

Case Report

Surgical treatment of a rostral mandibular complex odontoma in a 3-year-old horse

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Summary

Complex odontomas are rare odontogenic tumours in horses comprised of a combination of mesenchymal and epithelial tissues. Examination, radiographic and histopathological findings in this patient all represent the typical behaviour of a complex odontoma. Oral tumours in horses may have treatment limitations due to tumour size and location. Similar to treatment in other species, surgical enucleation was curative for this type of tumour.

Introduction

Oral tumours are rare in equine patients and consequently there is a limited database for reference (Dillehay and Schoeb 1986; Pirie and Dixon 1993). Primary oral tumours are usually odontogenic, osteogenic or soft tissue in origin. Odontogenic tumours are usually found to consist of pure epithelial or mixed epithelial and mesenchymal tissues (Regezi *et al.* 2003a; Sowers and Gengler 2005). There are few reports of complex or compound odontomas in the horse, with these being primarily in young animals (Dillehay and Schoeb 1986; Dubielzig *et al.* 1986; Knottenbelt and Kelly 2005). These tumours are frequently not detected until they are advanced and life threatening (Dillehay and Schoeb 1986).

Case details

History

A 3-year-old, 614 kg Holsteiner filly was presented for evaluation of a hard, nonpainful, round-to-oval mass on the left mandible. The owners reported that the mass was slow growing and had been present for 9 months. The patient was not having any difficulty with mastication or displaying signs of

dysphagia. The horse was current on immunisations and had no concurrent medical conditions.

Clinical findings

General physical examination was within normal limits. Extraoral examination revealed a firm, smooth, round, raised mass on the buccal aspect of the left mid-ventral mandibular body, at the level of the second premolar tooth (Triadan 306). The mass measured 5 x 6 x 5 cm and was nonpainful on palpation. Intraoral examination failed to reveal any abnormalities. Tooth 706 appeared to be present and did not exhibit mobility or pathological periodontal pocketing. Radiographs made of the left hemimandible confirmed the presence of tooth 706 and that the mass was centred over the reserve crown of tooth 306. The mass consisted of a combination of radio-opaque rim material interspersed with lucent and mineralising opaque areas, similar to the radiodensity of enamel (**Figs 1 and 2**). Normal dental sac structures were noted on all mandibular cheek teeth. Differential diagnosis for the mass included: periapical abscessation, cyst, ossifying fibroma, malignant neoplasia (invasive squamous cell carcinoma and myxomatous tumours) (Gibbs 2005; Knottenbelt and Kelly 2005) and odontogenic tumour (ameloblastoma, ameloblastic odontoma, complex odontoma) (Gibbs 2005; Knottenbelt and Kelly 2005). Surgical staging (biopsy) under general anaesthesia was recommended to provide a definitive diagnosis before evaluating treatment options, but was declined. Biopsy with surgical enucleation and debulking was discussed and elected.

Treatment

Preoperative medications included i.v. antibiotics: crystalline penicillin¹ (22,000 iu/kg bwt) and gentamicin² (6.6 mg/kg bwt) and phenylbutazone³ (4.4 mg/kg bwt). The horse was restrained under general anaesthesia and placed in right lateral recumbency (**Fig 3**).

A 10 cm long, U-shape curvilinear skin incision was created around the mass, with the convexity directed ventrally, in order

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to preserve the cutaneous blood supply and establish drainage if necessary. Soft tissue dissection permitted elevation and retraction of the superficial buccal portion of the buccinator muscle and localisation and avoidance of the mental foramen



Fig 1: Preoperative lateral skull radiographs depicting the mandibular mass centered over the left mandibular second premolar (tooth 306).

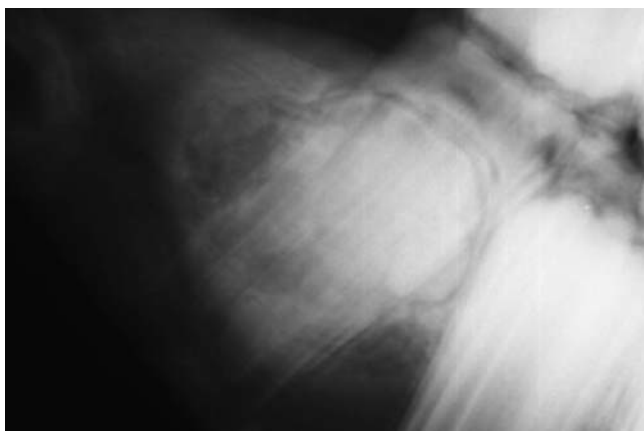


Fig 2: Preoperative close-up evaluation of left mandibular mass. Note the interspersed radio-dense material.



Fig 3: Anaesthetised patient positioned in right lateral recumbency. The mandibular mass has been shaved in preparation for surgery.

and its associated neurovascular bundle (Pasquini *et al.* 1991). An osteotome was used to create a 2 cm square window through the buccal cortical bone, revealing a cystic cavity filled with scant amounts of yellow-tinged, viscous fluid, fragments of suspended soft tissue and pale, white mineralised tissue. The wall of the lesion was continuous, forming a fibrous covering that extended as a solitary lesion beneath all edges of the fenestration. A 5 x 6 cm buccal osteotomy was performed to provide a fenestration capable of removing the entire mass without sectioning (**Fig 4**). Once the plate of cortical bone was incised, the mass was elevated and removed *en bloc* (**Fig 5**). The cavity was debrided with a curette, then copiously lavaged with sterile saline, revealing a thin plate of alveolar bone covering the reserve crown and roots of tooth 306.

The cavity was packed with providone-iodine soaked gauze which was exteriorised at the most ventral margin of the incision. Subcutaneous tissues were apposed with 2-0 polyglactin 910⁴ in 2 segments, leaving a 2 cm gap ventrally for drainage and gauze packing removal (Gaughan 1998). The skin edges were apposed in a similar manner. The patient recovered uneventfully from general anaesthesia and was

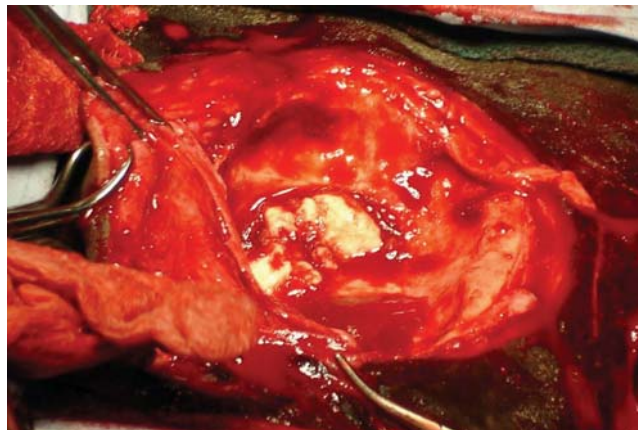


Fig 4: Intraoperative view and exposure of the mandibular mass after fenestration. Note the pale appearance of mineralised tissue.



Fig 5: Removal of the mandibular mass overlying tooth 306. The lesion was able to be separated from buccal cortical bone and removed as a single mass.

treated post operatively with oral phenylbutazone (1.5 mg/kg bwt *per os* b.i.d.) and trimethoprim sulphamethoxazole (25 mg/kg bwt *per os* b.i.d., for 14 days).

The gauze packing was removed 18 h after surgery. A topical adhesive bandage was then placed over the drainage site and was replaced daily for 3 days. Clear to yellow-tinged serous drainage decreased daily. Post operative radiographs confirmed complete mass removal (**Fig 6**).

Histopathological findings

The tissue examined contained abundant odontogenic epithelium and stellate reticulum. Woven eosinophilic, extracellular matrix with many empty lacuna were present, resembling necrotic cementum (**Fig 7**). Scattered throughout the stroma were nests of epithelial rests and chords of epithelial cells showing early differentiation into ameloblastic cells. Occasional small islands of chondroid-like mineralising material, thought to be abnormal enamel matrix, were seen surrounded by elongate cells (ameloblasts). The histopathological description was consistent with a complex

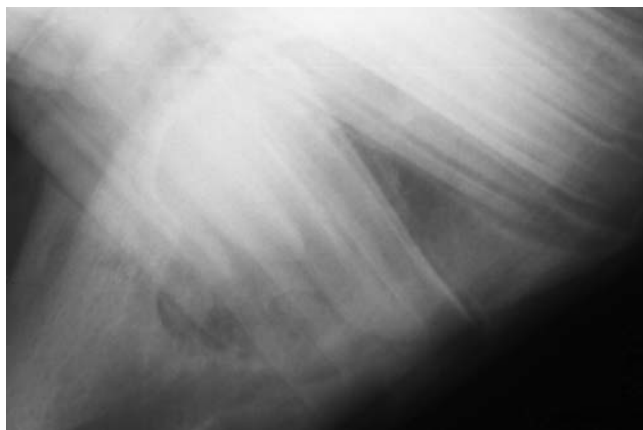


Fig 6: Post operative evaluation of the surgical site 48 h after surgery. Lack of radio-opaque material demonstrates complete removal of the gross lesion.

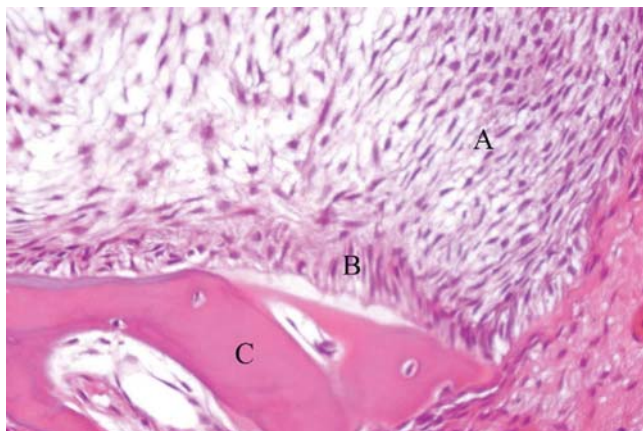


Fig 7: Histopathological evaluation of oral mass consistent with a complex odontoma. A. stellate reticulum, B. odontogenic epithelium, C. cementum.

odontoma. Immunohistochemistry was performed on the sample to evaluate for the epithelial and mesenchymal cell populations. Positive cytokeratin staining characterised both the epithelial cells rests and the stellate reticulum (Regezi *et al.* 2003b). Positive vimentin staining was seen in stellate reticular cells (Regezi *et al.* 2003b). The diagnosis of complex odontoma was made because of the presence of well differentiated odontogenic tissues of both epithelial (ameloblast and stellate reticulum) and mesenchymal (cementum and pulp) origin (Dubielzig *et al.* 1986; Regezi *et al.* 2003a,b).

Additional clinical findings

Approximately 6.5 months after surgical excision, the patient presented for oral and radiographic re-evaluation. Oral examination revealed the occlusal surface of tooth 306 to be level to the gingival margin with a remnant of the dental cap present and loosely attached to the buccal gingiva. Tooth 306 was found to be under erupted compared to the contralateral tooth (406) and the remaining left mandibular cheek teeth. A smooth, firm bony swelling over the surgery site was palpable and suspected to be associated with osseous remodelling. Deciduous dental caps remained on teeth 306 and 308, (but not 307), confirming delayed eruption of tooth 306 (**Fig 8**). The apex of tooth 306 remained similarly rounded compared to the pre- and post operative radiographs. Radiographs made of the region revealed bone remodelling of the surgical site, with no indication of tumour recurrence.

Discussion

Since oral tumours in the horse are rare, our experience with treatment is somewhat limited. Most oral tumours look similar grossly, and require histopathological examination for differentiation (Knottenbelt and Kelly 2005). Symptoms and clinical signs associated with oral tumours are not specific to horses. Dysphagia, weight loss, oral bleeding, grossly abnormal

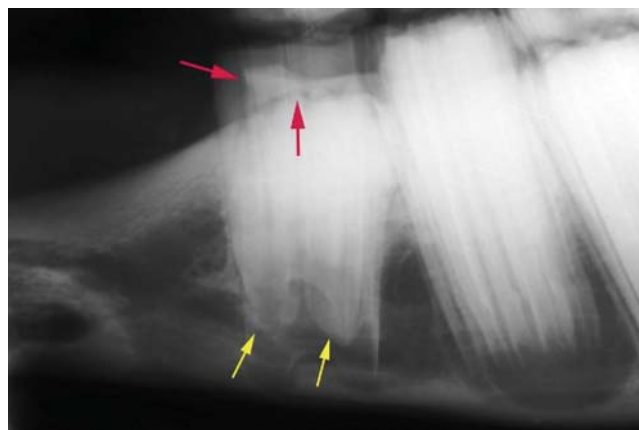


Fig 8: Six month recheck evaluation. Close evaluation demonstrates blunting of the apices of tooth 306 (yellow arrows). The left deciduous second premolar tooth (dental cap) remains present (red arrows). Contralateral tooth 406 can be seen superimposed over tooth 306 with normal apices and a clinical crown of normal height.

tissue, facial asymmetry and respiratory distress are all variable clinical signs of equine, canine or feline patients (Wiggs and Lobprise 1997; Knottenbelt and Kelly 2005). Delayed detection in equine patients may limit treatment options, even in instances of benign lesions of untreatable size (Knottenbelt and Kelly 2005). In a limited number of cases involving large or invasive lesions, radical mandibular resection and stabilisation with orthopaedic bone plates and *type II* external fixation have been used (Beard 1999). Mandibular fracture itself serves as a potential complication of surgical treatment (Brounts *et al.* 2004). Diagnostic imaging and biopsy remain the mainstays of diagnosis and treatment planning (Knottenbelt and Kelly 2005).

Imaging modalities used in diagnosing oral and dental tumours include radiography, computed tomography, gamma scintigraphy and ultrasonography for detailing soft tissue masses (Knottenbelt and Kelly 2005). Radiography was chosen for its accessibility, ability of producing diagnostic images of the rostral mandible and ease of obtaining images under sedation (Gibbs 2005).

Biopsy offers the most reliable method of tumour diagnosis (Knottenbelt and Kelly 2005). Fine needle aspirates are frequently performed in the field because they are easy to perform and are relatively atraumatic. With such a limited number of cells able to be collected, fine needle aspirates may not always confirm a diagnosis (Knottenbelt and Kelly 2005). Other biopsy techniques include hollow track (Trucut), wedge or sectional biopsies. Excisional biopsy is usually performed under general anaesthesia. Although this technique risks contamination of the surgery site with tumour cells, it may demonstrate excellent results with complete excision (Knottenbelt and Kelly 2005). The cystic nature of the lesion radiographically and the owners' reluctance to undergo multiple procedures made excisional biopsy the patient's best chance for a curative procedure under a single anaesthetic episode.

Odontomas are mixed odontogenic tumours made up of epithelial and mesenchymal dental tissues (Regezi *et al.* 2003a; Sowers and Gengler 2005). These tumours are biologically classified as hamartomas (proliferation of normal tissue with abnormal organisation) instead of neoplasms (Dubielzig *et al.* 1986; Crabill and Schumacher 1998; Regezi *et al.* 2003a). The radiographic and histopathological commonalities of complex odontomas extend across species lines. Histopathologically they display normal appearing enamel, dentin, cementum and occasionally pulp in a chaotic arrangement (Dillehay and Schoeb 1986; Dubielzig *et al.* 1986; Lobprise and Lemmons 2007). Radiographically these tumours typically appear as solid opacified lesions with similar radiodensity to teeth (Regezi *et al.* 2003a; Knottenbelt and Kelly 2005). Radiographic evaluation of mandibular tumours in horses should not solely be used to conclusively identify the specific type of tumour (Pirie and Dixon 1993) but may provide some diagnostic value. Biologically, complex odontomas have limited growth potential but may reach significant size (Regezi *et al.* 2003a). Cases of complex odontomas in horses have been described to include the presence of multilobulated masses within a well-differentiated cyst-like structure, consistent with this patient's radiographic findings (Dillehay and Schoeb 1986; Dubielzig

et al. 1986; Knottenbelt and Kelly 2005). Tumour growth may be locally aggressive but does not metastasise (Wiggs and Lobprise 1997) and complete enucleation is curative with rare recurrence (Regezi *et al.* 2003a; Knottenbelt and Kelly 2005). Compound odontomas demonstrate similar histological findings except that epithelial and mesenchymal derived tissues are organised into recognisable (normal or abnormally appearing) tooth structures (Dillehay and Schoeb 1986; Wiggs and Lobprise 1997; Knottenbelt and Kelly 2005). Odontomas tend to be reported in younger animals including: equine, canine, feline and human patients (Wiggs and Lobprise 1997; Regezi *et al.* 2003a; Brounts *et al.* 2004). They are considered rare in these species and may only contribute 0.05% of biopsy specimens in dogs (Sowers and Gengler 2005). It has been suggested that odontogenic tumours may be more common in horses than other species (Knottenbelt and Kelly 2005).

Ameloblastoma was a major differential diagnosis for the lesion in this patient. These lesions are also considered to be rare tumours of domestic animals and man (Dillehay and Schoeb 1986). Ameloblastomas are defined by containing large amounts of odontogenic epithelial tissue (Dillehay and Schoeb 1986) and lack inductive differentiation of dentin and enamel (Crabill and Schumacher 1998; Knottenbelt and Kelly 2005). Epithelial tissues giving rise to ameloblastomas may originate from the enamel organ, rests of Malassez, reduced enamel epithelium or the epithelial lining of odontogenic cysts (Regezi *et al.* 2003a). These oral tumours are more common in the mandible of older horses, but have been reported in foals (Crabill and Schumacher 1998) and frequently contain a central cystic region (Wiggs and Lobprise 1997; Knottenbelt and Kelly 2005). Radiographically, lesions tend to be osteolytic and uni- or multilocular (Crabill and Schumacher 1998; Regezi *et al.* 2003a). Aggressive surgical treatment with wide excisional margins is commonly thought to be the only potential curative procedure (Bjorling *et al.* 1987; Knottenbelt and Kelly 2005) due to the tumour cells' ability to invade bone (Crabill and Schumacher 1998). If excision is not complete, prognosis is poor and slow regrowth may be anticipated (Bjorling *et al.* 1987; Regezi *et al.* 2003a; Knottenbelt and Kelly 2005). Surgical excision remains the treatment of choice since treatments such as adjunctive radiotherapy (Knottenbelt and Kelly 2005), interlesional chemotherapy (De Cock *et al.* 2003) or sole treatment with curettage (Regezi *et al.* 2003a) all provide poor results.

Various subsets of ameloblastomas have been reported. Ameloblastic odontomas tend to be radiolucent or partially mineralised and may contain foci of enamel (Knottenbelt and Kelly 2005). Keratinising ameloblastomas develop large amounts of epithelial keratin throughout the lesion (Knottenbelt and Kelly 2005). Ameloblastic carcinomas demonstrate a high nucleus/cytoplasm ratio, high mitotic index and differentiation toward ameloblastic epithelium (De Cock *et al.* 2003). Common to the entire subset of ameloblastomas are the predominance of odontogenic epithelium and the necessity for wide surgical excision for treatment (Knottenbelt and Kelly 2005). Immunohistochemical staining aids in differentiating complex odontoma from ameloblastoma by positively identifying mesenchymal tissues (Regezi *et al.* 2003b).

Delayed eruption of the reserve crown of tooth 306 and possible devitalisation secondary to the oral mass or excisional treatment may necessitate future exodontia.

Equine exodontia is technically challenging, requires specialised equipment and historically has a high rate of complication (Tremaine and Lane 2005). In this case, comparison of preoperative and 6-month follow-up radiographs remained remarkably similar with an increased blunting of the periapical tissue of tooth 306 suggesting premature apical closure with possible devitalisation and arrested eruption (Tremaine and Lane 2005). Eruptive dates and expected periods of deciduous tooth exfoliation confirmed the abnormal eruption of tooth 306. The clinical crown height flush with the gingival margin, presence of a portion of the retained dental cap (deciduous tooth 706) and confirmed exfoliation of the deciduous tooth overlying 307 all support abnormal development of tooth 306. Exodontia may be required if 306 develops signs of apical abscessation or progressive periodontal disease (Tremaine and Lane 2005).

Odontogenic tumours in horses are rare, with few complex odontoma cases having been reported (Dubielzig et al. 1986; Pirie and Dixon 1993). The chaotic, disorganised manner in which the stellate reticulum and odontogenic epithelium were arranged are consistent with complex, rather than compound odontomas (Dubielzig et al. 1986; Regezi et al. 2003a). Recognisable, mineralised cementum within the specimen demonstrates inductive qualities of mixed mesenchymal-epithelial tumours, histological findings absent in ameloblastomas. This case demonstrates typical pathological findings consistent with a complex odontoma. Signalment, history, physical examination findings, radiographic findings, location of the lesion, histopathological findings and post operative tumour-free interval of at least 6 months are all supportive of a complex odontoma in this horse. As more odontogenic tumours are reported in equine patients, the database of histopathological descriptions will continue to grow.

Manufacturers' addresses

¹Fort Dodge Laboratories, Fort Dodge, Iowa, USA.

²Schering-Plough Animal Health Corporation, Kenilworth, New Jersey, USA.

³Phoenix Pharmaceuticals, St. Joseph, Missouri, USA.

⁴Ethicon Inc, Johnson & Johnson, Piscataway, New Jersey, USA.

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