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

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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.

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.

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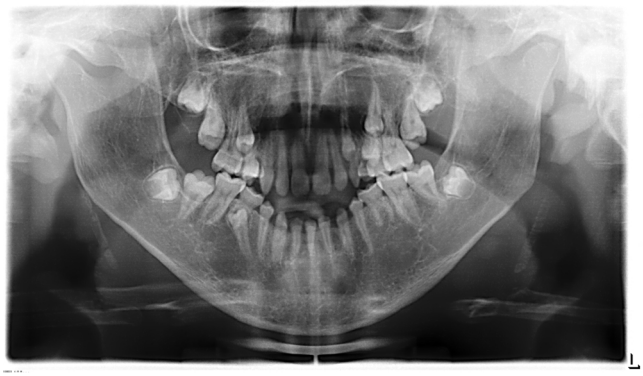

Fig. 1. The preoperative orthopantomogram.

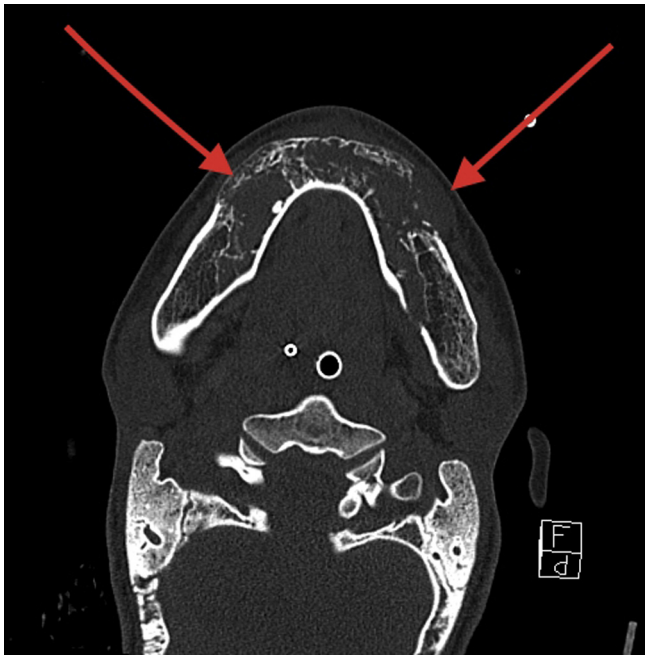

Fig. 2. The axial CT view of the mandibular lesion.

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 angioma. At several points, the resulting structural alterations had remodelled

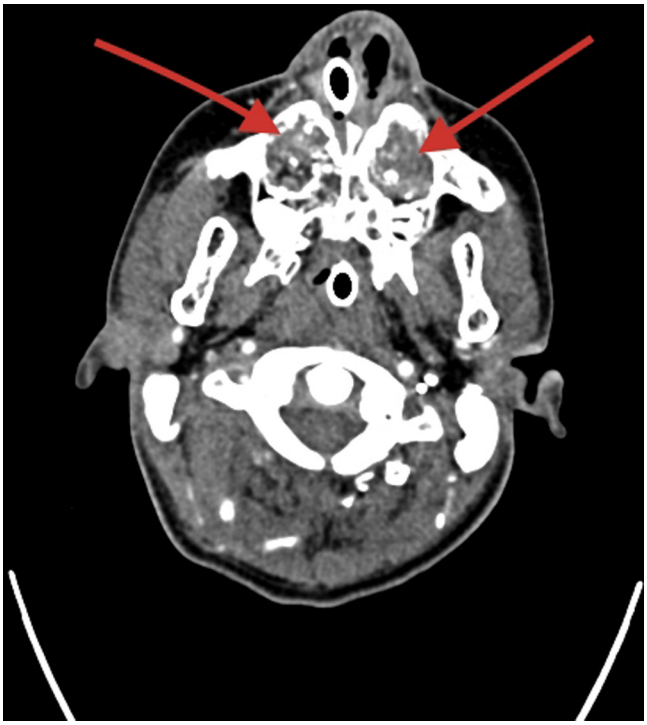

Fig. 3. The angio-CT axial view of the maxillary lesion.

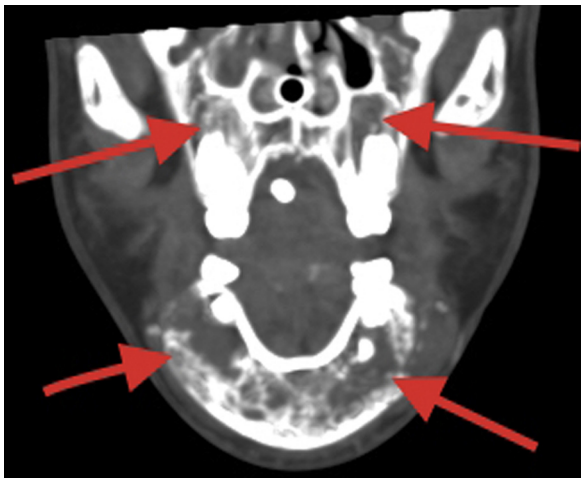

Fig. 4. The angio-CT coronal view of the mandibular and maxillary lesions.

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.