CASE REPORT

Post-traumatic near-complete aseptic necrosis of the maxilla: a case report and review of the literature

N Khan¹, W Memon^{*1}, M Idris¹, M Ahmed² and M Taufiq¹

¹Department of Radiology, Aga Khan University Hospital, Karachi, Pakistan; ²Department of Dentistry, Aga Khan University Hospital, Karachi, Pakistan

Osteonecrosis of the mandible and the maxilla is known; however, aseptic necrosis of the maxilla after traumatic fracture is hardly reported. This case aims to help clinicians realize the need to closely follow up and treat such patients with trauma as it can lead to osteonecrosis of the bone and cause difficulty in daily activities. We have also carried out a thorough review of the literature to give a concise understanding of all disease processes that may lead to osteonecrosis of the maxilla. Such an article has not previously been published. *Dentomaxillofacial Radiology* (2012) **41**, 429–431. doi: 10.1259/dmfr/12765535

Keywords: post-traumatic necrosis; maxilla; osteonecrosis; aseptic necrosis

Introduction

Post-traumatic necrosis of the maxilla is rare.¹ Only two case reports have been published describing necrosis of the maxilla after traumatic fracture.^{1,2} Osteonecrosis in the maxilla or anywhere else in the body develops when the vascularity is compromised. This may occur from a traumatic fracture; however, it is also a known complication after surgical osteotomy.^{3,4} Other known causes include infections, tumours and radiation.^{5–9} Recently, bisphosphonate-associated osteonecrosis has been reported as an emerging oral complication in cancer patients.^{10–13} We report a case of aseptic necrosis of the maxilla after mid-face trauma.

Case report

A 38-year-old male with no known comorbidities presented to the maxillofacial dental clinic with difficulty in chewing on the right side of his jaw. He complained that he was able to freely move a portion of his right upper gum, including the teeth, separately from the rest of the jaw. This caused difficulty in chewing. He had no other complaints. He had been in a road traffic accident 2 months previously where he was sitting in a car in the passenger seat without a seatbelt and was struck against the dashboard. He received first

aid from a nearby primary healthcare centre and had, among other injuries, trauma to his mid-face, which on examination was diagnosed as a fracture of the right maxilla. He was advised to see a maxillofacial or dental surgeon but, as the bruising and pain over his cheek subsided without any difficulty and he recovered from his injuries without any deformity, he did not follow this advice. He did not have an infection in the cheek and felt completely well. However, in the last few weeks he started noticing the abnormal mobility of his jaw.

A non-contrast CT scan of the face was performed which showed near-complete necrosis of the right maxillary bone. The body of the maxilla with the maxillary sinus, the alveolar process, zygomatic process and palatine process were completely absent (Figure 1a–d). Only the most superior part of the right maxilla above the maxillary sinus, which included the infraorbital foramen, was present (Figure 1c) and the frontal process of the maxilla was intact; these were forming the rim and walls of the orbit. Only a fine lace-like remnant of bone was seen along the outermost margins of the maxilla. The absence of alveolar processes gave the appearance of a floating upper right incisor and canine and molar teeth (Figure 1a–d).

No soft-tissue swelling or mass was present and no sclerosis, fracture or evidence of previous surgery was visible. On the basis of these findings a diagnosis of aseptic necrosis of the maxilla was made. He was referred to the maxillofacial clinic where he was prescribed prophylactic antibiotics. A maxillary necrosectomy with maxillary reconstruction was offered; however, at the last follow-up he still had not undergone surgery.

*Correspondence to: Dr Wasim Memon, Department of Radiology, Aga Khan University Hospital, Stadium Road, PO Box 3500, Karachi 74800, Pakistan. E-mail: wasim.memon@aku.com

Received 9 July 2010; revised 7 October 2010; accepted 28 October 2010

Figure 1 (a–d) Non-contrast CT scan of face in bone window settings shows nearly complete absence of the right maxilla. (a) Axial section shows complete absence of palatine process of the right maxilla up to midline. (b,c) Reformatted coronal sections demonstrate absence of zygomatic and alveolar processes of the right maxilla causing the floating appearance of the right upper jaw teeth. (c) Coronal section shows the presence of the uppermost part of the right maxilla which includes the infraorbital foramen and frontal process. (d) Reformatted sagittal section shows the absence of the walls of the maxillary sinus and floating teeth

Discussion

To the best of our knowledge, osteonecrosis of the maxilla from traumatic fracture has only previously been reported twice.^{1,2} In one case described by Cornah and O'Hare,¹ the patient had extensive maxillary fractures associated with Le Fort I and II-type fractures. This patient also developed maxillary infection. The maxilla was excised and appeared to have undergone complete necrosis.¹ In the other case, the patient sustained a Le Fort I-type fracture that was inadequately treated and resulted in a large maxillary defect.²

Osteonecrosis of the maxilla occurs when the vascularity is compromised.¹⁴ The vascular supply of the maxilla arises from the descending palatine artery and internal maxillary artery.¹⁴ There are a few known conditions that can affect this vascularity resulting in osteonecrosis of the maxilla. The causes can be broadly classified into septic and aseptic

necrosis. Septic osteonecrosis can occur from a variety of infections and aseptic necrosis includes malignancy, radiation, trauma, surgery and bisphosphonates therapy.^{5–9}

Septic necrosis of the maxilla can be caused by fungal infections such as mucormycosis and aspergillosis.^{5,6} Viral infections such as herpes zoster^{7,8,15} affect immunocompromised patients, including those who have uncontrolled diabetes, lymphomas, organ transplants and long-term corticosteroid and immunosuppressive therapy.^{5,6} Infections such as mucormycosis form thrombi within the blood vessels and result in decreased blood supply and necrosis.⁵ Osteonecrosis from herpes occurs when the varicella zoster virus reactivates from latency and becomes an active viral replication. Our patient did not have any signs or symptoms of infections in his post-traumatic period and he was not immunocompromised.

Malignancy and radiation in the region of the maxillary sinus can result in aseptic necrosis of the maxilla. Malignancy, such as squamous cell carcinoma of the maxillary sinus, can cause direct destruction of the maxilla and also occlude the vasculature, resulting in osteonecrosis.¹⁴ Radiation of tumours in the head and neck region can also result in osteonecrosis, which is considered as the worst long-term complication of radiotherapy.^{9,12}

Le Fort I osteotomy has become a routine procedure in elective orthognathic surgery.⁴ Although rare, ischaemia is one of the known complications of this procedure. In a study involving 1000 patients with Le Fort I osteotomy, ischaemic complications affected 10 patients, of whom 2 (0.2%) experienced partial aseptic necrosis of the alveolar process.⁴ One theory suggests that the blood flow to the maxilla after osteotomy is reduced, especially in diabetic patients, which may be a cause of necrosis in osteotomy patients.¹⁶

Recently, there have been reports in medical literature supporting the association between bisphosphonate use and osteonecrosis.¹¹ This is most commonly reported in patients who are receiving bisphosphonates for metastatic disease.¹¹ Bisphosphonates inhibit resorption by osteoclasts and cause induction of apoptosis.¹⁷ The mandible is the most common bone affected followed by the maxilla.¹¹ Osteonecrosis in patients taking bisphosphonate can show focal lytic changes on conventional radiographs and CT.¹¹ A positive association is seen between osteonecrosis and length of intravenous bisphosphonate therapy, oral hygiene and dental extraction.^{11–13}

Lesser-known causes of osteonecrosis include Wegener's granulomatosis, which is an inflammatory disease affecting blood vessels.⁵ Acute necrotizing ulcerative gingivitis arising from the gingiva can involve the maxilla,⁵ while Gorham's disease is another rare disorder of unknown aetiology that results in destruction

and resorption of bone from proliferation of non-neoplastic vascular or lymphomatous tissue.¹⁸

Surgical reconstruction of the maxilla is a daunting task for the surgeon because it needs to include the restoration of the original form and function that has been lost.¹⁹ The masticatory function, the resonance of the voice and the architecture of the face has to be restored so that the quality of life after the operation is optimal.¹⁹ Many local flaps have been described which achieve soft-tissue closure of the defect. Local flaps of the palatal mucosa from the contralateral side or ipsilateral cheek mucosa can be used to close small defects. Pedicled regional flaps are used for larger defects; the most common is the ipsilateral temporalis myofascial flap. Microvascular free-tissue transfer from the radical forearm flap and the latissimus dorsi flaps have also been advocated, but they rarely provide a functional solution. Osseous reconstruction of the maxilla has been attempted with little success.²⁰ Titanium mesh implants have also been tried as carriers for osseous tissue and the bone can then be used to place dental osseointegrated implants.²⁰ The reconstruction of the maxilla and the surrounding tissues using a prosthetic maxillary obturator device fabricated to include the lost dental arch still remains the method of choice because it achieves the best possible results compared with the other methods.²⁰

Conclusion

Trauma to the face is common in road traffic accidents. Mid-face trauma can result in fractures of the maxilla. Such injuries should be properly managed given that they can result in osteonecrosis of the maxilla. Although rare, it should not be taken lightly as it can lead to significant anxiety for the patient and necessitate major surgery to fill the defect.

References

- Cornah J, O'Hare PM. Total maxillary necrosis following severe facial injury. A case report. *Br J Oral Surg* 1981; **19**: 148–150.
- Migliorisi JA. Closure of a post-traumatic maxillary defect. A case report. *Int J Oral Maxillofac Surg* 1997; **26**: 336–337.
- Lanigan DT, Hey JH, West RA. Aseptic necrosis following maxillary osteotomies: report of 36 cases. *J Oral Maxillofac Surg* 1990; **48**: 142–156.
- Kramer FJ, Baethge C, Swennen G, Teltzrow T, Schulze A, Berten J, et al. Intra- and perioperative complications of the LeFort I osteotomy: a prospective evaluation of 1000 patients. *J Craniofac Surg* 2004; **15**: 971–977.
- Auluck A. Maxillary necrosis by mucormycosis. A case report and literature review. *Med Oral Patol Oral Cir Bucal* 2007; **12**: E360–4.
- Napoli JA, Donegan JO. Aspergillosis and necrosis of the maxilla: a case report. *J Oral Maxillofac Surg* 1991; **49**: 532–534.
- Owotade FJ, Ugboke VI, Kolude B. Herpes zoster infection of the maxilla: case report. *J Oral Maxillofac Surg* 1999; **57**: 1249–1251.
- Garty BZ, Dinari G, Sarnat H, Cohen S, Nitzan M. Tooth exfoliation and osteonecrosis of the maxilla after trigeminal herpes zoster. *J Pediatr* 1985; **106**: 71–73.
- Reuther T, Schuster T, Mende U, Kübler A. Osteoradionecrosis of the jaws as a side effect of radiotherapy of head and neck tumour patients—a report of a thirty-year retrospective review. *Int J Oral Maxillofac Surg* 2003; **32**: 289–295.
- Migliorati CA, Casiglia J, Epstein J, Jacobsen PL, Siegel MA, Woo SB. Managing the care of patients with bisphosphonate-associated osteonecrosis: an American Academy of Oral Medicine position paper. *J Am Dent Assoc* 2005; **136**: 1658–1668.
- Estilo CL, Van Poznak CH, Williams T, Bohle GC, Lwin PT, Zhou Q, et al. Osteonecrosis of the maxilla and mandible in patients with advanced cancer treated with bisphosphonate therapy. *Oncologist* 2008; **13**: 911–920.
- Durie BG, Katz M, Crowley J. Osteonecrosis of the jaw and bisphosphonates. *N Engl J Med* 2005; **353**: 99–102.
- Van Poznak C, Estilo C. Osteonecrosis of the jaw in cancer patients receiving IV bisphosphonates. *Oncology (Williston Park)* 2006; **20**: 1053–1062.
- Traxler H, Windisch A, Geyerhofer U, Surd R, Solar P, Firbas W. Arterial blood supply of the maxillary sinus. *Clin Anat* 1999; **12**: 417–421.
- Badjate SJ, Cariappa KM, Sheno SR, Nakhate S. Ramsay-Hunt syndrome complicating osteonecrosis of edentulous maxilla and mandible: report of a rare case. *J Maxillofac Oral Surg* 2009; **8**: 188–191.
- Buckley JG, Jones ML, Hill M, Sugar AW. An evaluation of the changes in maxillary pulpal blood flow associated with orthognathic surgery. *Br J Orthod* 1999; **26**: 39–45.
- Fleisch H. Bisphosphonates—preclinical. In: Fleisch H, ed. *Bisphosphonates in bone disease: from the laboratory to the patients*. 4th edn. San Diego, CA: Academic Press; 2000. pp 34–51.
- Patel DV. Gorham's disease or massive osteolysis. *Clin Med Res* 2005; **3**: 65–74.
- Nekora-Azak A, Evlioglu G, Ozdemir-Karatas M, Keskin H. Use of biofunctional prosthetic system following partial maxillary resection: a clinical report. *J Oral Rehabil* 2005; **32**: 693–695.
- Parr GR, Tharp GE, Rahn AO. Prosthodontic principles in the framework design of maxillary obturator prostheses. *J Prosthet Dent* 1989; **62**: 205–212.