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Implant failure, osteomyelitis and a pathologic mandibular fracture in a patient with extensive submandibular and cervical venous malformations; a case report

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Abstract

Osseo-integrated implants are common place in dental rehabilitation due to their long-term success and safety and now represent the gold standard for tooth replacement. With more implants being placed now than ever before in both general and specialty dental practice, unsurprisingly we are seeing a proportionate rise in implant related complications. The following case report is an example of one such complication of surgery, performed in a 54-year-old female with an atrophic mandible and submandibular/cervical venous malformations resulting in implant loss, secondary osteomyelitis and a pathological fracture.

Introduction

Whilst osseo-integrated implants are a common and successful procedure in dentistry, placement of implants in an edentulous atrophic mandible pose several additional risks due to the reduced bone height/volume and decreased vascular supply as a consequence of alveolar ridge resorption (Spencer, 2018). This loss of vascularised bone mass can potentially increase the risk of pathologic fracture at the time of surgery due to fracture propagation through cortical weakening, originating at the osteotomy site (Boffano et al., 2013). Furthermore, the decreased blood supply, which is worsened from periosteal stripping during the procedure, increases the risk of osteomyelitis, further weakening the mandible and potentially leading to a delayed pathologic fracture among other complications (O'Sullivan et al., 2006; Boffano et al., 2013).

There are several complex and confusing classifications of osteomyelitis of the maxillofacial skeleton, which generally speaking, aim to classify it based on aetiology. A detailed discussion of these is beyond the scope of this paper. However, the mechanism of infective osteomyelitis has been well studied and described by several authors (Marx, 1991; Bernier et al., 1995; Topazain et al., 2002; Baltensperger et al., 2004).

We discuss a case and the management of established mandibular osteomyelitis as a complication of dental implant surgery which resulted in the weakening of an already atrophic edentulous mandible, leading to its pathologic fracture. The case presented an additional challenge to the surgeon due to a large venous malformation in the neck which was not considered during the initial assessment prior to implant placement, and through which surgical access was required to treat the fracture and infection.

Case report

We present a 54-year-old New Zealand European female who was referred to the Auckland Regional Oral and Maxillofacial service by her general dentist in September, 2019. She presented with a primary complaint of significant pain and swelling in the left submandibular region that developed acutely with no preceding trauma.

Three months prior to her presentation she had three endosseous dental implants placed into her anterior edentulous atrophic mandible by her general dentist, who was experienced with the surgical placement of implants. Treatment was completed under local anaesthetic and the intent was to support a mandibular overdenture. The patient had been completely edentulous for 14 years prior to implant placement and pre-operative cone beam computed tomography imaging identified a residual ridge height of 12 mm at the planned surgical site. The implants were placed without complication. The patient was noted to have a very large and mobile tongue which made achieving primary closure over the implant fixtures difficult. Consequently, the mucoperiosteal flap overlying the fixtures dehisced on three occasions in the two weeks following her surgery and the site was left to heal by secondary intention. The patient repeatedly presented to her general dentist in the three months preceding her presentation to hospital with recurrent episodes of pain at the surgical sites. Despite the dentists best efforts and regular reviews, no clinical findings could explain the patients discomfort.

The patients medical history included cerebral palsy and multi-focal, non-syndromic venous malformations involving her brain, tongue and the soft tissues of the neck and submandibular region. She was not taking any regular medications, had no known drug allergies, was a lifelong non-smoker and non-drinker.

Clinical examination revealed a tender and diffuse extra-oral swelling involving the left submandibular and submental regions. There was gross mobility at the left mandibular parasymphysis, and an intra-oral fistula in

this region. A dental panoramic radiograph (Figure 1) and conventional computed tomography image (CT) (Figure 2) revealed a fracture of the left mandibular parasymphysis with surrounding osteolytic change of the fracture segments. The implant originally inserted in this region was reported by the patient to have fallen out several days before presentation. She was systemically well and showed no significant biochemical or haematological abnormalities.

Based on the history, clinical findings, and radiographic assessment, the working diagnosis was a pathologic fracture of the mandible secondary to suspected underlying chronic osteomyelitis or a pathological fracture due to implant placement in the atrophic mandible, leading to an infected non-union of the mandible. After informed consent, a decision was made for an extra-oral approach to the mandible (Figure 3), debridement of infected and necrotic bone, primary bone grafting with a cortico-cancellous iliac crest block graft and rigid-internal fixation utilising a titanium reconstruction plate and locking screws (Figure 4).

The surgical approach was complicated by the venous malformations in the submandibular, submental and cervical regions, which obviously posed an increased risk of major haemorrhage. This also represented an increased risk of damage to the marginal mandibular nerve due to the aberrant anatomy of the venous malformation and its distortion of the surrounding tissues.

Tissue cultures taken at the time of the procedure returned showing growth of *Staphylococcus aureus* as well as other oral commensal bacteria and the histologic features were non-specific, showing a chronic inflammatory infiltrate with no viable bone identified in the specimen. Given the clinical and radiographic findings, positive tissue cultures, and the intra-operative identification of non-viable bone, a diagnosis of chronic osteomyelitis was given. The patient was reviewed by the infectious diseases team and treated with six weeks of oral Augmentin.

Follow up reviews at one, two, three, and twelve months postoperatively revealed both clinical and biochemical resolution of infection. Dental panoramic radiograph (Figure 5) and cone beam computed tomography (CBCT) imaging twelve months following the procedure revealed interval healing consistent with bony union and successful treatment. Unfortunately, but not surprisingly, there was total and persistent loss of sensation to the patient's lower lip and chin bilaterally due to a combination of the pathological fracture and the need for bicortical fixation for mandibular stabilisation. The facial nerve function was normal.

Discussion

Infective osteomyelitis of the jaws (OMJ) is an inflammatory process, which typically initiates as a bacterial or fungal infection of the medullary bone of the mandible or maxilla that extends into the cortical bone and periosteum (Baltensperger and Eyrich, 2009). In established infections, the accumulation of pus and oedema in the medullary cavity leads to vascular compromise, tissue ischaemia and eventual

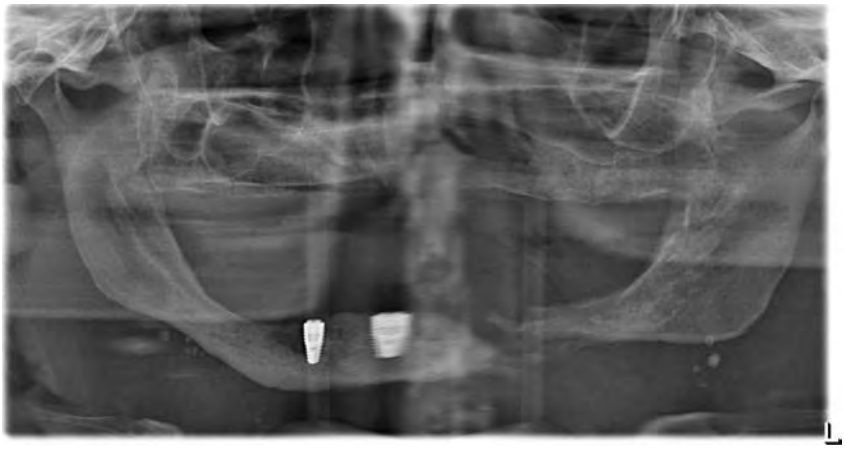


Figure 1 *(left).* Preoperative dental panoramic radiograph.

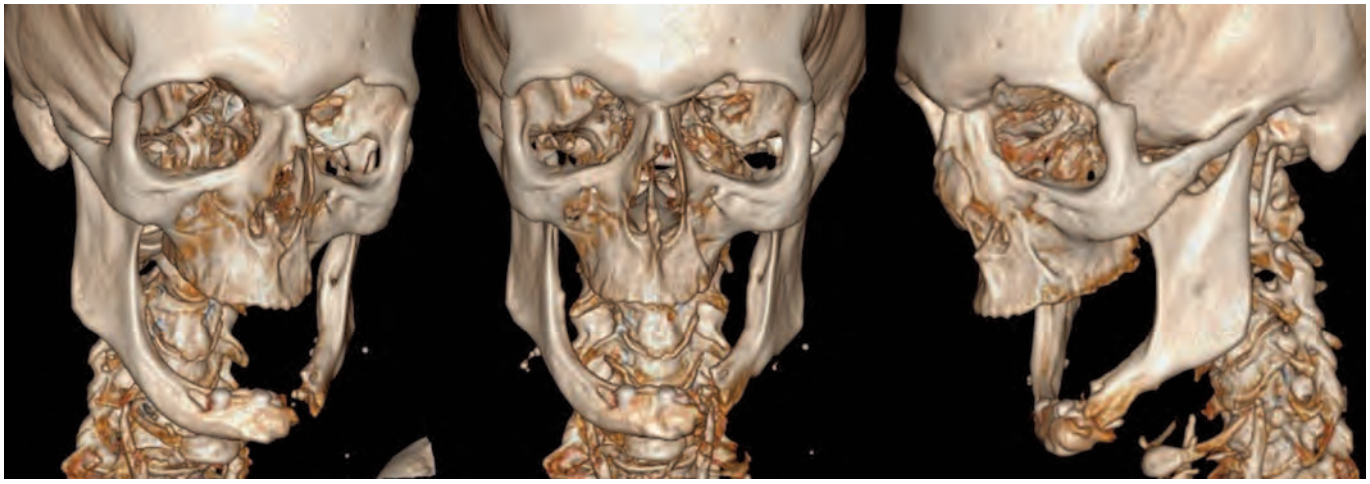
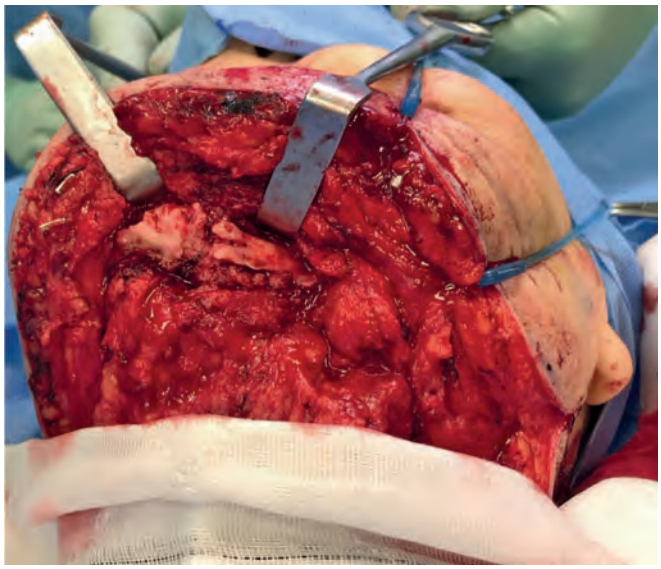
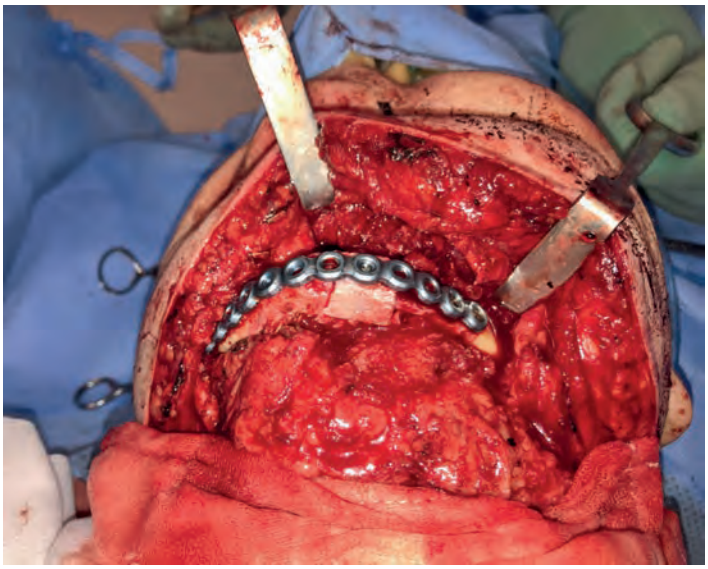


Figure 2 *(below).* Preoperative CT imaging.





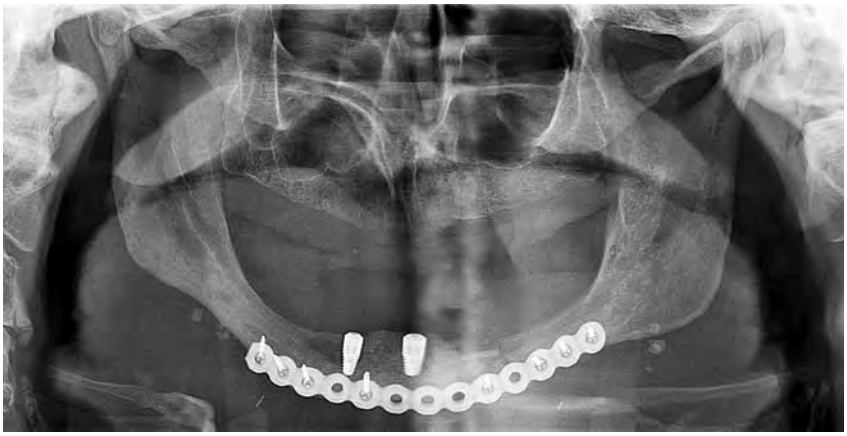


Figure 3. Exposure of the mandible and left parasymphysis defect.

Figure 4. Reconstruction plate fixating the mandible and autogenous graft.

Figure 5. 10-month post-operative CBCT image.

necrosis resulting in sequestra formation which is a hallmark sign of osteomyelitis (Yahalom et al., 2016). If osteomyelitis has been present for one month or less it can be classified as acute and progression past this arbitrary time point defines chronic osteomyelitis (Baltensperger et al., 2009). OMJ is often a polymicrobial infection (Semel et al., 2016) and the introduction of the causative microorganisms into the jaw bones is most frequently the result of trauma or untreated odontogenic infections (Balanger et al., 2017). The placement of endosseous dental implants represents a rare, but potential aetiology for the development of OMJ which may arise following the introduction of microbes into the medullary cavity during the surgical placement of the fixture (Yahalom et al., 2016) or as a consequence of untreated peri-implantitis (Doll et al., 2015). Individual risk factors include conditions that alter the vascularity of the mandible and maxilla, such as bone resorption as outlined, systemic diseases or acquired states that alter the host defence and lifestyle factors such as smoking (Marx, 1991; Kellesarian et al., 2018).

There have been 47 cases of OMJ associated with dental implants described in the literature to date (Kellesarian et al., 2016; Chatelain et al., 2018; Blanger et al., 2017; Schlund et al., 2017). Of these, there appears to be a female predilection and it is encountered most frequently in individuals in their sixth decade of life. The overwhelming majority of cases have been reported to occur in the mandible, with only 2 presenting in the maxilla (Piattelli et al., 1995; Yahalom et al., 2016). This is consistent with cases of OMJ of all other causes, believed to be due to the mandible's decreased vascularity, compared to that of the maxilla (Hudson, 1993). Only two cases of mandibular osteomyelitis associated with a dental implant progressing to a pathologic fracture have been reported in the literature (O'Sullivan et al., 2006; Doll et al., 2015); now we present the third.

The treatment philosophy regarding OMJ and infected mandibular fractures has changed considerably over recent decades. Historically, infected fractures and cases of OMJ were managed via closed techniques with the establishment of drainage, promotion of sequestration and the removal of compromised teeth. The infective process was left to run its course and it was numerous months until union occurred or reconstruction with autologous grafts was considered (Benson et al., 2006). However, these principles changed when emerging evidence suggested that the pathogenesis of the infective process was not caused by contamination, but by fracture instability (Koury and Ellis, 1992). Therefore, treatment principles have evolved to what they are today with the removal of the source of infection, surgical eradication of the affected bone and, in the presence of a fracture, rigid internal fixation with immediate primary bone grafting followed by adjuvant systemic antimicrobial therapy (Koury and Ellis, 1994; Benson et al., 2006; Marschall et al., 2019).

The extent of surgery is largely determined by the severity of the infection and the size of the defect. As seen in the present case, severe established infections with complications necessitate aggressive surgical intervention in the first instance. Baur and colleagues (2015) reported a 50% recurrence rate of OMJ after marginal resection versus a 5.5% recurrence rate after segmental resection (Baur et al., 2015). Therefore, the use of a seemingly more aggressive approach may be more "conservative" in the long run, due to more favourable outcomes and fewer returns to theatre (Ellis et al., 2008). Immediate autologous bone grafting used in conjunction with rigid internal fixation at the time of surgical debridement has been identified as effective definitive treatment, with Benson and colleagues (2006) reporting the outcomes of this approach, comparable to those with noninfected sites (Benson et al., 2006).

When a venous malformation is present in the region where a patient is requiring surgical intervention, various anatomical considerations need to be made to avoid complications. Venous malformations arise through errors in endothelial cell morphogenesis, which results in disorganised angiogenesis and intimal smooth muscle proliferation leading to the formation of dilated networks of venous lakes (Coletti et al., 2017; Hage et al., 2018). Venous malformations are not uncommon with an incidence estimated between 1:5000-10,000 (Coletti et al., 2017; Park et al., 2019). They can be categorised as sporadic or inherited and unifocal or multifocal (Park et al., 2019). Sporadic venous malformations are predominantly unifocal and form the vast majority of cases, whereas inherited forms are frequently multifocal and represent between 1-7% of cases (Dompmartin et al., 2010; Park et al., 2019), with the present case falling into this category.

Large infiltrating venous malformations have fragile walls that will easily rupture, and obtaining haemostasis in this scenario is complex, which can quickly lead to life threatening haemorrhage (Colletti et al., 2017). Up to 45% of head and neck venous malformations will have poorly defined margins (Park et al., 2019) and if the lesion does not originate from muscular tissue (i.e., it is cutaneous or mucosal in origin), almost all will invade surrounding skeletal muscle (Park et al., 2019). As a consequence, the venous malformation will distort normal tissue anatomy and anatomical landmarks of the neck. Therefore, when carrying out an extraoral approach to a mandible in these patients, subplatysmal dissection and identification of vital anatomical structures such as the marginal mandibular nerve, submandibular gland and facial artery and facial vein is complex. A careful, well-planned approach with appropriate three-dimensional pre-operative imaging is recommended to avoid potentially life threating complications.

To the best of our knowledge, this report represents the first published case of an extra-oral approach to, and reconstruction of, a mandible in a patient with a large venous malformation of the neck in the context of failed implant therapy and pathological fracture.

Conclusion

We present the third published case of a pathologic mandibular fracture arising in a site of osteomyelitis due to a failed dental implant, and the only published case describing the extra-oral neck approach to the mandible through large venous malformations. Although serious complications in dental implant surgery generally occur infrequently, we hope our paper will serve as a cautionary tale. The intent is not to dissuade or discourage the dental surgeon, but further empower them to make good decisions, whilst minimising the possibility of complications, leading to better outcomes for their patients.

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