PComA aneurysm [13]. Also, the mechanism of recovery by surgical clipping was known to be effective by relieving the mass effect. As the method of endovascular embolization has been more widely used over time, more cases have been reported along with a minor case series of patients who have made a functional recovery of TCN after coiling of an PComA aneurysm [5–7]. Recovery mechanism of TCN following coil embolization is related to loss or decrease of aneurysmal pulsation, despite the fact that the mass effect was not completely relieved. There is also evidence indicating that after embolization the aneurysm volume decreases by 30% within the period lasting from 2 to 12 months [14].

Although some studies suggested that surgical clipping was associated with a higher incidence of recovery of TCN and higher recovery level in comparison with endovascular treatment [15], meta-analysis of all similar available studies showed that there were no statistically significant differences in clinical outcome and rate of complete recovery between the two groups of patients following coil embolization and surgical clipping [9, 10]. The findings suggested that oculomotor nerve palsy may result not only from mechanical compression by coils but also from inflammation induced by perpendicular thrombosis occurring immediately after endosaccular embolization [11].

The prognosis of the TCN palsy mainly depends on the degree of preoperative deficit. Gender, age and size of the aneurysm had no influence on the functional recovery of the nerve [6, 7, 10, 16, 17]. The importance of interval between the onset of palsy and the time of operation/embolisation seems to be contradictory when compared to the recovery of the nerve according to the results obtained by various authors [7, 12]. Patients usually experienced complete functional recovery of TCN within 3 months of surgery/embolization. However, full recovery may also take two years. [6, 7, 17].

Ptosis is generally the first symptom, and it frequently shows the earliest recovery of all other disturbed oculomotor functions after surgery. The restitution of the single ocular muscle functions shows a fairly constant course: the *musculus* (*m) levator palpebrae* and the *m. rectus medialis* show rapid recovery. The parasympathetic fibres follow next, but normal function of elevation and depression of the ocular bulb (*m. rectus sup*., *m. obliquus inf*. and *m. rectus inferior*) is often delayed. The above mentioned clinical course of the TCN recovery in patients after clipping completely correlates with the clinical data of the patient reported here, regarding his/her functional recovery [13].

Endovascular embolization of PComA aneurysms imposes a problem of anatomical specificity of the location of an aneurysm, based on which the following types of aneurysms can be identified: „true PComA aneurysms", aneurysms which arise on the posterolateral wall of the ICA located proximal to the origin of the PComA and ICA communicating segment aneurysms distal to the origin of the PComA [18]. In the first aneurysmal type neck of the aneurysm originate of the PCom artery itself and is often associated with large or fetal PComA arteries. In the most common second type the neck of the aneurysm can partially incorporates the PComA artery [9], which was not present in our reported patient, although the aneurysm neck was closely related to the PComA origin. Although it is a seemingly irrelevant classification of a small segment of blood vessels at the base of the brain, its significance is highlighted in the era of endovascular embolization. As for recommendations given for the first two aforementioned subgroups of aneurysms, microsurgical treatment performed by placing adequately modified clips is being recommended. Endovascular embolization of the mentioned aneurysms is often incomplete due to the efforts invested in order to save the PComA origin. This is followed by aneurysm recurrence, while stent placement may compromise perforant branches. The location of the branches cannot be determined without performing neurosurgical microdissection. ICA communicating segment aneurysms distal to the origin of the PComA can be adequately treated by microsurgical treatment and endovascular embolization [18].

An increasing number of aneurysms demanding operative treatment immediately after the performed procedure of endovascular embolization, has been reported in the past few years. In our viewpoint, it is a result of inadequate diagnostic indications for aneurysm coiling procedures. Based on the literature, the most common indications for an operative treatment of previously coiled aneurysms were the following: incomplete aneurysm occlusion, aneurysmal regrowth and coil herniation [19]. A few case series of patients demonstrated that microsurgical treatment of the given aneurysms was associated with a low incidence of serious complications and favourable clinical outcome in most cases. When reviewing the mass effect of an aneurysm, numerous authors consider utilizing a aneurismal sac evacuation technique to be their first choice regardless of the aneurysm location and nerve deficits it causes [20]. Taking into consideration all the available literature, we have not been able to identify a case of microsurgical treatment of a PComA aneurysm where ophthalmoparesis progressed to acute TCN palsy immediately following the coiling procedure.

Due to all mentioned above, we are facing the following clinical dilemma: to wait for a spontaneous recovery of developed ophtalmoplegia after the coiling of a PcomA aneurysm, or not, and how long we should wait. In addition, is it necessary for patients to undergo operative treatment as soon as possible? Or, to rephrase our question: can our patient's state be led to the point of the irreversible TCN

damage by taking an expectation approach? The literature published so far has not given clear guidelines related to a reasonable time frame to expect the recovery of the nerve after endovascular embolization. Although it has been noticed that the recovery of the TCN palsy can take even a whole year after the embolization was performed, it is all about cases where initial regression of weakness symptoms has appeared in the first few weeks after the intervention [6, 7]. Taking into consideration all the findings revealed up to this point, we believed that concerning our patient's case we had run out of time and that microsurgical intervention was necessary. The progression of ophthalmoparesis to ophthalmoplegia that did not improve within ten weeks, was considered in our opinion to be an indicator of long-standing compression of the TCN. In the following postembolization course, it could lead to the irreversible TCN damage. Having completed further analyses of the clinical course and intraoperative findings, we concluded that the right decision, related to the further patient's treatment was made at the given moment. The decision was made for the purpose of prevention of permanent nerve deficit. Still, one question remains to be answered - did we react in a timely manner? Additionally, is the progression of ophthalmoparesis to ophthalmoplegia after coiling considered to be a early predictor of unfavourable outcome of the recovery of TCN which demands operative treatment in the first few days? All the additional questions that may arise concerning the given topic remain unanswered for the state being due to the lack of reported cases of patients with the clinical course similar to the one presented in our case report.

Conclusion

Despite the increasing frequency of use of endovascular embolization in the treatment of unruptured PComA aneurysms presenting with the TCN palsy, it is our viewpoint that concerning the cases with weakness showing marked progression towards complete ophtalmoplegia after EE is performed – the patients are required to undergo surgery. The surgery is to be performed by placing a clip over the neck of an aneurysm and by the use of the technique of coil extraction in order to decompress the oculomotor nerve. Simultaneously, it is neccessary to take into account the time factor, because the acute complete ophtalmoplegia occuring after the coiling of a PComA aneurysm should be taken as an indicator of a predominantly expressed compression of the TCN. The compression may lead to irreversible nerve damage, resulting in permanent nerve deficit. In the case of the aforementioned clinical course microsurgical procedure is advised, by means of which the aneurysm mass effect could be relieved. Unfortunately, based on our own experience and the given literature, we have to state that nothing can be said about the period of time recommended for the patient to undergo surgery which is considered to be more invasive treatment modality. However, in this case, according to the authors of the paper itself – the utilized modus of treatment was highly necessary.

REFERENCES

1. *Morris Z, Whiteley WN, Longstreth WT Jr, Weber F, Lee YC, Tsushima Y,* et al. Incidental findings on brain magnetic resonance imaging: systematic review and meta-analysis. BMJ 2009; 339: b3016.
2. *Good EF*. Ptosis as the sole manifestation of compression of the oculomotor nerve by an aneurysm of the posterior communicating artery. J Clin Neuroophthalmol 1990; 10(1): 59–61.
3. *Lanzino G, Andreoli A, Tognetti F, Limoni P, Calbucci F, Bortolami R*, et al. Orbital pain and unruptured carotid-posterior communicating artery aneurysms: the role of sensory fibers of the third cranial nerve. Acta Neurochir (Wien) 1993; 120(1−2): 7−11.
4. *Im SH, Han MH, Kwon OK, Kwon BJ, Kim SH, Kim JE,* et al. Endovascular coil embolization of 435 small asymptomatic unruptured intracranial aneurysms: procedural morbidity and patient outcome. AJNR Am J Neuroradiol 2009; 30(1): 79−84.
5. *Gu DQ, Luo B, Zhang X, Long XA, Duan CZ*. Recovery of posterior communicating artery aneurysm-induced oculomotor nerve paresis after endovascular treatment. Clin Neurol Neurosurg 2012; 114(9): 1238−42.
6. *Chalouhi N, Theofanis T, Jabbour P, Dumont AS, Gonzalez LF, Starke RM,* et al. Endovascular treatment of posterior communicating artery aneurysms with oculomotor nerve palsy: clinical outcomes and predictors of nerve recovery. AJNR Am J Neuroradiol 2013; 34(4): 828−32.
7. *Hanse MC, Gerrits MC, van Rooij WJ, Houben MP, Nijssen PC, Sluzewski M.* Recovery of posterior communicating artery aneurysm-induced oculomotor palsy after coiling. AJNR Am J Neuroradiol 2008; 29(5): 988−90.
8. *Nam KH, Choi CH, Lee JI, Ko JG, Lee TH, Lee SW*. Unruptured Intracranial Aneurysms with Oculomotor Nerve Palsy : Clinical Outcome between Surgical Clipping and Coil Embolization. J Korean Neurosurg Soc 2010; 48(2): 109−14.
9. *Golshani K, Ferrell A, Zomorodi A, Smith TP, Britz GW*. A review of the management of posterior communicating artery aneurysms in the modern era. Surg Neurol Int 2010; 1: 88.
10. *Khan SA, Agrawal A, Hailey CE, Smith TP, Gokhale S, Alexander MJ*, et al. Effect of surgical clipping versus endovascular coiling on recovery from oculomotor nerve palsy in patients with posterior communicating artery aneurysms: A retrospective comparative study and meta-analysis. Asian J Neurosurg 2013; 8(3): 117−24.
11. *van Rooij WJ, Sluzewski M.* Procedural morbidity and mortality of elective coil treatment of unruptured intracranial aneurysms. AJNR Am J Neuroradiol 2006; 27(8): 1678−80.
12. *Okawara SH*. Warning signs prior to rupture of an intracranial aneurysm. J Neurosurg. 1973; 38(5): 575−80.
13. *Leivo S, Hernesniemi J, Luukkonen M, Vapalahti M*. Early surgery improves the cure of aneurysm-induced oculomotor palsy. Surg Neurol 1996; 45(5): 430−4.
14. *de Oliveira JG, Borba LA, Rassi-Neto A, de Moura SM, Sanchez-Júnior SL, Rassi MS,* et al.. Intracranial aneurysms presenting with mass effect over the anterior optic pathways: neurosurgical management and outcomes. Neurosurg Focus 2009; 26(5): E3.
15. *Chen PR, Amin-Hanjani S, Albuquerque FC, McDougall C, Zabramski JM, Spetzler RF*. Outcome of oculomotor nerve palsy from posterior communicating artery aneurysms: comparison of clipping and coiling. Neurosurgery 2006; 58(6): 1040−6; discussion 1040−6.
16. *Nishino K, Ito Y, Hasegawa H, Shimbo J, Kikuchi B, Fujii Y*. Development of cranial nerve palsy shortly after endosaccular

embolization for asymptomatic cerebral aneurysm: report of two cases and literature review. Acta Neurochir (Wien) 2009; 151(4): 379−83.

17. *Hamer J.* Prognosis of oculomotor palsy in patients with aneurysms of the posterior communicating artery. Acta Neurochir (Wien) 1982; 66(3−4): 173−85.
18. *He W, Gandhi CD, Quinn J, Karimi R, Prestigiacomo CJ.* True aneurysms of the posterior communicating artery: a systematic review and meta-analysis of individual patient data. World Neurosurg 2011; 75(1): 64−72; discussion 49.
19. *Romani R, Lehto H, Laakso A, Horcajadas A, Kivisaari R, von und zu Fraunberg M,* et al. Microsurgery for previously coiled aneurysms: experience with 81 patients. Neurosurgery 2011; 68(1): 140−53; discussion 153−4.
20. *Waldron JS, Halbach VV, Lawton MT.* Microsurgical management of ncompletely coiled and recurrent aneurysms: trends, techniques, and observations on coil extrusion. Neurosurgery 2009; 64(5 Suppl 2): 301−15; discussion 315−7.

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