A sagittal split osteotomy approach for removal of a large cementoblastoma at the mandibular angle.

Précis

This case report demonstrates the effectiveness of sagittal split osteotomy in the removal of a mandibular cementoblastoma.

Abstract

Benign lesions at the angle of the mandible are frequently removed by a conventional intra-oral approach to gain access and achieve complete visualisation. This method is quick and effective when dealing with small, benign lesions that are superficially located at the angle of the mandible. The removal of large and deeply located lesions with a conventional intra-oral approach, however, brings about a unique set of challenges, particularly when the third molar is displaced towards the inferior border of the mandible, including: lack of complete visualisation of the lesion; difficulty in identification and protection of the inferior alveolar nerve; and, the necessity of removing a considerable amount of osseous structure, thus increasing the risk of a mandibular fracture. Alternative techniques for such lesions include an extra-oral approach, but this could potentially create a cosmetic defect from cutaneous scarring and can result in facial nerve injury.

This case report describes the use of a unilateral sagittal split osteotomy (SSO) in the removal of a mandibular cementoblastoma. This is a safe and effective technique allowing optimal access to the tumour with complete visualisation, identification and protection of the inferior alveolar nerve, and with minimal bone removal, while maintaining mandibular integrity, strength and facial aesthetics.

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Introduction

A cementoblastoma, also referred to as a true cementoma, is a rare, benign odontogenic tumour arising from ectomesenchymal cells.[1] They have also been referred to in the literature as: sclerosing cementoma; peri-apical fibro-osteoma; and, peri-apical fibrous dysplasia. Disorganised proliferation of cementoblasts results in subsequent deposition of cement-like tissue around the roots of teeth. Cementoblastomas account for between 0.69% and 8% of all odontogenic tumours and tend to occur between the second and third decades of life, with a median age of 20 years and an age range of eight to 44 years. Some studies show no no gender preference,[2] while others show a higher rate of occurrence in males.[3] Cementoblastomas tend to occur more frequently in the posterior mandible, involving the roots of premolar and molar teeth. They are asymptomatic lesions, which demonstrate a slow and expansile growth, and are usually discovered as an incidental radiographic finding.[4] However, cortical bone expansion can result in facial asymmetry and symptomatic painful lesions when facial nerves become involved.[5]

Because cementoblastomas have unlimited growth potential, treatment includes tumour resection with the extraction of the associated tooth. If the tumour is small at the time of diagnosis, treatment may consist of surgical removal with endodontic therapy and retention of the involved tooth. The traditional surgical approach for excision of a cementoblastoma at the mandibular angle is removal of bone to gain access to the tumour. However, the surgical risk increases with removal of larger lesions via traditional techniques, which will involve the removal of larger amounts of bone,

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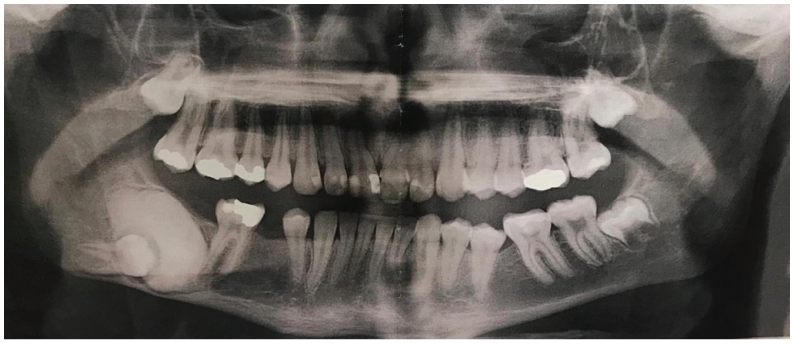

FIGURE 1: The patient's OPG revealed a large, radio-opaque mass, with a radiolucent rim measuring approximately 3cm mesiodistally, at the right angle of the mandible, extending from the lower border of the mandible to the alveolus.

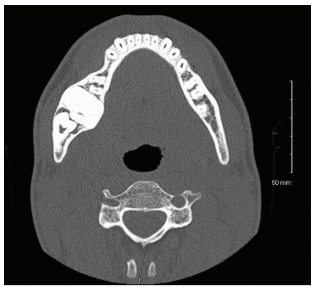

FIGURE 2a: CT axial view.

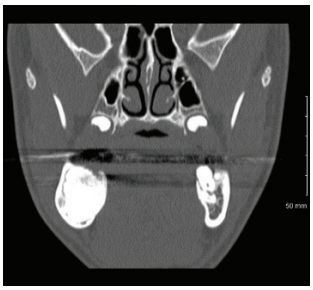

FIGURE 2b: CT coronal view (1).

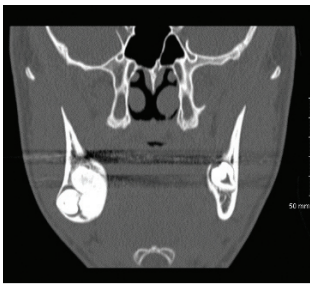

FIGURE 2c: CT coronal view (2).

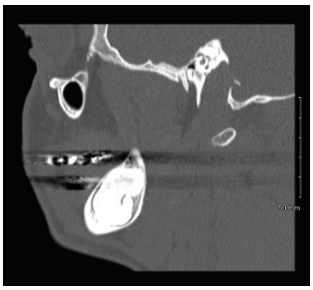

FIGURE 2d: CT sagittal view.

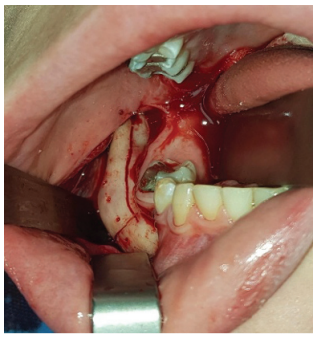

FIGURE 3a: The mandible was pre-plated before completing the mandibular split

FIGURE 3b: The split was uneventful and the buccal segment was lifted off the tumour, providing direct visualisation.

making the mandible weak with an increased potential for fracture. The integrity of the cortical plate remains critical in these circumstances to maintain mandibular strength. To avoid such complications, the use of a sagittal split osteotomy (SSO) technique was introduced by Rittersman and van Gool in 1979 for removal of a large non-malignant lesion.[6] Presented in this case report is the use of an SSO approach to remove a large radio-opaque lesion at the angle of the mandible with a favourable outcome.

Case report

A 17-year-old male was referred to the Oral and Maxillofacial Surgery Department at Cork University Hospital, after an incidental finding on orthopantomogram (OPG) during an orthodontic appointment. The OPG revealed a large, radio-opaque mass, with a radiolucent rim measuring approximately 3cm mesiodistally, at the right angle of the mandible, extending from the lower border of the mandible to the alveolus (**Figure 1**). The patient was asymptomatic and had no relevant medical or surgical history.

Clinical examination revealed a non-tender and firm expansion of the buccal cortex in the mandibular angle area with no facial asymmetry. The lower right third molar (LR8) was displaced towards the inferior border of the mandible and was intimately related to the inferior alveolar nerve (IAN), which had intact sensation. The extent of the lesion placed the patient at risk of a mandibular fracture and he was thus advised against involvement in contact sports.

Computerised tomographic (CT) images revealed a large, well-delineated, radio-opaque mass at the right angle of the mandible, originating from LR8 and causing thinning of the buccal and lingual cortical plates. The IAN was displaced inferiorly on the lingual aspect of the lesion (**Figures 2a-2d**).

Following discussion with the patient and a family member, consent was obtained for surgery, with a full written and verbal explanation of the risks and benefits. A right-sided SSO was performed under general anaesthetic, as this technique would provide adequate surgical access to the lesion, as well as preserving the buccal and lingual cortices. A buccal flap was raised and standard SSO bone cuts were made with a reciprocating saw. The mandible was pre-plated before completing the mandibular split (**Figure 3a**). The split was uneventful and the buccal segment was lifted off the tumour, providing direct visualisation (**Figure 3b**). Despite having a radiolucent rim around the radio-opaque lesion on imaging, the tumour could not be shelled out as a whole and the lateral half had to be cut down into multiple small pieces with a fissure bur in order to remove them. The

FIGURE 4: The remaining medial half of the tumour was shelled out carefully after identifying and protecting the IAN.

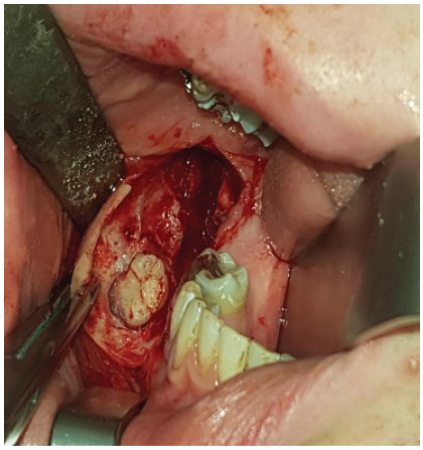

FIGURE 5a: LR8 was sectioned with a fissure bur and removed completely without any damage to the thin buccal cortex.

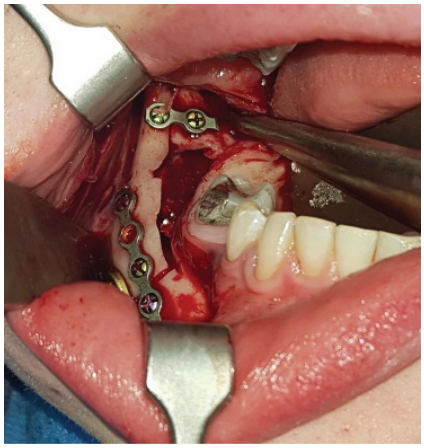

FIGURE 5b: The mandible was plated back into the pre-plated position with a six-hole 2.0mm plate buccally, a two-hole mandibular reconstruction plate buccally at the lower border of the mandible, and a two-hole 2.0mm plate inserted in the retromandibular area across the sagittal bony cut.

remaining medial half of the tumour was then shelled out carefully (**Figure 4**) after identifying and protecting the IAN. LR8 (**Figure 5a**) was sectioned with a fissure bur and removed completely without any damage to the thin buccal cortex. The mandible was then plated back (**Figure 5b**) into the pre-plated position with a six-hole 2.0mm plate buccally, a two-hole mandibular reconstruction plate buccally at the lower border of the mandible, and a two-hole 2.0mm plate inserted in the retromandibular area across the sagittal bony cut. The excised specimen was sent for histopathology.

Postoperatively, the patient had reduced sensation along the distribution of the right IAN, affecting the lower lip. This had been explained to the patient as one of the potential complications. The patient was discharged home next day on prophylactic antimicrobial therapy, analgesia, chlorhexidine mouthwash, and instructions regarding maintaining good oral hygiene and a soft diet (**Figure 6**). At one-week postoperative review he reported to be managing well, was taking a soft diet, had normal mouth opening, and reported no changes to his occlusion. Some sensation was returning to his lower right lip. At one-month review, a further improvement was reported in lower lip sensation, and at two months a further improvement again, with the affected region demonstrating satisfactory clinical healing. Six months postoperatively, OPG demonstrated excellent boney infill and the patient reported full resolution of parasthesis/anaesthesia of lower lip (**Figure 7**). The histopathology report confirmed the diagnosis as cementoblastoma with presence of bony-type material and cementum, with associated well-vascularised fibrous connective tissue stroma.

Discussion

Benign cementoblastoma was described as early as 1927 by Dewy[7] and in 1930 by Norberg,[8] who defined it as a true neoplasm of cementum or cementum-like tissue and formed on a tooth root by cementoblasts. The World Health Organisation first named this neoplasm "benign cementoblastoma" in its 1971 classification, and defined it as "a neoplasm characterised by formation of sheds of cementum-like tissue, which may contain a very large number of reversal lines and may be unmineralised at the periphery of the mass or in the more active growth areas".[9]

As previously stated, cementoblastomas are rare, slowly growing but expansile lesions with unlimited growth potential. They are often asymptomatic and mostly discovered incidentally on radiographic investigation, as in this case. However, they can also cause displacement and mobility of teeth, encroach on the nerve pulp, lead to root resorption, invade the maxillary sinus or orbital floor, cause facial asymmetry, and result in pain and paraesthesia, as well as potential pathological fracture of the jaw, as has been reported by Chrcanovic *et al.* in 2017,[10] on systematic review of 258 cases in the literature. They also concluded that cementoblastomas occur more commonly in the mandible, usually within the molar and premolar region, in a younger cohort of patients, reporting a mean age of 20.7 years, findings consistent with the patient in this case report.

Radiographically on OPG a cementoblastoma typically presents as an area of radiodensity or mixed density with a rounded or sunburst appearance and a relatively radiolucent rim. More often than not there is loss of the periodontal ligament space as well as root resorption of the associated tooth. Displacement and involvement of adjacent teeth and cortical erosion can also occur. When the attachment to the root of the involved tooth is apparent, this radiographic finding is nearly pathognomic. Further imaging via CT or cone-beam CT (CBCT) scan is commonly requested as an adjunctive aid to diagnosis. As well as an adjunct to clinical diagnosis, CT imaging is also useful in the planning of surgical intervention and it is critically important for planning excisions of larger lesions, as it will provide radiographic details about the lesion and its relationship with important structures in three dimensions.[11]

Histopathologically, cementoblastomas are typically characterised by masses of hypocellular cementum within a fibrovascular stroma, usually surrounded by a well-defined cementoblastic rim. Within the fibrovascular stroma, multinucleated osteoclast-type giant cells and plump cementoblasts can

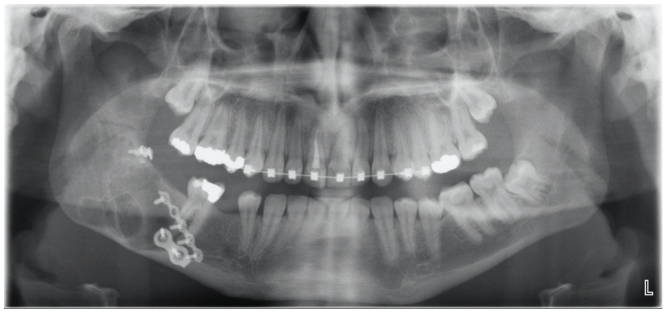

FIGURE 6: OPG recorded at day 1 postoperatively.

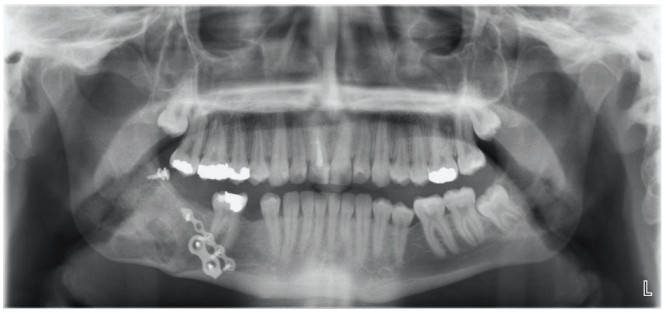

FIGURE 7: OPG recorded at six months postoperatively showing excellent bony infill.

occur. Prominent basophilic reversal lines within the cementum result in a pagetoid appearance. Peripherally to the lesion, the radiating columns of cellular unmineralised tissue account for the radiolucent zone. The spectrum of radiographic appearance of a cementoblastoma depends on its degree of mineralisation. Early-stage lesions generally appear more radiolucent and should be differentiated from periapical inflammatory lesions such as focal sclerosing osteotitis and focal osteomyelitis. In mature stage this lesion may be difficult to distinguish from hypercementosis, cemento-ossifying fibroma, osteoma, benign osteoblastoma, odontomas, and calcifying epithelial odontogenic tumours.[12]

Definitive diagnosis is made on a combination of clinical findings, radiographic investigations and histological analysis. It is imperative to include osteosarcoma in your differential diagnosis, although this would be an unusual site for that to occur.

A number of surgical techniques have been utilised in the removal of benign mandibular lesions, including:[13]

A. the intra-oral buccal approach, in which the buccal cortex is removed and the lesion is adequately exposed before enucleation;
B. the intra-oral lingual approach, in which the lingual cortex is removed and the lesion is exposed, with care being taken not to injure the lingual nerve;
C. segmental osteotomy, which involves an extra-oral submandibular approach with partial bone resection and reconstruction with a bone graft; and,
D. the unilateral mandibular SSO approach, as in this case.

The intra-oral buccal approach may be a reasonable option for removal of superficially placed lesions at the angle of the mandible, but for larger and deeply placed lesions it will create a large bony defect, increasing the chances of intra- or postoperative mandibular fracture. It can also lead to exposure of the lingual aspect of the mandible, resulting in dysaesthesia of the ipsilateral tongue. The intra-oral lingual approach has the inherent risk of damage to the lingual nerve, and the access and visibility it provides will be limited. The extra-oral approach provides good access and visibility but increases morbidity by the added disadvantages of creating a cosmetic defect by cutaneous scarring and risk of damaging the mandibular branch of the facial nerve. In cases of segmental osteotomy where reconstruction with bone graft is required, the morbidity increases with the addition of a donor site, and there is a risk of graft failure, in contrast to other approaches.[14]

Trauner and Obwegeser introduced the intra-oral approach to SSO for the correction of dentofacial anomalies.[15] Rittersman and Van Gool initially used the SSO approach to enucleate a large, multinucleated keratocyst from the mandible in 1979 as stated earlier, and the technique was subsequently used by Barnard in 1983 to access and remove a large complex composite odontoma. The modification and evolution of the SSO technique has resulted in its proven safety and effectiveness in the removal of pathological mandibular lesions.

Radiographic imaging in this case showed that LR8 was displaced towards the lower border of the mandible in a buccal position, and the IAN was pushed inferiomedially by the tumour. The CT scan also revealed thinning of both cortices of the mandible by the expanding tumour, which affected the lingual cortex more than the buccal cortex. This made it crucial to preserve the buccal cortical plate in order to minimise the chances of a mandibular fracture intra- or postoperatively. A conventional approach, therefore, could not be applied in this case as it would involve the removal of a substantial amount of buccal bone in order to gain access to the tumour, thus putting the mandible at high risk of fracture. Thus, the SSO approach was utilised, which provided an excellent solution to this problem and enabled complete preservation of the buccal cortical plate. This access also facilitated extraction of the LR8 with minimal bone removal and prevented a fracture of the already thin buccal cortex. It provided superior access and direct visualisation of the tumour, which facilitated its excision, as well as clear identification of the IAN and subsequent protection, permitting a reduced risk of damage. However, it should be noted that it does not eliminate the risk of paraesthesia entirely, which still occurs in approximately 34% of cases four days following surgery, but persists in only 8% of cases six months post surgery. Patients report more dissatisfaction with the paraesthesia of the lingual tissues that occurs in the intra-oral lingual approach in comparison with that associated with SSO.[16]

Conclusion

Access for removal of large pathologies at the mandibular angle presents a unique set of challenges to the surgeon. Creating a large bony defect with conventional approaches will put the mandible at risk of intra- or postoperative fracture. The SSO approach is an ideal alternative to the conventional methods, providing excellent access and direct visualisation of the tumour, minimising the risk of a fracture by minimal bone removal, decreasing the risk of damage to the IAN by identification and protection of the nerve, and providing the possibility of attaining primary wound healing. The size and location of the cementoblastoma in this case made it ideal for the SSO approach to be applied to achieve a favourable outcome. This case

illustrates that the SSO approach can be safely utilised for excision of large cementoblastomas as well as other benign lesions at the angle of the mandible. We recommend that the SSO approach should be promoted for the excision of deeply placed mandibular lesions as compared to conventional approaches.

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CPD questions

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CPD

1. Cementoblastomas arise from organised cementoblast proliferation.

- ○ A: True
- ○ B: False

2. Radiographically, on orthopantomogram (OPG), a cementoblastoma typically presents as an area of radiodensity or mixed density with a rounded or sunburst appearance and a relatively radiolucent rim.

- ○ A: True
- ○ B: False

3. Saggital split osteotomy is a safe and effective technique for the removal of pathological mandibular lesions.

- ○ A: True
- ○ B: False