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Case Report

A case of disseminated intravascular coagulation after two-jaw surgery

Arisa Yasuda, Hitoshi Sato*, Takanobu Inada, Yuzo Abe, Motohiro Tanaka, Mai Kurihara, Tatsuo Shiota

Department of Oral and Maxillofacial Surgery, School of Dentistry, Showa University, Tokyo, Japan

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ABSTRACT

Disseminated intravascular coagulation (DIC) is a serious condition that causes persistent widespread coagulation activation in the presence of underlying disease. Most patients who undergo orthognathic surgery are young and have no underlying disease. Therefore, serious postoperative complications such as DIC rarely arise. We describe a patient with DIC that developed after two-jaw surgery in the absence of underlying disease. A 31-year-old man was referred to our hospital with a chief complaint of an occlusal abnormality. He had no underlying disease and his preoperative laboratory findings were unremarkable. The patient provided presurgical, written, informed consent to orthognathic surgery including Le Fort I osteotomy and bilateral sagittal split ramus osteotomy. Perioperative findings and events were also unremarkable. The surgical duration was 4 h and 31 min. Blood loss was 435 mL and he was transfused with 400 mL of hemodiluted autologous blood. The patient developed obvious facial swelling and ecchymosis from the infraorbital area to the cheek in both sides of the face on postoperative day (POD) 5. Blood findings on POD 6 were as follows: RBC, $365 \times 10^4/\mu\text{L}$; WBC, $9,200/\mu\text{L}$; PLT, $0.3 \times 10^4/\mu\text{L}$; FDP, $21.25 \mu\text{g/mL}$; Fibrinogen, 501 mg/dL ; and D-dimer, $10.67 \mu\text{g/mL}$. Treatment for early symptomatic improvement was started, since these findings met several criteria for diagnosis of DIC. Blood parameters improved (PLT, $6.5 \times 10^4/\mu\text{L}$; FDP, $11.05 \mu\text{g/mL}$; D-dimer, $5.24 \mu\text{g/mL}$) one day after the intravenous administration of prednisolone (60 mg) and transfusion with 10 units of platelet concentrate. Thereafter, the clinical course was uneventful.

1. Introduction

Disseminated intravascular coagulation (DIC) is a serious condition which, in the presence of underlying disease, causes persistent and widespread coagulation activation and frequent formation of microthrombi in small blood vessels [1–6]. Relatively common causes of DIC include sepsis, surgery, major trauma, cancer, and complications of pregnancy. It can arise in the oral and maxillofacial region after head trauma, infection, metastatic disease, and surgery [7–9]. Most patients who undergo orthognathic surgery are young and have no underlying disease. Therefore, serious postoperative complications such as DIC rarely arise. We describe a patient without underlying disease who developed DIC after two-jaw surgery.

2. Case report

A 31-year-old Japanese man (height, 170 cm; weight, 68.0 kg) with a chief complaint of an occlusal abnormality began treatment in the orthodontic department at our institution in September 2011. His personal and family medical histories were unremarkable. We diagnosed

facial asymmetry with class III malocclusion accompanied by skeletal mandibular prognathism (Table 1, Fig. 1A and B). Frontal cephalometric X-rays showed the left upper and lower first molars positioned about 2.5 mm lower than those on the right, and the occlusal plane inclined upwards, toward the right. Thus, orthognathic surgery, including Le Fort I osteotomy and bilateral sagittal split ramus osteotomy, was scheduled for November 2013 after the patient completed pre-surgical orthodontic treatment. His nutritional status was favorable, and laboratory findings at three weeks before surgery revealed no abnormal values (Table 2) or abnormal findings in either panoramic dental radiographs (Fig. 2) or the intraoral mucosa (Fig. 3). Le Fort I osteotomy and bilateral sagittal split ramus osteotomy proceeded under general anesthesia in November 2013. The left first molar region was elevated 2.5 mm and the maxilla was rotated making the right first molar as a center. The mandible was repositioned posteriorly (right and left, 5.0 and 2.5 mm, respectively). Interfering bone around the descending palatine artery and maxillary tuberosity was carefully removed with a round bar and a bone rongeur. The volume of intraoperative blood loss was 435 mL, the surgical duration was 4 h 31 min, and the patient was transfused with 400 mL of hemodilutional autologous

* Corresponding author at: Department of Oral and Maxillofacial Surgery, School of Dentistry, Showa University, 2-1-1 Kitasenzoku, Ota-ku, Tokyo, 145-8515, Japan.
E-mail address: h.sato@dent.showa-u.ac.jp (H. Sato).

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Table 1
Preoperative cephalometric analysis of lateral X-rays.

| | Mean | SD | Data |
|---------------------------------|--------|------|--------|
| SNA (degree) | 81.82 | 3.09 | 81.56 |
| SNB (degree) | 78.61 | 3.14 | 82.82 |
| ANB (degree) | 3.28 | 2.66 | -1.26 |
| Facial angle (degree) | 85.07 | 5.76 | 92.17 |
| Angle of convexity (degree) | 5.6 | 4.33 | -4.49 |
| Mandibular plane angle (degree) | 26.25 | 6.34 | 28.15 |
| Ramus inclination (degree) | 87.36 | 4.14 | 83.71 |
| S'-PTM' (mm) | 19.77 | 3.04 | 19.62 |
| A'-PTM' (mm) | 51.02 | 2.62 | 45.53 |
| Gn-Cd (mm) | 128.52 | 4.39 | 129.50 |
| Pog'-Go (mm) | 82.05 | 3.76 | 84.73 |
| Cd-Go (mm) | 69.62 | 4.89 | 64.42 |

Abbreviations: A, A point; ANB, A point nasion B point; Cd, condylo; Gn, gnathion; Go, gonion; Pog, pogonion; PTM, pterygomaxillary fissure; S, sella; SD, standard deviation; SNA, sella nasion A point; SNB, sella nasion B point.

* Scores beyond the range of 1 SD.

blood. Osteosynthesis (Fig. 4) proceeded using titanium plates (AO compact MF system; Johnson & Johnson K.K., Tokyo, Japan). No adverse events such as nosebleeds and ecchymosis occurred at the completion of surgery, and hemorrhage was not evident on POD 1. Therefore, intermaxillary fixation was applied until POD 5 (Fig. 5). A continuous suction device inserted into the bilateral mandibular bodies drained 165 mL during the first 24 h after surgery. This volume decreased to 15 mL during the next 24 h, so the suction device was removed on POD 2. Although the patient had a high WBC count (15,160/ μL) on POD 2, values for RBC, Hb, Ht, and PLT were within normal ranges (Table 2). A nosebleed occurred on POD 4 after the patient coughed with a vigorous head movement, and small nosebleeds occurred several times each day thereafter. The infraorbital area and both cheeks became remarkably swollen on POD 5 along with periorbital ecchymosis (Fig. 6A) and bloody spots in the upper lip, maxillary gingiva (Fig. 6B), and gingivobuccal fold (Fig. 6C). No bloody spots were found in any other region. We considered that the excessive coughing had induced these adverse postoperative findings. However, the spots in the facial area worsened on POD 6, indicating a hemorrhagic tendency, and blood samples were analyzed. The findings showed PLT, $0.3 \times 10^4/\mu\text{L}$; FDP, $21.25 \mu\text{g/mL}$; Fib, 501 mg/dL ; and D-dimer,

$10.67 \mu\text{g/mL}$, indicating DIC (Table 2). A hematology consult confirmed this indication, and the patient was subsequently transferred to that department. Thereafter, anti-inflammatory treatment proceeded with intravenous infusions of prednisolone (60 mg/day; 1 mg/kg/day) and 10 units of transfused platelet concentrate. On POD 7, PLT ($6.5 \times 10^4/\mu\text{L}$), FDP ($11.05 \mu\text{g/mL}$), and D-dimer ($5.24 \mu\text{g/mL}$) values improved and a bone marrow analysis showed a megakaryocyte count of $50/\mu\text{L}$, which was within the normal range. Facial swelling and the bloody spots also improved and the nosebleeds were no longer occurring by POD 11. At that time, PLT ($33.7 \times 10^4/\mu\text{L}$) was within the normal range and the patient was discharged. Blood analysis on POD 25 showed PLT $27.6 \times 10^4/\mu\text{L}$, which was within the normal range. The patient has remained free of adverse events and abnormalities at two years after the surgery.

3. Discussion

Disseminated intravascular coagulation is a hemorrhagic disease characterized by imbalanced blood coagulation [10], and it is broadly coagulation- or fibrinolysis-dominant based on the degree to which the coagulation and fibrinolytic systems are activated [11]. Although PLT sharply decreases in coagulation-dominant DIC, FDP and D-dimer mildly increase, FDP and D-dimer strikingly increase, and PLT slowly decreases in fibrinolysis-dominant DIC. The clinical characteristics of coagulation-dominant DIC comprise hemorrhage from orifices such as the nose, mouth, ears, and veins, as well as hematuria and bloody feces. It can be induced by severe trauma, burns, and underlying diseases such as sepsis. Fibrinolysis-dominant DIC is often clinically asymptomatic, and abnormal blood parameters are the only signs of its existence [12,13]. The clinical findings of the present patient, namely sharply decreased PLT and nosebleeds, were consistent with the characteristics of coagulation-dominant DIC.

Early diagnosis and treatment of DIC are very important because it worsens during progression [14], and a tendency towards DIC should be treated first. However, a gold standard of diagnostic criteria for DIC has not yet been established. Some DIC scoring systems have been introduced via guidelines published in international journals by the Japanese Ministry of Health, Labour and Welfare (JMHLW) [15], the Japanese Association of Acute Medicine (JAAM) [16], and the International Society on Thrombosis and Haemostasis (ISTH) [17].



Fig. 1. Preoperative facial photographs. Frontal (A) and lateral (B) views.

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