

Partial Resection of Maxillary Ossifying Fibroma in a Thoroughbred Stallion

Sang-Kyu Lee^{***}, Byung Hyun Kim^{***}, Richard Luong^{****}, Bok-Sun Jung^{**},
Hyung-Ho Im^{**}, Jeonghun Lee^{***}, Eo-Jin Im^{***} and Inhyung Lee^{*1}

**Department of Veterinary Clinical Sciences, College of Veterinary Medicine, Seoul National University, Seoul 08826, Korea*

***KRA Equine Hospital, Korea Racing Authority, Gwacheon 13822, Korea*

****Busan KRA Equine Hospital, Korea Racing Authority, Busan 46745, Korea*

*****IDEXX Laboratories, West Sacramento 95605, USA*

(Received: January 27, 2018 / Accepted: June 08, 2018)

Abstract : A 5-year-old Thoroughbred race horse was presented to Busan Korea Racing Authority equine hospital with a 3-year history of a slow-growing left rostral maxillary mass. The location and progressive growth of the mass eventually resulted in poor food prehension, quidding and mouth bit placement. The mass was solitary and hard, and covered by normal smooth oral mucosa. Radiographic examination of the maxillae showed a flocculated and mixed radiolucent lesion protruding outward and displacing the 202 and 203 teeth caudally. The 202 tooth was in normal size and the 203 tooth was hypoplastic on radiography. Under general anesthesia, a partial surgical resection of the mass was performed to minimize functional loss and facilitate prompt return to track. After surgery, there was improvement in food intake, mouth bit placement, and cosmetic appearance. Histopathological examination determined the resected maxillary mass to be an ossifying fibroma. However, there was continued growth of remnant mass in the maxilla. Equine ossifying fibroma is a rare condition and primarily affects the rostral mandible, and less commonly, the maxillae. In this case, the lesion was slow-growing, and caused cosmetic and functional impairments, including poor food intake and reduced trainability. Surgical resection was performed, but the effect of treatment was limited due to advanced size / stage of the tumor. Early dental care is suggested for horse owners to prevent belated identification and improve successful treatment of oral disorders like ossifying fibroma.

Key words : Ossifying fibroma, Thoroughbred, Partial resection, Maxillae.

Introduction

Oral neoplasia is rare in the horse and can be associated with teeth (odontogenic), bone (osteogenic), or soft tissues (4,9). Most are not identified until in an advanced state, and selection of treatment methods is often limited due to late detection (4). The gross appearance of equine oral neoplasms is very similar and definitive diagnosis is made with histological examination (4). Due to an overall rare incidence of these conditions and limited number of reports, it is difficult to decide the most optimal treatment with respect to specific tumor type and prognostic considerations.

Ossifying fibroma is one of type of oral neoplasia in horses, and is mostly reported in young horses (4,5,8,10). Ossifying fibroma is a proliferative, fibro-osseous, tumor-like lesion with predilection for the rostral mandible in horses (4,5).

The purpose of this report is to describe the unusual presentation of ossifying fibroma in maxillae in a 5-year-old Thoroughbred race horse with clinical signs and the result of partial resection as a compromise treatment.

Case

History

A 5-year-old stallion, Thoroughbred race horse was presented to Busan Korea Racing Authority Equine hospital with 3-year history of a slow-growing mass in the left rostral maxillae. The owner reported that left incisor aspect was hard and swollen, with absence of the 201 and 202 teeth, and a small 203 tooth. The horse was bought at the age of 2 years at an overseas auction sale, so the exact onset of the lesion was unknown but assumed to be earlier than two years. Reduced trainability by due to poor mouth placement of a bit was noted. Food intake was affected, noted as prolonged time to eat, quidding and saliva dribbling while eating. The owner sought veterinary intervention to resolution of the condition with return to trainability and rapid recovery.

Clinical findings

A 7 × 3 × 3 cm mass around the gingival tissue of the upper left incisors was identified (Fig 1). The mass was solitary, firm, and large enough to have protruded beyond the left upper lip, interfering with prehension and biting. Ultrasonography revealed a hyperechoic bone like mass in the underlying gingival mucosa. On radiographs, there was a protruding heterogeneous opacity mass in the left rostral maxillae, displacing 202 caudally and 203 laterally (Fig 2). No other abnormal

¹Corresponding author.
E-mail : inhyunglee@snu.ac.kr



Fig 1. Enlargement of left rostral maxillary mass on admission. The mass was protruded laterally out of lips in normal closure of mouth (A, arrow heads). The mass (arrow heads) was covered with normal oral mucosa, and 202 tooth was missing and 203 tooth (arrow) was hypoplastic (B). The lips had been pulled out by horse handler's hands.

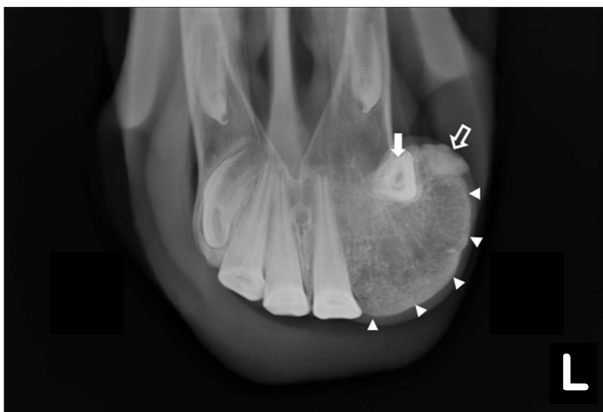


Fig 2. Dorsoventral intraoral radiograph of rostral maxillary mass on admission. Heterogeneous opacity mass (arrow heads) was located on left maxillary. There were caudally detrued 202 tooth (white arrow) in the mass and hypoplastic 203 tooth (black arrow).

lesions were identified in the oral cavity. No other abnormalities were observed on physical examination. Hematologic and serum biochemistry test results were within normal ranges except for high total bilirubin (5.1 mg/dL; reference interval: 0.5-2.3 mg/dL). A tentative diagnosis of ameloblastoma, osteoma, fibrous dysplasia, and ossifying fibroma was made based

on location, gross appearance, and diagnostic imaging results. Histologic examination with surgical intervention of the tumor was recommended for treatment and prognostication.

Surgical technique

Initially, full hemimaxillectomy was recommended to eliminate the recurrence of the tumor, but the owner was concerned with a poorer post-surgical outcome, related to worsened food prehension and loss of incisor support for proper mouth bit placement, which may ultimately cause reduced athletic performance. Subsequently, partial maxillectomy was elected instead.

No abnormal results were found in preoperative complete blood count and serum chemistry tests. Phenylbutazone (4 mg/kg intravenously; IV), procaine penicillin (22,000 U/kg intramuscularly; IM) and gentamicin (6.6 mg/kg IV) were administered as preoperative treatment. After sedation with detomidine (0.02 mg/kg IV), general anesthesia was induced with diazepam (0.1 mg/kg IV) and ketamine (2.2 mg/kg IV) and maintained with isoflurane in oxygen and positive pressure ventilation through an endotracheal tube in dorsal recumbency. The surgical site was cleaned with a 0.05% chlorhexidine solution. Left infraorbital nerve block was performed with 5 mL of 2% lidocaine. A mouth gag made of plastic pipe was placed in the right aspect of incisors. A horizontal incision over the gingival mucosa of the left maxillary mass was made and undermined to separate the mass from the mucosa. Using an oscillating saw, the exposed mass was resected in manner to restore overall normal contours of the left rostral maxillae. The oral mucosa was reapposed to cover resected aspects of mass with vertical interrupted mattress sutures. The patient recovered from anesthesia uneventfully. The resected left maxillary surgical specimen was immersed in 10% neutral buffered formalin and histological examination was performed.

Gross and microscopic findings

Gross examination of the formalin-fixed resected mass revealed a 7 × 3 cm, pink, firm and cancellous bony specimen. Portions of the resected mass were transected into smaller pieces for decalcification. The decalcified subsamples were then processed for routine light microscopic examination of hematoxylin and eosin-stained sections.

Microscopically, the neoplasm consisted entirely of a fibroosseous proliferation characterized by abundant anastomosing trabeculae of new woven bone, the surface of which was lined by a single layer of osteoblasts (Fig 3). The woven bone was supported by a moderate amount of irregular, loose, collagenous stromal tissue that contained regularly distributed plump reactive fibroblasts (Fig 3). The transition between the fibrous and osseous components was abrupt. Based on histologic examination, this mass was determined to be an ossifying fibroma.

Post-operative care

Procaine penicillin (22,000 U/kg IM) and phenylbutazone (4 mg/kg IV) were administered for 7 days after surgery. The horse received 4 meals a day, made of water soaked feed and finely cut grass. The mouth was rinsed with diluted chloro-

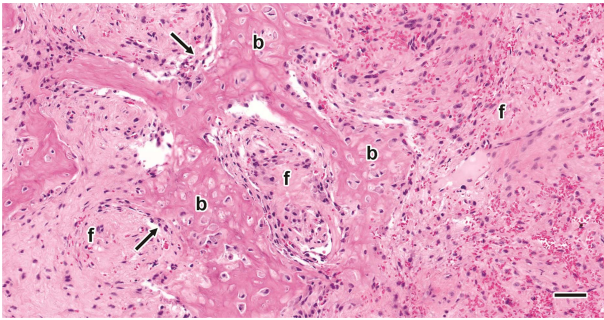


Fig 3. Photomicrograph of biopsy of maxillary mass containing an ossifying fibroma. The fibrous component proliferation consisted of disorganized fibroblasts that produced large amounts of collagenous tissues (f). The fibroblasts transformed into osteoblasts that produced the osseous component, represented by anastomosing trabeculae of woven bone (b). Some of the osteoblasts existed as a single layer of cells that palisade along the surface of the bony trabeculae (arrows). H&E stain. Scale bar = 50 μ m.

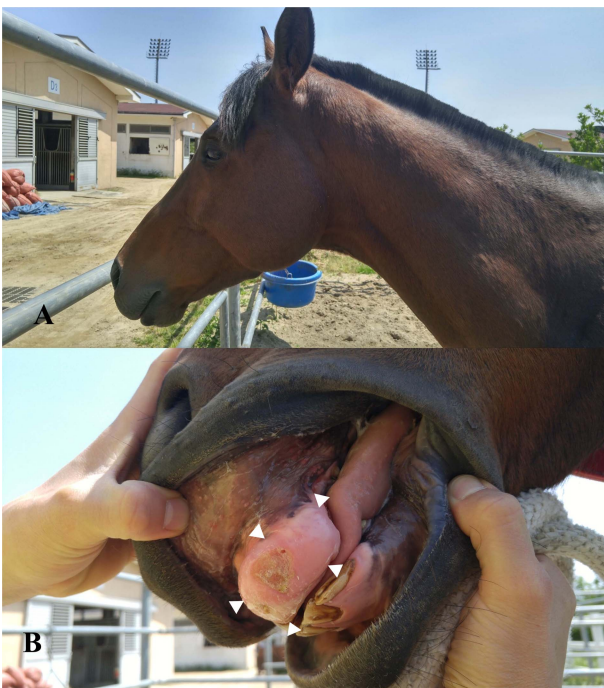


Fig 4. Photograph of the horse taken 10 weeks after surgery. The lips are closed normally (A). The regrowth of the mass was presented on surgical site (B. arrow heads). The lips had been pulled out by horse handler's hands.

hexidine solution after each feeding to prevent post-surgical infection. Vitality and appetite were not impaired by the surgery. Dehiscence of the suture site occurred 6 days after surgery, and sutures were removed. Continuous and frequent mouth lavage was recommended.

At 2-weeks post-surgery, the mucosa apposed partially and there was contraction of the exposed surgical site. At 10-weeks post-surgery, regrowth and protrusion of the mass were noted (Fig 4). Again, hemimaxillectomy was recommended to prevent recurrence of the neoplasm, but the owner

declined further surgical intervention due to financial reasons and uncertainty of return to full athletic performance.

Discussion

Ossifying fibroma is a benign, proliferative, fibro-osseous tumor originated from soft tissues (4). Ossifying fibroma is a rare condition in humans and animals, and amongst animals, is most commonly recognized in young horses (4,8,10). Ossifying fibroma has also been reported in the dog, cat, rabbit, llama, greater kudu, sheep, and goat (7,14). It is commonly reported to affect the mandibular region in humans (13). Similarly, the lesion has been reported to mainly affect the rostral mandible in young horses (11,15), although a single case of ossifying fibroma in the rostral maxillary in an older horse was reported recently (6). Although there is a female sex predilection in humans (13), no sexual predilections have been reported in animals. This lesion may distort normal bone shape, displace teeth, cause tooth loss, and result in difficulty of prehension / mastication (14) and inability to place a bit in the mouth.

It is thought that the development and occurrence of ossifying fibroma may be related to an abnormal intramembranous ossification of bone, defect with dental eruption, and/or trauma (5,8). Recently, a genetic mutation was discovered as a cause in humans (2).

Definitive diagnosis was made based on clinical, diagnostic imaging, and histologic features. Clinically, this large unilateral maxillary tumor was covered by normal oral mucosa, and resulted in dysphagia due to gross distortion of the lips and incisor teeth. Radiographically, the mass presented as a dense opacity mass with calcification. Histologically, the mass was a fibroosseous proliferation consisting of new woven trabecular bone lined by osteoblasts and irregular collagenous stroma populated by plump reactive fibroblasts. There was abrupt transition from fibroblastic stroma to osteoblasts in this case (4). This case was diagnosed as ossifying fibroma.

Surgical removals, including variations of mandibulectomy and maxillectomy, have been frequently described for the treatment of equine oral tumors (15). Specifically, mandibular ossifying fibromas can be successfully treated cases with rostral mandibulectomy, with literature reporting good results and little surgical complications (1,8). Successful outcome has even been documented with standing mandibulectomy (1). However, fewer cases of maxillectomy for the treatment of ossifying fibromas have been reported (6,12,15), and is considered more complicated because of the care and precision required to preserve major palatine and incisive arteries penetrating the rostral maxillae, as well as the need for recumbency with general anesthesia (1). While Cousty and Tricaud reported one case of successful hemimaxillectomy resulting in good outcome with respect to food prehension (1), functional deficits related to maxillectomy should be considered for an athletic horse : e.g., such as the placement of a mouth bit for training; adequate food intake.

In this case, full rostral hemimaxillectomy was advocated as the surgical treatment of choice to best prevent recurrence of the lesion. However, other track performance considerations related to mouth bit placement and food intake were

taken into account in this case, which lead to the owner electing for a partial resection of the neoplasm instead. The owner was cautioned regarding a less successful outcome with partial maxillectomy, in terms of potential neoplasm recurrence depending on the ultimate histologic diagnosis. The resected neoplasm was submitted to histological examination and diagnosed as ossifying fibroma, and therefore local recurrence was highly probable due to the partial maxillectomy approach.

Surgical excision, radiation using cobalt-60 teletherapy and surgical excision followed by adjuvant radiation therapy were reported for successful ossifying fibroma treatment (4,15). Robbins *et al.* reported a case of mandibular ossifying fibroma treatment case in young horse using by cobalt 60 radiation (10). Unfortunately, there was no locally or regionally available radiation therapy facility for this equine patient, and thus adjuvant radiation therapy could not be provided after surgical intervention in this case.

Throughout ten-week postoperative check, recurrence of the neoplasm was confirmed and fast growth of remnant neoplasms was noted. It was likely that the fast post-surgical growth was a consequence of surgery-related elaboration of angiogenic cytokines, which promoted remaining tumor cells/tissue growth (3).

Ossifying fibroma is a benign, fibro-osseous mass which mainly develops in the rostral regions of the mandible of young horses (8,15). However, the lesion is prone to be identified late such as in this case at the advanced stage because of deficient oral care and underestimated significance of oral tumors. Oral care should be provided appropriately to young horses, which may help identify oral lesions like ossifying fibroma earlier, and therefore facilitate more appropriate and allow for more successful treatment at the earlier stages, and minimizing functional deficits and complications. More extensive excision and adjuvant radiation therapy may be required at more advanced stages of the lesion to minimize unacceptable cosmetic and functional deficit (4,11).

References

1. Cousty M, Tricaud C. Resection of a premaxilla and rostral portion of the maxilla in a horse. *Equine Vet Educ* 2014; 26: 269-273.
2. Dominguet PR, Meyer TN, Alves FA, Bittencourt WS. Juvenile ossifying fibroma of the jaw. *Br J Oral Maxillofac Surg* 2008; 46: 480-481.
3. Karl HB, Struikmans H, Barten-Van Rijbroek AD. Surgical stress and accelerated tumor growth. *Anticancer Res* 2008; 28: 1129-1132.
4. Knottenbelt DC, Kelly DF. Oral and dental tumors. In: *Equine dentistry*. Edinburgh: Elsevier Saunders. 2005: 127-143.
5. Knottenbelt DC, Pascoe RR. Conditions of the alimentary tract. In: *Color atlas of diseases and disorders of the horse*. Edinburgh: Mosby. 1994: 28.
6. Lechartier A, Steele E, Vallefucio R, Mespoulhès-Rivière C. Resection of the incisive bone and rostral maxillae for removal of an ossifying fibroma in an 18-year-old warm-blood gelding. *Equine Vet Educ* 2015; 27: 574-578.
7. Miller MA, Towle AM, Heng G, Greenberg CB, Pool RR. Mandibular ossifying fibroma in a dog. *Vet Pathol* 2008; 45: 203-206.
8. Morse CC, Saik JE, Richardson DW, Fetter AW. Equine juvenile mandibular ossifying fibroma. *Vet Pathol* 1988; 25: 415-421.
9. Pence P, Wilewski K. Mature horse dentistry. In: *Equine dentistry: a practical guide*. Philadelphia: Lippincott Williams and Wilkins. 2002: 161-162.
10. Robbins SC, Arighi M, Ottewell G. The use of megavoltage radiation to treat juvenile mandibular ossifying fibroma in a horse. *Can Vet J* 1996; 37: 683-684.
11. Sanchez LC. Disorders of the gastrointestinal system. In: *Equine internal medicine*, 3rd ed. St. Louis: Saunders. 2010: 895.
12. Schumacher J, Kemper DL, Helman RG, Edwards JL. Removal of the premaxillae and rostral portions of the maxillae of a horse. *J Am Vet Med Assoc* 1996; 209: 118-119.
13. Slootweg P. Fibro-osseous lesions. In: *Pathology of the maxillofacial bones: A guide to diagnosis*. Cham: Springer. 2015: 132-133.
14. Whitten KA, Popielarczyk MM, Belote DA, McLeod GC, Mense MG. Ossifying fibroma in a miniature rex rabbit (*Oryctolagus cuniculus*). *Vet Pathol* 2006; 43: 62-64.
15. Witte S. Maxillectomy and mandibulectomy in the horse: Indications and necessity of post-operative adjunct therapy. *Equine Vet Educ* 2014; 26: 274-279.