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Prophylactic measures during oral surgery in patients with Moyamoya

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ABSTRACT

Introduction: Moyamoya disease is an uncommon cerebrovascular condition in Western populations; therefore, it is not well known. Patients with Moyamoya require preventative measures when undergoing oral surgery. *PRESENTATION OF CASE*: A 15-year-old girl with Moyamoya disease was referred to our department of Oral and Maxillofacial Surgery for removal of a lateral incisor and four impacted wisdom teeth. An empathic approach, a low-stress environment, conscious sedation and adequate local anesthesia were provided during surgery. The procedure was uneventful.
Discussion: In patients with Moyamoya, it is imperative not to cause a variations in cerebral blood flow (eg by hyperventilation), since this might lead to ischemic attacks or cerebral hemorrhage with detrimental consequences. Maintenance of blood pressure is achieved by providing a tranquile environment and adequate patient information. Furthermore, the use of conscious sedation is advised. Use of adequate local anesthetic is important. Regardless of the concentration of epinephrine, there is no significant impact of the local anesthetic on blood pressure, as long as the maximum dose is not surpassed. Anti-platelet therapy can be continued safely without a significant increase in post-operative bleeding. There are no indications for antibiotic profylaxis in patients with this disease.
Conclusion: Oral surgery in patients with Moyamoya can be performed safely. Local anesthesia and post-operative analgesia should be adequate to prevent complications during and after surgery. The use of intravenous sedation in a low-stress environment is advised.

1. Introduction

Moyamoya disease was introduced as an entity in the 1950's. It is a rare condition that involves a narrowing and/or blockage of intracranial arteries. Predominantly, the internal carotid artery and its branches are affected. In response to the relatively low blood flow, collaterals are formed, but the walls of these vessels are thin and prone to bleeding. Cerebrovascular incidents, such as ischemic events or intracranial hemorrhage are very common. Preventative measures consist of antiplatelet therapy and avoidance of significant fluctuations in blood pressure or hyperventilation.

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Here, we describe a patient with Moyamoya that required removal of a lateral incisor and impacted third molars in the upper and lower jaws.

2. Presentation of case

A 15-year-old girl (a dichorionic/diamniotic twin), with a height of 1.58 m and a weight of 44.4 kg, was referred to the department of Oral and Maxillofacial Surgery by the treating orthodontist. She required the removal of the right upper lateral incisor (due to agenesis of the left incisor) and four fully impacted wisdom teeth (Fig. 1). Her medical history revealed hemiparesis on the right-hand side and left-sided facial nerve palsy, which developed at the age of 13, after a period of persistent, progressive headaches. This event led to subsequent investigations and revealed the diagnosis of Moyamoya disease, albeit with an atypical cerebral angiograph (Fig. 2). Based on a review of anomalies in the angiograph combined with past symptoms, the treating Neurology team estimated that the girl had experienced no less than 24 transient ischemic attacks before diagnosis. Secondary prevention consisted of acetylsalicylic acid 160 mg once daily.

The oral surgery was carried out under conscious intravenous sedation with 4 mg midazolam and 0.06 mg fentanyl. The patient was non-invasively monitored with blood pressure measurements, an oxygen saturation monitor and capnography. A nasal cannula provided additional oxygen. During the procedure, we also administered 80 mg methylprednisolone and 20 mg ketorolac to minimize post-operative swelling and pain. After sedation was induced, local anesthesia was administered with a vasoconstrictor (articaine hydrochloride 40 mg/ml + adrenaline tartrate 10 μg/ml). The right upper lateral incisor was readily removed with forceps. Two impacted molars were removed by raising a local mucoperiosteal flap, performing limited bone trepanation, and splitting the lower molar. Gelatin foam sponges were applied, and the wounds were sutured with resorbable polyglactin material. The procedure went well, without patient anxiety, and the blood pressure, oxygen saturation, and CO2 levels remained stable. The parents, fearing post-operative bleeding, demanded that we only removed 2 wisdom teeth in the first treatment. A second appointment was made for removing the last two teeth with a procedure similar to that performed for the first treatment.

All wound healing was uneventful, with only limited pain and swelling post-operatively.

3. Discussion

Moyamoya disease is a chronic cerebrovascular occlusive disease with a largely unknown pathogenesis. The pathogenesis involves a combination of genetic and acquired conditions; indeed, in this case, the patient's twin sister did not have Moyamoya. The auto-immune system might also play a role in Moyamoya [1]. The disorder causes unique, very localized, bilateral stenosis of the internal carotid arteries. This stenosis leads to relative ischemia in distal regions, which causes the formation of collaterals. However, the collateral vessels are quite brittle, due to a fragmented elastic lamina, which makes them prone to arterial bleeding. The image on digital subtraction angiography showed the typical 'puff of smoke' sign (moyamoya in Japanese). Patients with Moyamoya initially present with transient ischemic attacks or ischemic strokes. Other possible symptoms include headache, seizures, or hemiparesis.

Fig. 1. Panoramic radiograph.

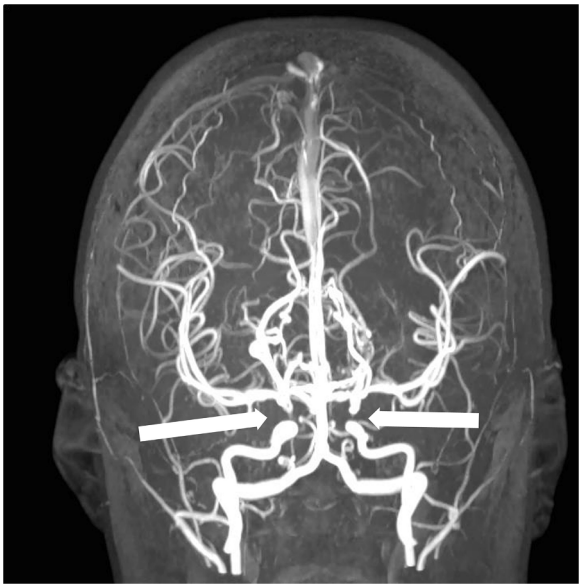

Fig. 2. MRI, TOF sequence. Note the severe stenosis bilaterally, within the internal carotid arteries (arrows).

Moyamoya disease is distinct from Moyamoya syndrome, which is also described in literature. Moyamoya disease is the term used when the arteriopathy is idiopathic; Moyamoya syndrome is the term used when the arteriopathy is secondary to an associated condition, like atherosclerosis, meningitis, autoimmune disease, cranial radiation, or genetic syndromes, such as neurofibromatosis type 1, trisomy 21, or Alagille syndrome [2]. Moyamoya disease is most common in children and young adults, whereas Moyamoya syndrome is more common in older patients. Moyamoya disease is characterized by bilateral symmetry, and Moyamoya syndrome can occur unilaterally.

Epidemiologically, Moyamoya disease shows an obvious racial preponderance; it is mostly observed in Asian races, particularly in Japanese populations. The incidence of Moyamoya disease in Japan is somewhere between 0.35 and 11/100,000 inhabitants. Unfortunately, the incidence of Moyamoya in European nations is unclear. Globally, Moyamoya disease has shown a slightly higher prevalence among males than among females [2,3].

Due to its low prevalence in European populations, Moyamoya disease can be difficult to diagnose. Multiple imaging features are at the disposal of the specialized clinician, but the diagnosis rests mainly on clinical symptoms and angiographic findings [4].

Currently, there is no known cure for Moyamoya disease. Antiplatelet agents are generally prescribed to improve microcirculation and prevent embolisms [1,5]. In some patients, neurosurgical revascularization may be performed, although the indications for patient selection and the choice of neurovascular technique are not clear. The preferred surgical treatment option is either direct bypass surgery or indirect bypass surgery. In direct bypass surgery, the superficial temporal artery is anastomosed with the middle cerebral artery. The most frequently performed indirect bypass surgery is an encephalo-duro-arterio-synangiosis (EDAS). In contrast to the direct bypass technique, where the superficial temporal artery is harvested, in the EDAS technique, the superficial temporal artery is not harvested; instead, it is placed in contact with the brain surface, which results in angiogenesis. Evidence is scarce, but the frequency of ischemic events tends to decline after these types of surgery. There are no clear data are available on associated complications [4].

Patients with Moyamoya disease have tenuous cerebral perfusion. They are at dual risk of ischemia, with hypotension, and cerebral hemorrhage, with hypertensive surges. Tight hemodynamic control is essential, and pre-operative blood pressure levels should be targeted. Adequate cerebral blood flow and cerebral perfusion pressure should be maintained at all times to avoid ischemic complications [6]. Additionally, as a preventative measure, it is important to avoid hyperventilation associated with crying secondary to pain. A high respiratory rate will diminish CO2 blood levels (hypocapnea), which causes cerebral hypoperfusion and could lead to ischemic stroke. Hypercapnea might also be deleterious in the setting of Moyamoya. Dehydration presents a danger and should be avoided, particularly post-operatively.

As described by Seto et al. [5], localized oral surgery may be performed with little risk of complication, when the correct preventative measures are followed. The aforementioned peaceful environment is particularly important in children that are easily frightened. This notion must be kept in mind, even when performing small medical acts, like placing intravenous lines [7]. Local anesthetics that contain epinephrine at doses adapted for children (i.e., 1:80,000 to 1:200,000) will not significantly influence blood pressure. Moore and coauthors [8] found no differences in cardiovascular profiles between children treated with 4% articaine and 1:100,000 or 1:200,000 epinephrine, when using a one-cartridge volume. However, with large volumes (nearly seven cartridges, which is the maximum allowable dose of 4% articaine), Hersh and coauthors [9] reported that the 1: 100,000 epinephrine dose produced significantly higher heart rates and systolic blood pressure at 10 min after injection. Post-operative pain management can be maintained at an adequate level by following the WHO guidelines for the analgesic ladder [10].

Moyamoya alone does not constitute an indication for prophylactic antibiotics.

4. Conclusion

This study demonstrated that oral surgery in a patient with Moyamoya disease can be performed safely, when the oral surgery team is aware of the risks of hyperventilation and high or low blood pressure. Hyperventilation and large variations in blood pressure could cause a cerebral ischemic attack or intracranial hemorrhage. The treatment environment should be calm and tranquil. When performing oral surgery, it is advisable to use intravenous conscious sedation, combined with adequate local anesthesia, with or without epinephrine.

Conflicts of interest

None.

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None.

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