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A massive haemorrhage developing during deciduous tooth extraction in a young child: A case report



Gabriele Bocchialini*, Luca Ferrari, Dante Burlini

Maxillofacial Pediatric Surgery Unit, Ospedale dei Bambini-Asst., Spedali Civili, Brescia, Italy

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ABSTRACT

INTRODUCTION: Major haemorrhage developing during oral or maxillofacial surgery is uncommon, but potentially life-threatening. Dental extractions in childhood are common, but blood loss is unusual.

CASE PRESENTATION: Here, we report a rare case of a complication developing during a trivial deciduous tooth extraction. We focus on the great difference between the preoperative orthopantomography and the maxillo-facial computed tomography obtained post-haemorrhage.

DISCUSSION: The surgical procedures most commonly associated with acute haemorrhage are orthognathic, oncological, and temporomandibular joint procedures. When acute haemorrhage develops, the surgeon must remain calm, temporarily arrest the bleeding, and ultimately definitively control it. Although ligation of the offending artery, vein, or vascular malformation is clearly the treatment of choice, identifying the offending vessel and gaining adequate access thereto are often difficult.

CONCLUSION: We emphasise that what we encountered could happen in routine daily practice.

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1. Introduction

Arteriovenous malformation occurs as a result of errors in vascular morphogenesis present at birth [1]: these grow proportionately with age [2,3] and manifest at any time during life due to an event like trauma, surgery or infection [4].

Rarely, a mandibular arteriovenous malformation may be potentially life-threatening. [5] The management of acute haemorrhage can be difficult; although ligation of the offending blood vessel is the treatment of choice, access is often limited or impossible [6].

The high vascularity of the face and the abundant midline anastomoses between the facial arteries can obscure the source of bleeding and delay haemorrhage control [7]. Approximately 51% of vascular malformations occur in the head-and-neck region; the male:female ratio is 1:1.5 [8].

We describe an intra-operative, potentially life-threatening complication that developed during deciduous tooth extraction in a 14-year-old girl with the Pierre Robin sequence and deletion of the long arm of chromosome 4. The preoperative workup featured only orthopantomography.

2. Case report

A 14-year-old girl with the Pierre Robin sequence and deletion of the long arm of chromosome 4 was referred to the Outpatient Unit of the Department of Maxillofacial Pediatric Surgery, Ospedale dei bambini Asst., Spedali Civili, Brescia, Italy. Physical examination revealed only mandibular hypoplasia with multiple deciduous teeth and caries; the patient was noted to be 'fragile' (syndromic child). Orthopantomography revealed mixed dentition with nothing of note in the mandible or maxillae (Fig. 1). The patient had no history of trauma, previous medication, gingival bleeding, blood transfusion, or elective surgery. The dentist planned avulsion of 85 to facilitate the eruption of permanent teeth and avoid abscess formation. The decision was made to perform extraction and deliver the necessary tooth care in the operating room, following our protocol for the treatment of "fragile children". Surgical intervention was performed under both sedation and local anaesthesia. During mobilisation of 85 with a dental elevator, massive bleeding (approximately 700 mL over a few seconds) was encountered, triggering tachycardia and hypotension. The next few moments were life-threatening; the team responded admirably. The patient was intubated immediately after the surgeon momentarily controlled the bleeding by compressing the bleeding area with a finger and aspirated the blood in the oral cavity because the airway was not protected. An acute transfusion was given to avoid hypovolemic shock. The haemorrhage was subsequently controlled by initial bipolar cautery and then by application of bone wax and fibrin glue.

* Corresponding author at: Piazzale Spedali Civili 1, 25123 Brescia, Italy.
E-mail address: gabriele.bocchialini@libero.it (G. Bocchialini).

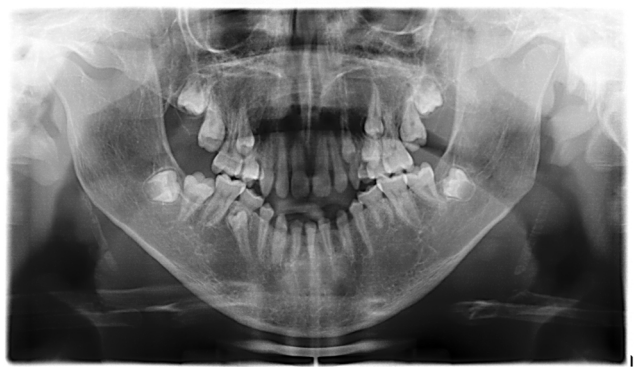


Fig. 1. The preoperative orthopantomogram.

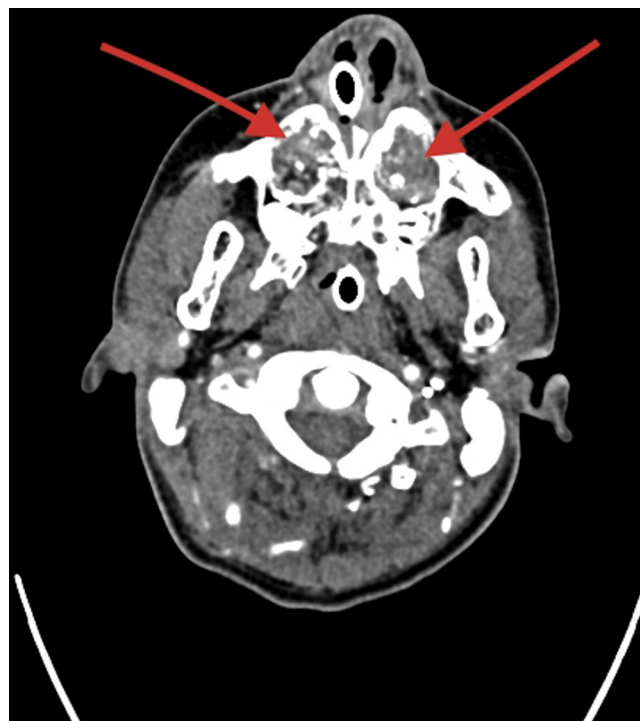


Fig. 3. The angio-CT axial view of the maxillary lesion.



Fig. 2. The axial CT view of the mandibular lesion.

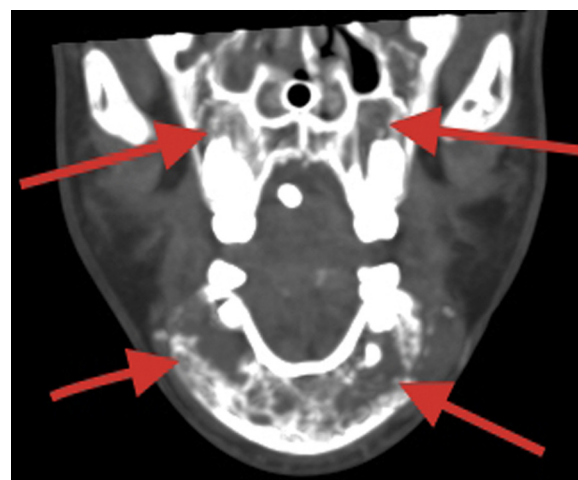


Fig. 4. The angio-CT coronal view of the mandibular and maxillary lesions.

Angio-computed tomography was performed on the intubated patient, with remarkable results. The horizontal branch of the mandible appeared to have expanded; the chin region normally filled with cancellous bone was completely composed of vascularised tissue. Although the tissue was solid, it lacked the typical appearance of high-flux MAV and was evidently a low-flux angiomatous formation. The outer cortical bone (between the premolars and left canines) was focally interrupted by solid, vascularised subcutaneous tissue. The expanded cortical bone exhibited marked thinning, and the solid vascularised tissue extended to the right mandibular angle. The maxilla also appeared to have expanded, containing similar, solid vascularised tissue. Even the sphenoid appeared to have partially expanded; the ethmoid was partially occupied by hypodense material (Figs. 2–4).

Two days later, the patient underwent contrast-enhanced magnetic resonance imaging, which revealed widespread structural subversion of the bones of the head and neck. Almost every bone was affected. The cancellous bony cores of the zygomatic and nasal bones, and those of the alveolar processes and horizontal branches of the jaw, were replaced by tissue with the signal characteristics, and the impregnation and bone development modes, of angiooma. At several points, the resulting structural alterations had remodelled

and disrupted the cortex (Figs. 5 and 6). A Chiari type I malformation was also evident.

The patient was extubated in our paediatric intensive care unit and monitored to prevent recurrence of bleeding. She returned to our department when she had stabilised, two days later. She did not develop neurological or other physical sequelae.

A biopsy of the lesion might be the next step; we are planning the ideal place for a biopsy in order to avoid another massive haemorrhage.

A genetic investigation is ongoing; we seek to discover the connections between the metabolic disease, the bone tumours, the vascular malformations, and any known (but rare) syndrome.

Once the diagnosis is clear, it will be easier to proceed with possible subsequent extractions.

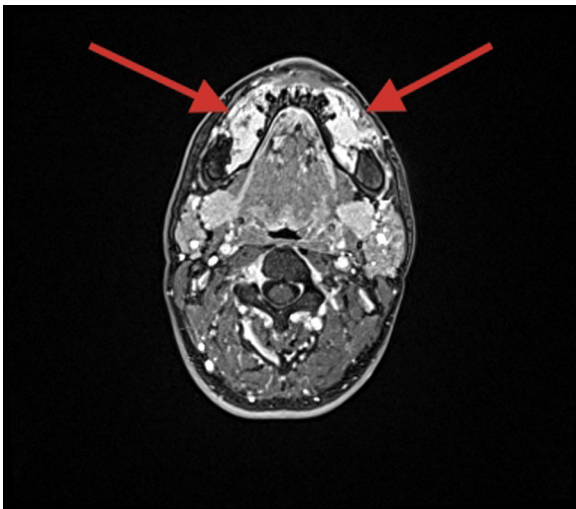


Fig. 5. The MR axial view of the mandibular lesion.

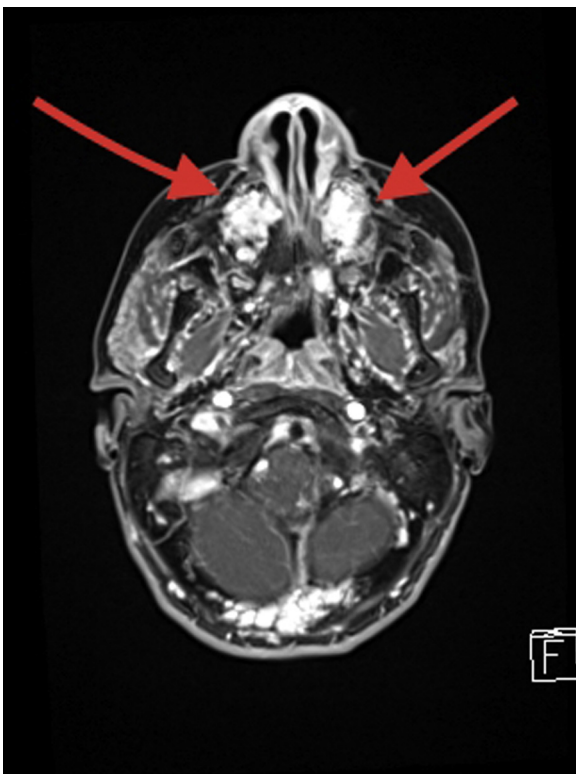


Fig. 6. The MR axial view of the maxillary lesion.

3. Discussion

A life-threatening facial haemorrhage may be associated with facial trauma or recent elective maxillofacial surgery and can trigger hypovolemic shock, tachycardia (heart rate >100), hypotension (systemic blood pressure <100 mm Hg), and a fall in the haematocrit to 24% or of the haemoglobin level to <8 g/dL. The current ATLS guidelines indicate that resuscitation and acute transfusion are essential. Uncontrolled profuse bleeding of the facial vasculature, creating both haemodynamic instability and airway compromise, is rare; the incidence is approximately 1% [7,9–13]. The high-level vascularity of the face and the abundant midline anastomoses between the facial arteries can obscure the source of bleeding and delay haemorrhage control [7].

Various evidence-based guidelines for dental clinicians deal with the use of general anaesthesia when treating children. The guidelines state when anaesthesia should be used and aid in planning dental interventions [14,15]. Young children with other comorbidities may be distressed by the numbness imparted by local anaesthesia and by bleeding associated with dental extraction [16].

Computed tomography showed that the lesions were located in the mandible, maxilla, nose, and zygoma. Extracranial arteriovenous malformations of the head and neck are high-flow lesions, and are among the most serious malformations, being difficult to diagnose, treat, and cure [1], especially in dental practice. Death occurs in 10–15% of patients who haemorrhage and morbidity of various degrees develops in 50% [17]. Multiple imaging modalities should be used to evaluate the malformations in terms of size, flow velocity, flow direction, relationships to surrounding structures, and lesional contents [18]. Angiography is the gold standard when determining the location and flow characteristics of a vascular lesion [1].

Although ligation of the offending vascular malformation is clearly the treatment of choice, definitive control of bleeding has been achieved historically with either ligation of the external carotid artery or selective embolisation; the choice is often determined more by the availability of a skilled interventional radiologist proficient in angiography and embolisation and the ability of a surgeon to perform ligation of the external carotid artery [6].

This case report was written following the SCARE guidelines [19].

4. Conclusion

The decision of how to manage a severe acute haemorrhage is influenced by many factors. The haemorrhage should be arrested at the bleeding vessel, but this is sometimes difficult. In our case, we applied packing and pressure and administered a haemostatic drug intra-alveolarly, partly because vessel ligation was impractical given the extent of vascular disruption evident on CT and partly because selective embolisation was impractical due to the tortuous nature of the bleeding vessels.

Critically, we had initially identified the patient as a “fragile child” (syndromic child); our protocol dictated that she should be treated in the operating room. This meant that, when the emergency developed, we had everything that we needed to hand.

We present this rare case to emphasise that any procedure that seems to be simple can go catastrophically wrong.

Therefore, in this article we would like to emphasise the extreme rarity of the case, focusing on the big difference between the preoperative panoramic radiograph and whole body computed tomography obtained post-haemorrhage

Author contribution

All authors have contributed in study concept, data collection, data analysis and interpretation and in writing the paper.

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Conflicts of interest

None.

Consent

The consent has been given.

Registration of research studies

None.

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