Peripheral osteoma of the mandible in a cat

Perifeer osteoma van de mandibula bij een kat

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ABSTRACT

A six-year-old, male, castrated domestic shorthair cat was presented for evaluation of an oral mass. Full physical examination revealed a large hard mass arising from the lateral aspect of the caudal part of the left mandible. Abdominal ultrasound and thoracic computed tomography (CT) were performed to rule out metastatic disease. In addition, CT of the skull with intravenous contrast agent was performed. Histopathological examination of extra-oral biopsies confirmed the tentative diagnosis of peripheral osteoma. A three-dimensional printed skull was used in conjunction with the CT images to determine the detailed location and extent of the mass and to identify anatomical spatial relationships with important neurovascular structures. Surgical debulking of the osteoma was performed. The cat recovered uneventfully but the mass regrew nine months after debulking therapy, sooner than expected. Nevertheless, CT scan of the skull was suggestive for regrowth of the peripheral osteoma rather than for malignant transformation. The owners did not elect to pursue caudal mandibulectomy.

SAMENVATTING

Een zesjarige, mannelijke, gecastreerde kat werd aangeboden voor evaluatie van een orale massa. Op het lichamelijk onderzoek werd een grote harde massa opgemerkt aan de laterale kant van het caudale deel van de linkermandibula. Abdominale echografie en computertomografie (CT) van de thorax werden uitgevoerd om metastasen uit te sluiten. Bijkomend werd een CT-scan van de schedel met intraveneuze contrastvloeistof genomen. Histopathologisch onderzoek van extraorale biopsieën bevestigde de waarschijnlijkheidsdiagnose van perifeer osteoma. Een driedimensionale (3-D), geprinte schedel in combinatie met CT-beelden werd gebruikt om de exacte locatie en omvang van de massa te beoordelen en om alle belangrijke neurovasculaire structuren te kunnen onderscheiden. Chirurgische debulking van het osteoma werd uitgevoerd. De kat herstelde zonder problemen, maar de massa kwam negen maanden na debulking terug. Dat was eerder dan verwacht. Desondanks was de CT-scan van de schedel suggestiever voor terugkeer van het perifere osteoma dan voor een maligne transformatie. De eigenaren kozen ervoor om niet verder te gaan met caudale mandibulectomie.

INTRODUCTION

In domestic animals, primary bone tumors are infrequently diagnosed. In cats, the incidence is estimated to be 4.9 per 100,000 individuals (Dernell et al., 2007). However, most primary bone tumors in cats are malignant (67-90%) (Dernell et al., 2007; Haynes et al., 2012; Thompson and Dittmer, 2016). The mean age at which bone tumors develop in cats is eight to ten years (Dernell et al., 2007; Haynes et al., 2012). Long bones are more often affected than the axial skeleton (Dernell et al., 2007), with only 6% of all primary bone tumors arising from bones of the skull (O'Brien et al., 1996).

Figure 1. Three-dimensional computed tomographic reconstruction of the head of a six-year-old, male, castrated domestic shorthair cat. The images show a solitary osseous mass, firmly attached to the caudal aspect of the left mandible.

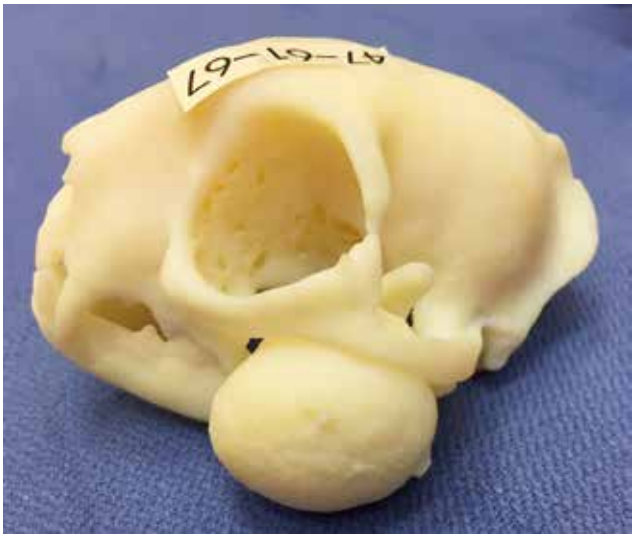

Figure 2. Three-dimensional reconstruction of the skull. The egg-sized mass is firmly adhered to the underlying bone of the left mandible and extends laterally along the mandibular bone. The caudodorsal extension ends immediately rostral to the temporomandibular joint.

Osteoma is a benign primary bone and cartilage tumor (Pool, 1978; Foley, 1993) that is characterized by continuous, slow growth (Mittal and Iyer, 2008) of cancellous bone (trabeculae), compact bone (dense lamellae) or a combination of both (Woldenberg et al., 2005). Osteomas can occur in all domestic animals, but they are more frequently diagnosed in horses and cows than in other species (Pool, 1978). In an early review article it has been reported that less than 2% of cats diagnosed with bone tumors are affected by osteoma (Quigley and Leedale, 1983). Osteomas usually arise from flat bones, with a predilection for the mandible, maxilla, nasal sinuses and the craniofacial region (Foley, 1993; Linden et al., 2016). In cats, the caudal portion of the mandible is the most frequently affected region (Fiani et al., 2011).

In humans, osteomas are classified based on their origin as peripheral, central and extra-skeletal (Johann et al., 2005). Peripheral osteomas grow on the surface of bones, whereas central osteomas arise from within the bone, and extraskeletal osteomas originate in muscle tissue (Woldenberg et al., 2005). Although a similar classification does not yet exist in veterinary medicine, two distinctive patterns resembling peripheral and central osteomas have been distinguished on computed tomography (CT) scans (Fiani et al., 2011).

In contrast to malignant primary bone tumors, the prognosis of osteoma after complete surgical excision is generally good. The prognosis of central osteoma is more reserved than for the peripheral variety because it is known to recur more frequently than peripheral osteoma and excisional surgery is more invasive (Larrea-Oyarbide et al., 2008). When the tumor is in an advanced stage, the extent of the surgery may be so radical that the patient's quality of life would be severely compromised. Therefore, it is important to diagnose and treat an osteoma in its early stage (Corgozinho et al., 2015).

In the current case report, an adult cat with a mandibular peripheral osteoma treated by surgical debulking is described.

CASE REPORT

A six-year-old, male, castrated, domestic shorthair cat (9.2 kg) was presented to the William R. Pritchard Veterinary Medical Teaching Hospital (VMTH) Dentistry and Oral Surgery Service (DOSS), University of California, Davis for evaluation of an oral mass. The owner presented the cat to his private practitioner approximately two months earlier, for evaluation of a visible swelling of the face that had been present for an unknown period of time. Physical examination revealed a round, egg-sized mass lateral to the caudal half of the left mandible. The veterinarian attempted to obtain a fine-needle aspirate, but was unsuccessful due to the hard texture of the mass. On radiographs, the bony nature of the egg-shaped mass was demonstrated. The cat was subsequently referred to a local hospital for CT scan of the skull and biopsies of the mass. A tentative diagnosis of osteoma was made since histopathology only revealed normal woven bone. The cat was subsequently referred to the UC Davis DOSS VMTH.

The owner reported that despite having a large mass, the cat was able to eat, drink and groom itself normally. Physical examination, blood work, urine collection and abdominal ultrasound were performed and were unremarkable. On oral examination, a firm mass was noted at the left caudal oral cavity, causing the side of the face to bulge. The mucosa overlying the mass was mottled red-pink and mildly ulcerated in patches. Moderate gingivitis with mild diffuse plaque and calculus accumulation was noted.

The next day, the cat was placed under general anesthesia. Thoracic CT demonstrated no evidence of metastatic disease or other comorbidities. CT of the skull with intravenous contrast agent revealed a 2 by 3

by 4 cm, smoothly margined, homogenously mineral dense mass arising from the left mandible caudal to the fist molar tooth (Figure 1). The mass had a pedunculated extension laterally along the mandible from the level of the left mandibular fourth premolar tooth to immediately rostral to the temporomandibular joint. Surrounding the mass, there was a halo of soft tissue density with peripheral contrast enhancement. The mass displaced the left masseter muscle medially and bulged ventral to the zygomatic arch to impinge on the ventral aspect of the left pterygopalatine fossa. The left eye was mildly displaced dorsolaterally. A three-dimensional model of the skull was printed for surgical planning (Figure 2). Based on the oral examination and CT findings, an osteoma of the left mandible was suspected. Other differentials included osteosarcoma and osteochondroma.

Dental radiographs confirmed the large bone-opaque mass arising from the left mandible. An incisional biopsy was obtained. Over the ventrolateral aspect of the mass, down to the level of bony tissue, an approximately 2.5-cm skin incision was made. The subdermal tissue and masseter muscle were bluntly dissected to enhance the visualization of the mass. Using an osteotome and mallet, a bone biopsy sample was collected. The surgical site was irrigated with sterile saline 0.9%. The masseter muscle was apposed with simple-interrupted sutures using poliglecaprone 25 4-0 (Monocryl®, Ethicon, US). The same suture was used for the intradermal layer, with the knot buried. Surgical skin glue was used for final skin apposition, as skin sutures would require heavy sedation to remove given the patient's temperament. The biopsy sample was placed in 10% buffered formalin and submitted for histopathology. Photographs were obtained postoperatively to demonstrate the location of the skin incision, which would need to be included in more definitive treatment. Awaiting the biopsy results, buprenorphine (Buprenex®, Reckitt & Colman, UK) 0.01 mg/kg was administered sublingually three times a day for the next three days. Amoxicillin and clavulanic acid (Clavamox®, Zoetis, US) 12.5 mg/kg per oral (PO) two times daily was administered while the incision was healing. It was also recommended to feed soft food, continue keeping the cat indoors, and for the patient to wear an Elizabethan collar to prevent self-trauma to the healing biopsy site.

Histopathological findings revealed bone that was composed of a dense network of lamellar and woven bone, forming expansive and disorganized trabeculae and islands. The surfaces of the trabeculae were variably lined by a single row of osteoblasts that rarely piled up two to three rows thick. The intertrabecular connective tissue was relatively paucicellular. Multifocally, there were vascular canals. Mitotic figures were not noted. The tentative diagnosis of osteoma was confirmed and surgical debulking of the osteoma was advised.

On the day of surgery, the cat was anesthetized and placed in dorsal recumbency under anesthesia.

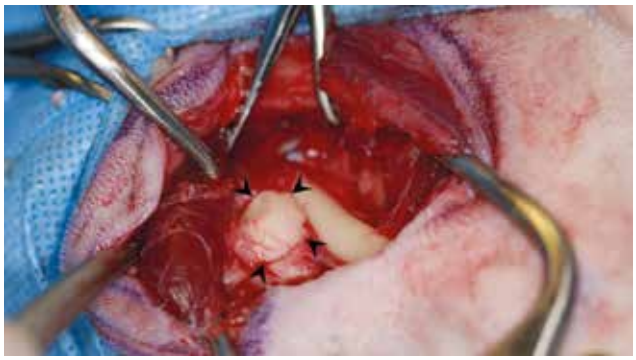

Figure 3. The overlying masseter and digastric muscles were dissected away, along with fibrous tissue, in order to improve visualization of the mass (arrow heads).

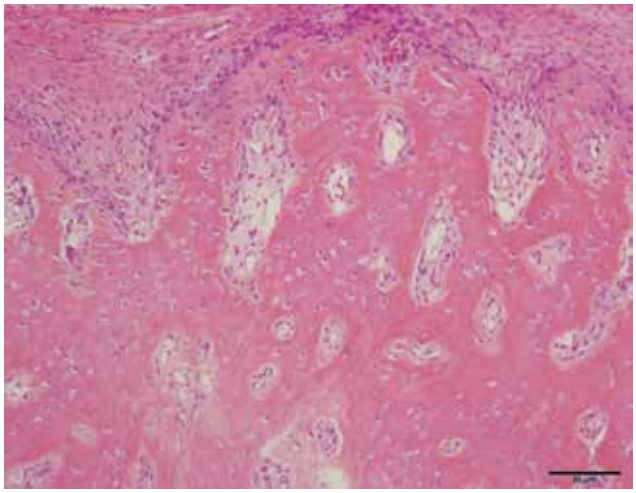

Figure 4. Photomicrograph of the mandibular osteoma of the cat. Note the mass is composed by fronts of woven bone lined by osteoblasts intimately interacting with connective tissue components lining the periphery of the mass (top of image). Also noted progressive maturation of woven bone towards the deeper portions of the mass (bottom of image). H&E stain; X10 magnification.

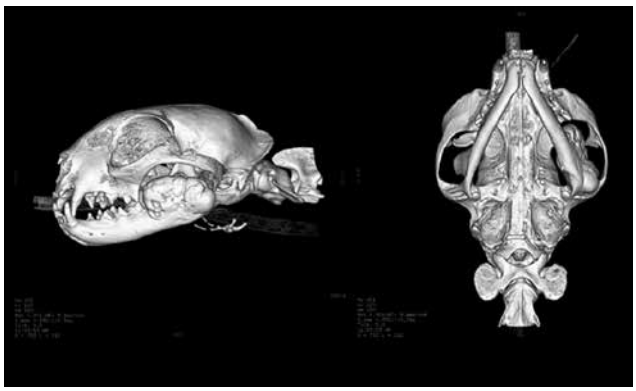

Figure 5. Three-dimensional computed tomographic reconstruction of the head of the domestic shorthair cat nine months after surgical debulking of an osteoma. The tumor recurred at the left caudal mandible with a more axial extension. The medial cortex immediately adjacent to the mass is severely thinned.

A fusiform skin incision was made overlying the left mandible. The masseter and digastric muscles and the fibrous tissue overlying the osteoma were bluntly and sharply dissected away from the surface of the mass (Figure 3). Using an osteotome and mallet, the mass was separated from the underlying mandible. A vessel "feeding" the mass was ligated twice with polyglactin 910 3-0 (Vicryl®, Ethicon, US) and transected. The osteoma was removed in a single piece, placed in 10% buffered formalin and submitted for histopathology. The underlying mandibular bone was further exposed and irregular bone was smoothed with a piezosurgery unit (Implant Center 2, Acteon, France). The surgical site was irrigated with sterile saline 0.9%. The deep and superficial muscle layers were apposed using poliglecaprone 25 4-0 in a simple-interrupted pattern. The overlying skin was closed in the same pattern with nylon 4-0 (Ethilon®, Ethicon, US), tacking most sutures into the underlying superficial muscle and fascia to reduce dead space and further reinforce the closure.

The cat was hospitalized overnight and additional pain management was achieved with meloxicam (Metacam®, Merial, Germany) 0.2 mg/kg IV at extubation and oxymorphone 0.05 mg/kg IV every six hours.

Histopathology revealed that the submitted tissue consisted of 80% immature woven bone (Figure 4). The margins were variably lined by pallisading osteoblasts. Superficially, the woven bone was interwoven with an overlying thin fibrovascular capsule. The clinical diagnosis of a periosteal osteoma was confirmed.

Nine months after debulking surgery, the owner returned to the UC Davis DOSS with the complaint that the mass had regrown. In the owner's opinion, the progression was slow. On physical examination, a bony mass attached to the left caudal mandible was appreciated, yet smaller than the previously diagnosed osteoma and potentially wrapping around slightly to the medial aspect. A cone beam CT scan was performed and showed a mass with a more axial extension than the osteoma of the year before and no impinging on the pterygopalatine fossa (Figure 5). The left caudal mandible was more severely affected by the mineralized mass than the year before. The mass expanded the affected portion of the mandible, causing severe thinning to complete loss of the medial cortex along the length of the mass.

Caudal mandibulectomy was discussed as the curative treatment of choice given the marked thinning of the cortical bone and hence the risk of mandibular fracture if debulking was considered for a second time. However, the cat was free of clinical signs and the owner elected not to proceed with additional surgery. The owner was advised that humane euthanasia should be considered if the cat's quality of life deteriorated. The owner was warned to watch for signs of diminishing quality of life, including decreased appetite or ability to prehend or masticate food, and temporomandibular joint ankylosis.

DISCUSSION

In this case report, a six-year-old domestic shorthair cat with a peripheral osteoma of the mandible is described. Based on previously published single case reports and one case series in cats, there does not seem to be a breed predilection; however, it appears that neutered males are affected more often than females (Maas and Theyse, 2007; Fiani et al., 2011; Corgozinho et al., 2015). The age of cats at presentation is widespread (1-23 years), although most diagnoses of peripheral osteoma in cats are reported around the age of nine years.

The clinical presentation of maxillofacial osteoma can be highly variable, ranging from asymptomatic to difficulties with eating, mandibular drift, mandibular swelling, fever, epiphora, and exophthalmos (Fiani et al., 2011). The cat in the current case report did not have any problems associated with the mass at the time of either presentation, but the owner consulted his primary care veterinarian due to an obvious facial asymmetry.

In both domestic animals and humans, osteomas are mostly described in the maxillofacial region (Vigorita et al., 2008; Thompson and Dittmer, 2016). They are characterized by continuous growth of bone, but can become quiescent for years. Peripheral osteomas can remain unnoticed for an unknown period of time, unless they begin to interfere with important adjacent anatomical structures due to their size (Thompson and Dittmer, 2016).

The mandible was the bone of origin in five out of seven cats in a case series of osteomas (Fiani et al., 2011). In single case reports, osteomas in cats in the nasal cavity, the maxilla, the zygomatic arch, the temporomandibular joint, periorbital, the ribs and the radius have been described (Pool, 1978; Foley, 1993; Maas and Theyse, 2007; Fiani et al., 2011; Haynes et al., 2012; Corgozinho et al., 2015; Cunha et al., 2015).

There is ongoing debate whether osteoma is a true neoplasm, a developmental anomaly or an osteogenic lesion triggered by trauma or infection (Sayan et al., 2002; Woldenberg et al., 2005; Boffano et al., 2012). In the case described here, a traumatic cause exacerbated by muscle traction could have been the trigger for osteoma formation, since the peripheral osteoma was located along the buccal aspect of the left mandible, a region susceptible to trauma, as described in the human literature (Sayan et al., 2002). However, the owner did not recall any history of trauma.

The diagnosis of peripheral osteoma should be made based on the combination of clinical findings, diagnostic imaging and histopathology (Fiani et al., 2011). In the case reported here, the only significant clinical abnormality was a marked bulging of the left side of the face. Usually, peripheral osteomas are not painful on palpation (Cunha et al., 2015). Diagnostic imaging of peripheral osteoma is based on radiological examinations or CT scan (Mittal and Iyer, 2008).

Conventional radiography typically shows a pedunculated, mushroom-shaped, radiopaque mass (Durão et al., 2012). Although the radiological features are suggestive, CT is preferred to understand the precise location and extent of the mass, as well as acting as an aid for surgical planning (Mittal and Iyer, 2008; Durão et al., 2012). On CT, a peripheral osteoma is usually characterized by bone density, smooth margins and a sessile base (compact peripheral osteoma) although it can be less hyperattenuating with rather irregular margins and attached to the bone via a pedicle (cancellous peripheral osteoma) (Richardson et al., 1999; Fiani et al., 2011; Thompson and Dittmer, 2016). Importantly, the definitive diagnosis of osteoma can only be made by proper histological examination (Durão et al., 2012; Thompson et al., 2016). Peripheral osteomas are histologically characterized by a deeper layer of lamellar bone trabeculae and a superficial layer of woven bone trabeculae (Linden et al., 2016; Thompson and Dittmer, 2016). There are two distinctive histological patterns of peripheral and central osteomas, the compact and the cancellous osteoma. Normal-appearing bone with limited marrow spaces and occasional Haversian canals is typical for compact osteoma. Cancellous osteoma consists of bone marrow with osteoblasts and trabeculae of bone (Richardson et al., 1999; Sayan et al., 2002). Based on CT findings and histopathology, the osteoma described in this case was a compact peripheral osteoma.

Two important lesions to be differentiated from osteoma and one another are ossifying fibroma and fibrous dysplasia. All three bony lesions mostly affect the craniomaxillofacial region (Linden et al., 2016). Other differential diagnoses for bone tumors reported in the literature are trauma-induced exostoses of the jaw, osteochondroma, and periosteal osteochondrosarcoma (Fiani et al., 2011; Haynes et al., 2012). Parosteal osteosarcoma (OSA) is another differential that should be taken into account in this specific case, since the mass in the cat described recurred sooner than expected in case of osteoma, which could indicate malignant transformation (Johann et al., 2005; Linden et al., 2016). It is important to note that malignant transformation of an osteoma has never been reported previously in any species (Pool, 1978; Sayan et al., 2002).

Mandibulectomy or debulking are two surgical treatment options that can be performed in cats with mandibular osteomas (Fiani et al., 2011). In humans, peripheral osteomas located on the mandible can be removed via an intraoral or an extraoral approach (Woldenberg et al., 2005; Starch-Jensen, 2017); the former being preferred for esthetic reasons (Woldenberg et al., 2005). The main disadvantage of an intraoral approach is decreased visualization and thus the higher risk of damaging adjacent anatomical structures. In the cat described, an extraoral approach was performed, as in all previously reported cases (Foley, 1993; Maas and Theyse, 2007; Fiani et al., 2011). Due to the complex anatomy of the maxillofacial region, preoperative planning is crucial (Winer et al., 2017). In this case, the authors opted for CT images that were used to create a three-dimensional (3-D) printed skull. The 3-D printing technique has been recently introduced into veterinary medicine to guide surgeons in their preoperative planning (Arzi et al., 2015; Harrysson et al., 2015). This technique makes it possible to convert a two-dimensional (2-D) image (e.g. CT) into a 3-D object (e.g. skull model) in order to achieve a better understanding of spatial relationships and patient-specific anatomy (Winer et al., 2017). This technique reduces the risk of complications (e.g. nerve damage, hemorrhage) (Winer et al., 2017) and may reduce surgery time (D'Urso et al., 1999). Three-dimensional printing is a useful tool for preoperative planning, but it still differs from intraoperative anatomy since it does not include soft tissues (Winer et al., 2017). Due to the location of the mass presented in this study, involving the ramus and body of the mandible and the close proximity to the temporomandibular joint, tumor-free margins could not be obtained without performing a unilateral caudal mandibulectomy.

The osteoma regrew nine months after the debulking surgery, which was not entirely unexpected since microscopic remnants were known to be left behind at the time of debulking surgery. However, the interval from surgery to regrowth was shorter than expected. The long-term prognosis for the cat in this case report is unclear since osteomas near the temporomandibular joint can induce temporomandibular joint ankylosis (Maas and Theyse, 2007). The surgical option of caudal mandibulectomy to achieve definitive cure was explained to the owner, but he elected to forgo further surgery.

CONCLUSION

Clinicians should not jump to conclusions when confronted with bony proliferations; a conclusive rather than a tentative diagnosis should be made before discussing the treatment options and long-term prognosis with the owner. Although relatively rare in dogs and cats, osteoma should be included in the list of differential diagnoses of bone tumors. Osteomas are benign tumors with a typically slow growth rate and generally good prognosis. They may be treated by either debulking surgery or curative radical excision, depending on the location, goals of the client and effect on the patient's quality of life.

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