Unexpected Perioperative Complication of Aneurysm Surgery: Armored Arachnoiditis Case Report

Anevrizma Cerrahisinin Perioperatif Beklenmedik Komplikasyonu: Zırhlı Araknoidit, Olgu Sunumu

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ABSTRACT
Numerous problems have been described during the aneurysm surgery. In this report, severe arachnoidal scarring (that is named as armored arachnoiditis) that made clipping of anterior communicating (ACom) aneurysm impossible in our case of study is presented.

A case with recurrent hemorrhage is hospitalized in our institution. Initial diagnosis was meningitis in a local hospital. The patient was operated after recurrent intraventricular hemorrhage in late phase due to unavailability of endovascular options at our institution. At surgery, surprisingly all of the cisterns were obliterated and no cleavage plane was found. Every effort for taking CSF was unsuccessful. The corticectomy of gyrus rectus did not solve the problem. Neither aneurysm nor proximal and distal controls could be exposed in spite of any dissection techniques used. We felt that any further attempts to dissect aneurysm and ACom complex would result in a catastrophic injury and rupture. Since, the armored arachnoiditis could not be detected before the surgery, patients should be treated by endovascular techniques following the surgery in the presence of this type of arachnoiditis. In this report, the pertinent literature related to perioperative complications of aneurysm surgery is shortly reviewed.

Key words: Aneurysm, arachnoiditis, surgery, complication, subarachnoid hemorrhage

ÖZET
Anevrizma cerrahisi esnasında çeşitli sorunlar tanımlanmıştır. Bu yazıda, anterior komünikan (ACom) anevrizmasının kapatılamaması olanaksız ve zırhlı araknoitid olarak isimlendirilen ciddi cisternal yapışıklığı olan bir olgu sunulmaktadır. Başka bir merkezden menenjit ön tanısı ile gönderilen tekrarlayıcı kanamalı bir olgu kliniğimize yatırıldı. Olgu, hastanemizde endovasküler tedavi seçenekleri olmadığından tekrarlayıcı intraventriküler kanama sonrası geç dönemde ameliyat edildi. Cerrahisi esnasında, sürpriz bir şekilde tüm sistemler kapalı idi ve klivaj kaybolmuştu. BOS boşaltmaya yönelik tüm çabalar başarısızdı. Girus rektusa uygulanan korticektomi sorunu çözmedi. The corticectomy of gyrus rectus did not solve the problem. Neither aneurysm nor proximal and distal controls could be exposed in spite of any dissection techniques used. We felt that any further attempts to dissect aneurysm and ACom complex would result in a catastrophic injury and rupture. Zırhlı araknoidit ameliyattan önce anlaşılamayacaktır, bu tıpte bir araknoiditte cerrahi sonrası endovasküler yöntemler kullanılmalıdır. Zırhlı araknoiditli bir olgu sununsunda, anevrizma cerrahisinin perioperatif komplikasyonları ilgili literatür taraması ile kısaca gözden geçirilmiştir. (Nöropsikiyatri Arşivi 2011; 48: 207-10)

Anahtar kelimeler: Anevrizma, araknoidit, cerrahi, kompleksiyon, subaraknoid kanama

Introduction
Surgical problems during aneurysms surgery can be seen in the early, mid and/or in the late phase of surgery and roughly classified as issues related to position (preincisional stage), during the craniotomy (incisional stage), and lastly, after the craniotomy (postincisional stage). Majority of problems in the beginning of the surgery (preincisional stage) and during the craniotomy (incisional stage) can be related to intubation and anesthesia delivery systems (1). In this situation, decreased oxygen saturation may lead to increased intracranial pressure and neurological events including (re)rupture of the aneurysm. Inadvertent waking up of the patient may cause sensing of pain and swelling of the brain and/or rebleeding. After craniotomy, problems are seen at predissection, during dissection or during clip application. Dissection of cisterns can be difficult in edematous and fragile brain tissue. At the same time, tendency of laceration or hemorrhage of red and angry brain may occur. There is an increased risk of rebleeding especially in the early phase of subarachnoid hemorrhage (SAH) due to retraction of tough brain. Problems will grow in the case of difficult and/or deep location and/or large size of the aneurysm. Extensive

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removal of the sphenoid ridge, hyperventilation, gradual withdrawal of CSF, and reduction of mean arterial pressure will minimize the rate of rebleeding. Sharp dissection should be used in exposing the aneurysm as well as removing clot from around it to reduce the risk of large tearing in its fundus or neck. Problems related to clip application are the use of inappropriate clip; clip failure; presence of perforating arteries in the clip’s leg; an aneurysmal remnant after clipping; slipping of the clip; fracture of atheromatous plaque; emboliform processes from atheromatous or thrombotic aneurysm; perforation of aneurysm; shrinkage, stenosis or kinking of parent artery and (manuplational) vasospasm after clip application. The aneurysm should be mobilized and inspected before the clip application, and a neck should be made by bipolar coagulation, if necessary. Surgical approach to the unruptured aneurysm first, can cause rebleeding of the ruptured one in the case of multiple aneurysms. The majority of perioperative difficulties are especially seen on the cases of early surgery. All complications would have high risk for mortality and morbidity when they occur before clipping.

Case Report

A 63-year-old man suffering from head and neck pain was admitted to a local hospital. The patient was hospitalized and followed up with suspicious diagnosis of meningitis. He was transferred to our hospital 15 days after his complaint began. GCS was 14 (WFNS grade III) when he was admitted to our hospital. CT scan showed triventricular hydrocephalus and no sign of hemorrhage (Figure 1). Lumbar puncture was performed and findings consistent with subarachnoid hemorrhage were revealed. He had a new bleeding attack 2 days after his hospitalization, before an angiography was performed. GCS score was dropped suddenly to 11 and patient was accepted as WFNS grade IV. Computerized tomography was repeated and intraventricular fresh hemorrhage was found in occipital horns but not in subarachnoid cisterns (Figure 2). External ventricular drainage (EVD) was performed to reduce intracranial pressure and bloody CSF was drawn intermittently to impede probable hydrocephalus. Daily CSF drainage amount varied between 90–300 cc (mean: 128.5 cc/day). But, the clinical status of the patient got worsens day by day and as a result mechanical ventilation was performed. He had no hematological abnormality. CT findings and persistent bloody CSF led us to believe that the patient had an aneurysm at anterior communicating artery (AComA). Further, the evidence pointed to the aneurysm directly projected and embedded to the third ventricle by lamina terminalis cistern. CT-angiogram performed 7th day of hospitalization for repeated fresh bleeding showed bilobulated AComA aneurysm that project superiorly towards to third ventricle (Figure 3). A nipple was present on the larger lobule. Microbiological follow up of CSF did not reveal infection, but patient had respiratory and urinary infections during this period of time in intensive care unit. Intracranial pressure was controlled and normalized by EVD before surgery. Surgical clipping was decided to treat aneurysm due to unavailability of either angiography, or endovascular options in our institution and also the risk of patient transfer. The patient with same GCS score of 11 was taken to operating room in 16th day of hospitalization and in second week of the second hemorrhage. During the surgery,
we expected to observe clear subarachnoid cisterns due to purely intraventricular hemorrhage as well as the late phase of bleeding. At surgery, surprisingly sylvian and all of the other cisterns were obliterated. Excessive adherence and solid clot made it difficult even impossible to dissect all cisterns. Every effort for taking CSF from cisterns including sylvian, carotid, chiasmatic were unsuccessful. There was no cleavage plane around the vessels and only superior part (in surgical plane) of right M1 and A1 was visible. Not only proximal cisterns (sylvian, carotid and chiasmatic) but all of the other cisterns could not be opened. Commonly simple and short procedures for any experienced surgeon were impossible and time consuming. The same problems mentioned above were also persistent around the aneurysm. We employed the corticectomy of gyrus rectus to no avail though it allowed us to see only the superior part of the right A1, A2 and Heubner arteries. Initial part of the AComA could also be exposed, but neither the starting point of the aneurysm neck, nor any perforating branches of AComA, A2’s or opposite A1 could be seen in spite of any dissection techniques used including sharp dissection or water dissection techniques (Figure 4). Instillation of saline by a vessel catheter to the mentioned cisterns to separate, cut and pass of them was unsuccessful. We felt that any further attempts to dissect aneurysm and ACom complex would result in a catastrophic rupture and bleeding, injury to perforating branches such as hypothalamic or Heubner arteries. We also believed that bleeding would not be controlled because of absence of exposure of aneurysm and proximal and distal controls. Surgical attempts to treat aneurysm were ended for this risky condition and endovascular route was planned. The appearance of the all surgical area resembled to an undissectable brain tumor. Endovascular coiling was carried out successfully 25 days after the surgery (Figure 5). The patient is still on a follow up with GCS score of 9 after 28 months of initial bleeding.

Figure 4. Severe (armored) arachnoiditis was shown with unmarked (a) and marked (b) intraoperative pictures and also on graphical drawing (c) that is not overcame in spite of corticectomy of the gyrus rectus on right side. (RA1 and RA2 = Right A1 and A2)

Figure 5. Digital subtraction angiogram during endovascular coiling after 25 days of surgery. Oblique subtracted (a) and nonsubtracted (b), and AP nonsubtracted (c) cuts show the shape of the aneurysm and suspended contrast media (c) after coiling

Discussion

Surgical treatment of aneurysm is a challenging procedure especially in older patients, and in patients with poor WFNS grade, as well as in the early surgery. Higher WFNS grade is related to severe neurodamage, vasospasm, hydrocephalus and other systemic diseases. Surgical treatment may add comorbidly or mortality in poor grade patient. The majority of physicians will prefer endovascular techniques in spite of surgery in risky patient. Endovascular treatment should be used, if possible.

One of the perioperative problems in aneurysm surgery is cisternal adherence due to glue effect of haemorrhage. Dense arachnoidal scarring could not be seen in all SAH patients and its exact incidence is not known. The arachnoidal obliteration may be seen in elderly and obese patients. The presence of solid form of clot makes surgery difficult and sometimes impossible. This condition is described as ‘cisternal=leptomeningeal=arachnoidal scarring’, ‘cisternal or sulcal obliteration’, ‘meningeal fibrosis’, ‘fibrosing arachnoiditis’ in the literature (2-4). Severe adherence is related to age, poor neurological status, vasoospasm, presence of acute hydrocephalus, CSF infections, necessity of EVD, rebleeding and most importantly intraventricular hemorrhage (IVH) (3). The most frequent reason of severe adherence is IVH. Besides, it can be seen after surgery and cottonoids as a foreign body reaction that can be left during surgery. Further, antiinflammatory response to antigenic material that is released from a cyst of intraventricular or subarachnoid neurocysticercosis is another cause of this condition (5-8). The other common causes of severe adherence are decreased serum T lymphocytes and increased serum IgA, IgG and P proteins, and CSF proteins; local (rhinitis, sinusitis) or systemic (tuberculosis) infections; Chiari malformation and syringomyelia; and leptomeningeal metastasis (5-11). Optochiasmal arachnoiditis (OCA) is well known complication of AComA cases. But, this kind of adherence is seen locally, especially in lamina terminalis and chiasmatic cistern. Since the extant literature does not present any terminology to describe this condition in the SAH patients, we name it “Armed Arachnoiditis”. Armor was used as a cloth to protect warriors in Medieval period in all over world. These clothes were worn by warriors and covered by helmet, shield and other personal handmade guns in addition to clothes at the theatre of war. In that time, any gun such as sword, arrow, mace, etc. couldn’t penetrate these clothes. Problems in the theatre of war were similar to our problems in the operating theatre. Any surgical instruments and techniques didn’t solve the severe scarring in our case. These similarities were taken to give the name of ‘armored arachnoiditis’ to explain this situation. Arachnoidal membranes were clad armor and arachnoidal barriers were similar to armored warriors. In our case, we speculated IVH and EVD as the potential causes of armored arachnoiditis. There were no CSF infection findings. Admittance of patient at a late phase (15 days later of initial SAH) might have hidden the findings of cisternal bleeding. Intermittent external drainage of some amount of daily produced CSF may cause obliteration and adherence of all...
cisterns. CSF may flow to cortical cisterns and sagittal sinus directly with bypassing blocked basal cisterns.

Our literature review reveals that cytokines play a significant role in armored arachnoiditis. Different subgroups of Transforming Growth Factor Beta (TGF-α, -β1, -β2, -β3) are well known cytokines (2,12-15). They are present in thrombocytes and naturally in clot and are released after IVH. Stimulatory effect of cytokines to fibroblast causes the production of extracellular matrix proteins such as fibronectin, laminin and vitronectin (2). These proteins show a glue effect in cisterns and cause cisternal blockage and hydrocephalus (13). Well known characteristics of extracellular matrix proteins are stimulation of production and storage of collagens (especially type I and III collagens that make fibrosis, and procollagen peptids of collagens); acceleration of fibrinosis and wound improvement; influence of development of fibrotic disease such as hepatic and pulmonary fibrosis, glomerulonephritis, fibrosing alveolitis and diabetic neuropathy (2,4). Clinical and experimental researches have revealed a low level of plasminogen which inverts endogenous fibrinosis. On the other hand, a high level of plasminogen activator inhibitor-1 (PAI-1) that impedes plasminogen activators causes inadequate fibrinolysis together (16,17). The importance of other proinflammatory cytokines such as tumor necrosis factor (TNF), Interleukin-1, -6, -8 and interferon in neuronal damage have been reported in patients with IVH (12). Fibrotic complication stimulated by TGF-β1 and interferon in neuronal damage have been reported in patients with IVH (12). Fibrotic complication stimulated by TGF-β1, can be reversed by some methods. Early and rapid removal of cisternal blood with drainage and irrigation, recombinant tissue plasminogen activator, urokinase and streptokinase can be used separately or together for this purpose (18).

Contrast enhancement of related region in CT or MRI can be the diagnostic method in severe arachnoiditis like in the OCA that is local form of adherence (9,11,19). Confirmation of cisternal adherence by cisternography using contrast or radioisotope media in suspicious cases before surgery will guide the surgeon in choosing the treatment options and certainly help him/her prevent this complication preoperatively.

Initial CT scans have revealed only hydrocephalus in our case. Subarachnoidal hemorrhage was confirmed by lumbar puncture. The reason of absence of any sign of cisternal bleeding undoubtedly consumed time. It was not possible to conclude that initial bleeding was subarachnoidal or intraventricular. However, it is well known and demonstrated that second attack was to intraventricular space. First speculation for presented case is cisternal blockage that was developed after the initial bleeding. Severe fibrotic arachnoiditis caused fixation of aneurysm to the surrounding tissues and hemorrhage to intraventricular system afterward. This type of arachnoiditis can be named as primary or spontaneous arachnoiditis. Second speculation is cisternal blockage that was developed after EVD. Externalization of daily produced CSF might have caused stopping of circulation at cisterns. Previous subarachnoidal hemorrhage may have resulted in a glue effect and severe arachnoiditis at these dried spaces. This type of arachnoiditis can be named as secondary or postdrainage arachnoiditis. Grading may also be possible for any type of arachnoiditis. If a grading will be used in the future, armored type of adherence would be the worst form of it.

Conclusion

Armored arachnoiditis or cisternal lost is a rare and unpredictable problem that can be faced with during surgery as we have confirmed in our case. Possible harmful effects of EVD and TGF that is used to prevent hydrocephalus must be kept in mind in this situation as they might be the reasons of blockage of normal CSF circulation and adherence at the cisterns. Contrast enhanced CT or MR imaging may reveal arachnoidal adherences preoperatively. Radiological procedures should be followed to expose this tough problem. Endovascular approaches should be chosen if such complication is recognized pre- or peroperatively in an aneurysm surgery.

References