Histopathological Characteristics of an Ossifying Fibroma Formed in the Maxilla of a Racehorse

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A 1-year-old male thoroughbred racehorse experienced swelling of the left upper lip. The swelling was attributable to enlargement around the incisive bone of the interdental space posterior to the third incisor in the left maxilla. Even after two operations to reduce the bulk of the mass, it continued to increase in size. Dyspnea caused by stenosis of the nasal cavity forced us to perform euthanasia, and a pathological examination was conducted. Macroscopic examination of a section of the mass revealed the formation of multiple areas of solid fibrous tissue, and trabeculae within the incisive bone which had displaced the cortical bone. On histology, the mass was composed of trabecular bone-like structures due to the proliferation and aggregation of fibroblasts. Therefore, we diagnosed it as an ossifying fibroma. Equine ossifying fibroma is characterized by development in the mandible, but was formed in the maxilla in this case. Equine ossifying fibroma has not been reported previously in Japan. This is the first case of equine ossifying fibroma identified in Japan.

Key words: bone, horse, maxilla, ossifying fibroma

In the World Health Organization International Classification of Tumors of Domestic Animals, among the tumors formed in the bones of the upper alimentary system and in bones and joints, osteoma, ossifying fibroma, myxoma, and chondroma are classified as benign, while osteosarcoma, chondrosarcoma, and multilobular bone tumor are classified as malignant [3, 9]. In addition, fibrous dysplasia and craniomandibular osteopathy are classified as tumor-like lesions [3]. Osteoma, ossifying fibroma, and fibrous dysplasia, which often develop in horses, should be diagnosed carefully because of their similar histopathological morphology [9]. Ossifying fibroma has been reported in horses, cattle, cats, dogs, antelope, and sheep, and occurs at high incidence in the mandibles of young horses, [1, 2, 4–6, 8, 11]. Nevertheless, there have been no reports of equine ossifying fibroma in Japan. Here, we summarize a rare case of a horse with an ossifying fibroma formed in the left incisive bone of the maxilla that was examined histopathologically.

A male thoroughbred horse born in Hokkaido first experienced swelling of the left maxilla at the age of 1 year. Over the following 3 months, the swelling became more conspicuous, and caused serious stenosis of the left nasal cavity, which was treated by surgery to partially reduce the enlarged bone tissue. However, over the next 9 months, the left maxilla again swelled gradually, leading to dyspnea associated with obstruction of the left nasal cavity. The horse also had difficulty in chewing, owing to considerable gingival enlargement between the incisor and premolar of the left maxilla (Fig. 1). Radiography revealed that the mass was relatively solid and contained radiopaque trabecular structures (Fig. 2). Another operation was performed to remove the tissues that had formed within the enlarged incisive bone, but the mass was too large to...
remove completely. The left maxilla rapidly enlarged again over the following 3 months. Dyspnea caused by obstruction of the nasal cavity forced us to perform euthanasia with an intravenous injection of an excess of sodium thiopental, and post mortem examination was performed.

Fig. 1. Clinical appearance of the case. A severely swollen left upper lip can be seen (before the second operation).

Fig. 2. Radiograph of the maxilla. Dilatation can be seen in the regional bone of and around the interdental space between the left third incisor and the maxillary bone. Sections 5 mm thick were prepared by slicing in a frontal direction at the arrowed site and were examined histopathologically.

Fig. 3. Macroscopic findings on a frontal-direction slice of the mass (arrows in Fig. 2). The tissue appears to be a mixture of soft areas showing gelatinous degeneration (black arrow) and areas with thickened or increased cancellous bone (red arrows).

Fig. 4. Histopathological findings regarding the mass. Trabeculae of woven bone are embedded in the fibrous tumor tissue. (Hematoxylin-eosin stain).

Fig. 5. High magnification image of the mass. Fibroblast-like cells are aggregated to form a trabecular structure. Numerous osteoblast-like cells adjoining or shifting from fibroblasts can be seen around the trabeculae. (Hematoxylin-eosin stain).
performed.

Macroscopic examination revealed a palm-sized mass in the incisive bone between the incisor and premolar of the left maxilla. The mass was white and indurated. In the frontal direction, in an excised portion of the interdental space of the enlarged incisive bone posterior to the left third incisor, an area of gelatinous soft tissue and another area of thickened or increased cancellous bone were observed (Fig. 3). Soft X-ray imaging of the excised portion of the interdental space of the enlarged incisive bone showed no bony structures in the area of the gelatinous tissue, while in the surrounding solid area that contained cancellous bone, thickening and multiplication of the trabeculae of the cancellous bone could be seen. However, the compact bone of the incisive bone was thinned by the proliferating mass.

On histopathology, the proliferating mass within the enlarged incisive bone was seen to consist of both fibrous and osseous elements (Fig. 4). In the fibrous area, proliferating fibroblast-like spindle cells which had oval middle-sized nuclei and fusiform slightly basophilic cytoplasm were seen in combination with collagen fibers and blood vessels. Few mitotic figures were observed.

In the osseous elements, there were irregularly shaped osteoid spicules and woven bone rimmed with osteoblasts. The fibroblast-like spindle cells underwent transformation to osteoblasts along the margins of the developing bone spicules (Fig. 5). Cartilage and necrosis were not observed in the mass. Osteoclasts were clearly observed at the trabecular margin. The fibroblast-like cells were partially aggregated, and some mild calcification was observed. In addition, the marginal trabeculae had numerous osteoclasts.

The gelatinous areas were composed of fibroblast-like cells and collagen fibers with edema. The fibroblast-like cells showed oval middle-sized nuclei and were fusiform. Some mitotic figures were seen. These cells showed morphological features that were the same as those of the proliferating fibroblast-like cells in the fibrous area. No osseous elements could be seen. Toluidine blue staining revealed no mucinous substance that would indicate metachromasia in the edematous connective tissue. Some hemosiderin deposits were observed in the connective tissue. No stellate cells, which resemble myxoma tumor cells, were observed. From these histological findings, the gelatinous area was considered to consist of the same proliferating fibrous tissue as the fibrous area. It is possible that the gelatinous area was the area of the surgical reduction, because hemosiderin deposition and edema are histopathological findings often observed after surgery.

We diagnosed this case on the basis of the histopathological findings as an ossifying fibroma. An osteoma is a tumor that generates collagen fibers and vessels in well-differentiated cancellous or compact bone and its stroma. On the other hand, fibrous dysplasia is generated from a substrate that consists mainly of collagen fibers and vessels, and shows tissue morphology with scattered, undifferentiated trabecular structures. Ossifying fibroma has morphological characteristics intermediate between osteoma and fibrous dysplasia in terms of histology [10]. In this case, the bone tissue was woven bone. Well-differentiated lamellar bone was observed in the osteoma. The trabeculae arise via metaplasia of fibrous connective tissue and are not typically rimmed by osteoblasts in fibrous dysplasia. Therefore, we diagnosed ossifying fibroma.

Ossifying fibroma is often reported in horses. In most cases, the tumor develops in the mandible. In many countries, because ossifying fibromas occur in horses as young as 2 to 14 months, this tumor is known as equine juvenile mandibular ossifying fibroma [5]. Ossifying fibroma is classified as a benign tumor histologically because it shows low anaplasia, no metastasis, and is characterized by local invasiveness, that is, enlarging fibrous bone growth that leads to destruction of preexisting trabeculae [3]. In this case, tumor tissue extended into the incisive bone, indicating that this ossifying fibroma was relatively invasive.

A preliminary diagnosis of ossifying fibroma can be made from its clinical course, its site of development, the age at onset, and the findings on macroscopic examination, palpation, and radiography, with subsequent definitive diagnosis after biopsy. Total removal via mandibulectomy is considered ideal for tumors of the mandible, and is reported to reduce recurrence and improve the outcome [7]. However, as in this case, when the tumor develops in the maxilla, total resection may be impossible depending on the range and invasiveness of tumor tissue into surrounding tissue, increasing the likelihood of recurrence. Therefore, radiotherapy should be combined with tumor tissue reduction surgery.
References


