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Case Report

Extended Subtotal Mandibulectomy in a Cat with Alveolar Osteomyelitis Characterized by a Spiculated Periosteal Reaction

Key words

mandibular mass;
alveolar osteomyelitis;
periosteal reaction;
mandibulectomy

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Abstract: A 10-year-old spayed female Scottish Straight cat presented with a firm, non-movable mass in the right mandible. Over the past month, it had progressively enlarged, leading to difficulty eating and subsequent weight loss. Computed tomography (CT) scans revealed that the mass was confined to the body of the mandible, with a prominent periosteal reaction characterized by a spiculated pattern. The extensive and irregular morphology of the periosteal reaction strongly suggested malignancy. No evidence suggestive of metastasis was identified on physical examination or diagnostic imaging. Surgical resection was prioritized over obtaining a definitive diagnosis, owing to the rapid increase in mass size and associated clinical signs. An extended subtotal mandibulectomy was performed for diagnostic and therapeutic purposes, revealing alveolar osteomyelitis. Postoperatively, the patient showed improved masticatory function with no apparent signs of oral pain, indicating a good surgical outcome. During the 16-month follow-up period, the patient remained in good general and physical condition without recurrence of the mass. To the best of our knowledge, this is the first report suggesting that osteomyelitis can present with an extensive, irregular periosteal reaction with radiating lines ("sunburst" subtype), typically associated with malignant tumors. Additionally, surgical resection may be critical for recovery of masticatory function and alleviation of oral pain, even in cases involving benign mandibular lesions.

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Introduction

Feline mandibular swelling has a 50% malignancy rate, and benign lesions mostly manifest as osteomyelitis (1). Mandibular bone lesions are expected to be more biologically aggressive when cortical margins are ill-defined, when the zone of transition is broad and indistinct, and when the periosteal reaction is irregular on diagnostic imaging (2, 3). Unfortunately, both malignant tumors and osteomyelitis can exhibit these aggressive characteristics (2, 3, 4, 5). Mandibulectomy is commonly recommended for malignant mandibular tumors because complete surgical excision is the only curative method (6).

Moreover, despite variations in the extent of distant metastasis among different types, most malignant tumors have low metastatic potential (6, 7, 8, 9), suggesting a higher likelihood of achieving curative ablation. Conversely, the extent of local invasion is similar among the different types of mandibular tumors (2). Therefore, there is a general consensus that a minimum margin of 1 cm should be aimed for when planning for surgery with curative intent (2, 7). When surgery is not appropriate, palliative care may temporarily improve quality of life but often leads to euthanasia due to tumor progression (6).

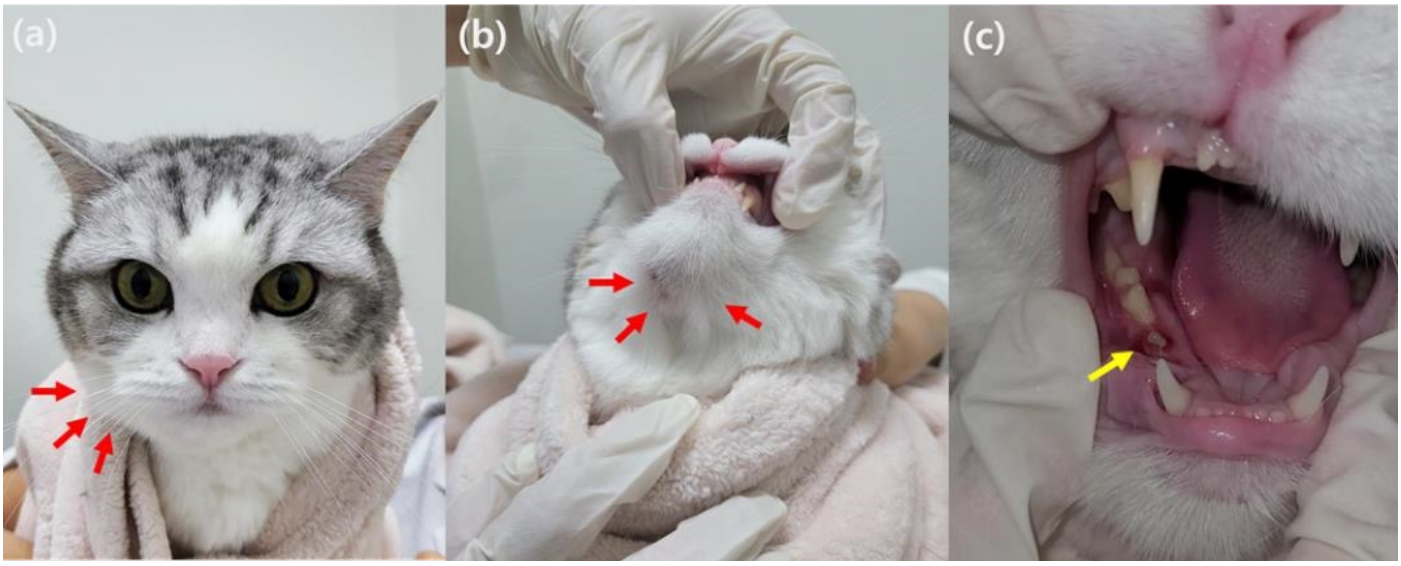

Figure 1: Preoperative examination reveals a firm mandibular mass (red arrow) and gingival erythema around the mandibular right third premolar (Triadan 407) (yellow arrow)

In the treatment of osteomyelitis, while general principles are followed, clinical decisions are often adapted to the patient's specific condition (10, 11). Antibiotic therapy alone may be sufficient, but surgical excision is required in more advanced cases (4, 12). When radical excision is chosen for mandibular osteomyelitis, at least 1 cm margin is generally recommended, although this remains controversial (6, 12). Ultimately, regardless of the presence of malignant tumors or osteomyelitis, mandibular excisions with margins >1 cm are believed to yield a good prognosis.

Among the various mandibulectomy methods, total mandibulectomy is the most invasive and technically demanding procedure (7, 13). It involves removal of the entire mandibular body and ramus with disarticulation of the temporomandibular joint (TMJ), requiring detachment of the masseter, temporalis, medial and lateral pterygoid, and digastricus muscles. Disarticulation of the TMJ can increase surgical risks and anesthesia time due to its technical complexity (13). Although typically reserved for more severe cases, total mandibulectomy has a complication rate of approximately 38%, exceeding that of partial resections (33% for segmental mandibulectomy and 12% for unilateral rostral mandibulectomy) (14). In contrast, subtotal mandibulectomy allows removal of the mandibular body while preserving the vertical ramus associated with TMJ, offering a balance between therapeutic efficacy and functional preservation. A recently described variation, the extended subtotal mandibulectomy, further refines this approach by enabling complete excision of the mandibular canal (13). This technique may minimize surgical morbidity without compromising therapeutic goals, making it an increasingly preferred alternative in appropriately selected cases.

Alveolar osteomyelitis refers to infection or inflammation involving the alveolar bone, including the mandibular canal and medullary spaces, which support the teeth and are commonly affected in association with dental or periodontal disease (4, 15). This case report presents a situation where alveolar osteomyelitis manifests with a spiculated periosteal pattern, a distinctive sign of malignancy. Surgery was required to alleviate the clinical signs, and a wide surgical resection in the form of an extended subtotal mandibulectomy allowed for successful control of the mandibular osteomyelitis.

Case description

A 10-year-old spayed female Scottish Straight cat weighing 3 kg was admitted for evaluation of a right mandibular mass. The patient had difficulty eating, leading to recent weight loss. On history taking, the owner reported that the patient had been diagnosed with a feline odontoclastic resorption lesion (FORL) affecting the right mandible teeth four years earlier. Although tooth extraction is the primary treatment for FORLs, it was not pursued in this case due to concerns regarding anesthesia. Instead, the patient was treated with doxycycline, meloxicam, and alendronate, as reported by the owner. The patient continued antibiotics and nonsteroidal anti-inflammatory drugs (NSAIDs) for the past four years until admission to our hospital.

Physical examination revealed a firm, non-movable mass of size 2 × 3 × 2 cm (W x L x H) in the right mandible. Gingival erythema was also observed, particularly around the mandibular right third premolar (Triadan 407) (Figure 1). The patient exhibited no systemic signs such as fever or lethargy. No abnormalities were detected on superficial lymph node palpation. Blood analysis revealed no significant findings. Radiography demonstrated an extensive, irregular periosteal reaction with radiating lines (the "sunburst" appearance) localized to the right mandibular body, with

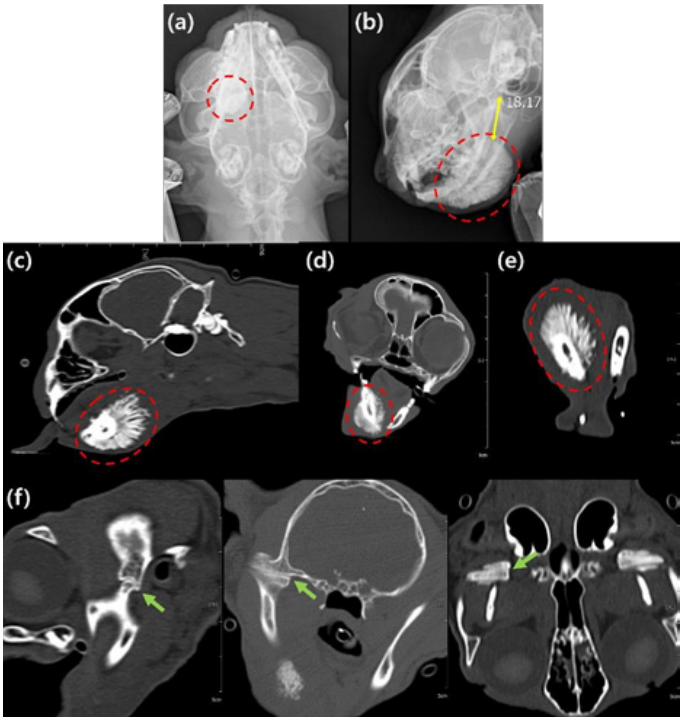

Figure 2: Radiography (a-b) and CT (c-f) reveal a spiculated periosteal reaction ("sunburst" type) on the right mandibular body (red dotted circle), 18.2 mm rostral to the TMJ (yellow arrow). The TMJ shows no evidence of involvement on CT (green arrow)

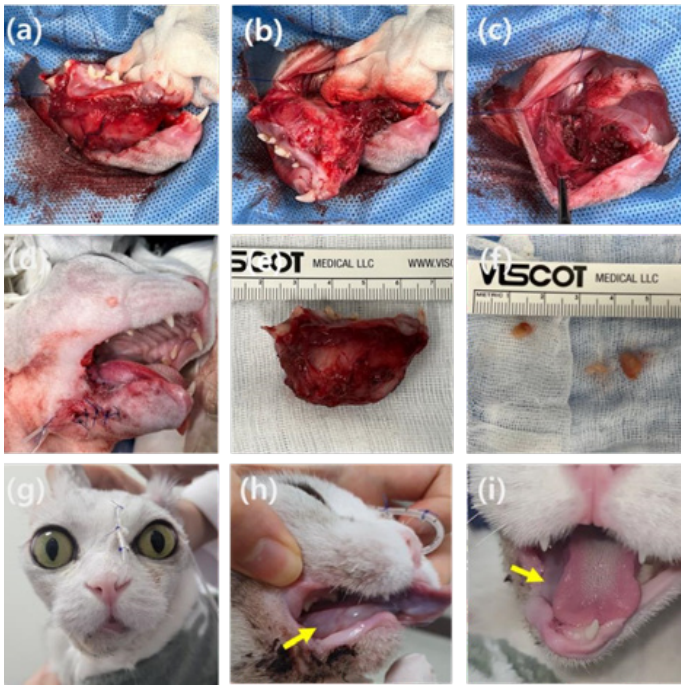

Figure 3: Intraoperative (a-f) and postoperative (g-i) photographs. The mandible is resected approximately 1 cm caudal from the mass (a-c), and a Penrose drain tube is placed (d). The right mandibular lymph nodes are also excised for metastasis evaluation (f). For postoperative care, a nasoesophageal tube is temporarily placed (g). A ranula-like lesion (yellow arrow) is noticeable after surgery (h-i)

the temporomandibular joint (TMJ) unaffected (Figure 2). The caudal border of the periosteal reaction was located approximately 18.2 mm rostral to the right TMJ, specifically the condylar process. Thoracic and abdominal radiography, abdominal ultrasonography, and computed tomography (CT) were performed to evaluate for possible metastasis and to better characterize the bony lesion. CT reconfirmed the periosteal reaction in the right mandibular body without evidence of additional bone involvement or distant metastasis (Figure 2). No signs of possible metastasis were noted across all imaging modalities. Due to the extent and irregularity of the periosteal reaction, a preliminary diagnosis of malignancy was strongly considered, although benign tumors and inflammatory conditions could not be entirely ruled out.

Although preoperative biopsy was considered, extended subtotal mandibulectomy was prioritized to address the rapid growth of the lesion and the potential risk of temporomandibular joint involvement. The mandible was resected approximately 1 cm caudal from the mass. Considering the high possibility of malignancy, the right mandibular lymph nodes were also excised to evaluate possible metastasis (Figure 3). The entire resected mandible and mandibular lymph nodes were sent for histopathological analysis.

General anesthesia was maintained with isoflurane following propofol induction. Analgesia was achieved with fentanyl CRI. The patient was positioned in left lateral recumbency, and inferior alveolar nerve block was performed using bupivacaine. A labial mucosal incision was made, extending from the mandibular symphysis to the caudal boundary of the planned ostectomy. Dissection was carried along the mandibular body, including the sublingual aspect, to expose the mandibular symphysis rostrally and the caudal boundary of the planned ostectomy. The anterior belly of the digastricus muscle was detached to access the mandibular body. Care was taken to avoid laceration of the inferior alveolar neurovascular structures. The mandibular symphysis was transected and the mandibular body was excised approximately 1 cm caudal to the gross lesion, using a combination of an osteotome and an oscillating saw. The ostectomy was extended caudally towards the angular process to include the mandibular foramen, enabling en bloc removal of the mandibular canal, consistent with an extended subtotal mandibulectomy technique. The vertical ramus, including the coronoid and condylar processes, was preserved, and disarticulation of the temporomandibular joint was not required. Margins were planned based on preoperative CT imaging and intraoperative gross appearance, aiming to achieve 1 cm of grossly normal tissue around the lesion. Margin evaluation was based on intraoperative gross assessment, as histologic confirmation of clean margins is inherently challenging in chronic osteomyelitis due to diffuse inflammatory remodeling. Wound closure was performed using 4-0 PDS® II (polydioxanone, Ethicon, Johnson & Johnson, USA) for the digastricus, oral mucosa,

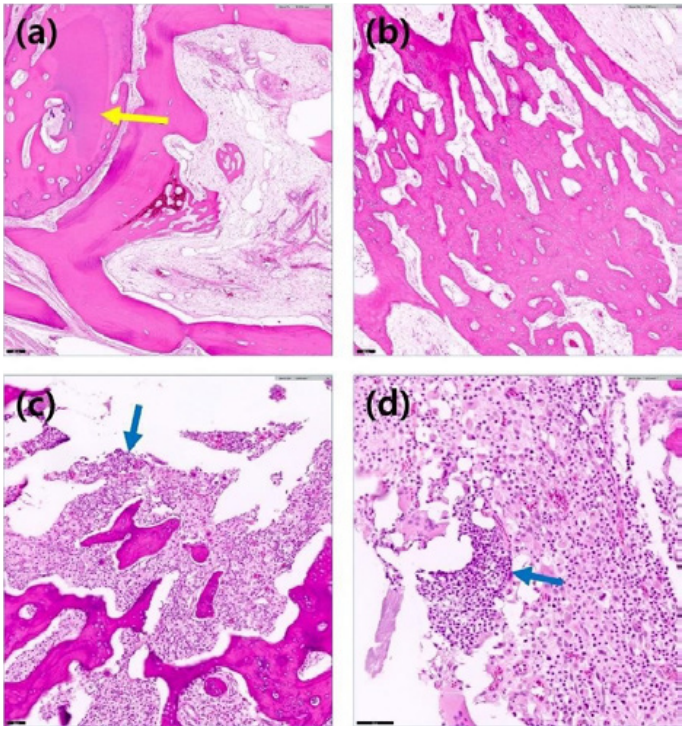

Figure 4: Histopathology of the resected mandibular specimen (H&E, 5x (a), 10x (b), 20x (c), 40x (d) magnifications). Alveolar osteomyelitis manifests as suppurative, marked, chronically active areas with microabscess formation (blue arrow), granulation tissue, chronic bony reorganization, and intratrabecular edema. Periosteal woven bone proliferation is observed in the mandible (a, b, c) and tooth (yellow arrow).

and subcutaneous layers, and 3-0 Dafilon® (monofilament polyamide, B. Braun, Melsungen, Germany) for the skin.

A Penrose drain tube was placed to manage the anticipated dead space, secured using two interrupted sutures, and removed on postoperative day 5 due to marked reduction in drainage and to minimize the risk of retrograde infection. A nasoesophageal tube was temporarily placed for postoperative care to support enteral nutrition (Figure 3). Return of voluntary appetite was confirmed on postoperative day 7. However, oral ingestion was postponed and the nasoesophageal tube was maintained until the day of suture removal (postoperative day 17), to minimize the risk of dehiscence caused by food-related trauma.

Histopathological examination of the resected mandibular specimen revealed alveolar osteomyelitis with periodontitis (Figure 4), as well as reactive lymphadenopathy of the right mandibular lymph nodes. Alveolar osteomyelitis manifested as suppurative, marked, chronically active areas with microabscess formation, granulation tissue, chronic bony reorganization, and intratrabecular edema. Marked chronic periosteal new woven bone formation was also observed. The mandibular lymph As a wide resection was performed and no clinical signs were observed postoperatively, the likelihood of residual disease was considered low. However, given the chronicity and extent of the lesion, and the inherent difficulty in assessing complete surgical margins in chronic osteomyelitis, adjunctive clindamycin therapy (22 mg/kg PO BID for 6 weeks) was administered postoperatively to minimize the risk of residual infection and recurrence.

After surgery, a transient ranula-like lesion developed and spontaneously resolved within 2 weeks. Persistent minor drooling was noted but required minimal care. (Figure 3) Although cheiloplasty was considered, further intervention was not pursued due to the owner's concern about anesthesia. The cosmetic changes associated with extended subtotal mandibulectomy were minimal (Figure 3), and no major complications were identified.

Postoperatively, the patient showed improved masticatory function, with no apparent signs of oral pain. Clinical improvement was clearly evident by the time of suture removal and remained stable throughout the 16-month follow-up period. The patient remained in good general and physical condition without recurrence, as assessed through periodic phone interviews with the owner. Although the patient did not return for in-clinic re-evaluation beyond the early postoperative period, the overall surgical outcome was deemed successful, with complete resolution of clinical symptoms and no major complications.

Discussion

In this case, the extensive periosteal reaction, typically characteristic of malignancy, led to the initial assumption of a neoplasm. Periosteal and endosteal new bone formation (reactive bone formation) is a common feature of neoplasia and osteomyelitis, along with osteolysis and osteosclerosis (3, 4, 5). However, although bone infections may also have an active periosteal reaction, extremely aggressive periosteal reactions are more commonly associated with tumors (3, 16). Furthermore, the immense degree of periosteal reaction observed in this case, is rare even in tumors, let alone in osteomyelitis. In cats, the “sunburst” subtype of periosteal reaction in the mandible has been described in a few cases of osteosarcoma (17), and squamous cell carcinoma (6), but not in osteomyelitis. In humans, strong periosteal reaction has been described in osteomyelitis but typically manifests as an “onion-skin” (laminated) appearance (18, 19, 20, 21). The spiculated pattern, including both “hair-on-end” and “sunburst” subtypes, is highly suggestive of Ewing’s sarcoma and osteosarcoma (20), respectively. To our knowledge, this is the first report to show that the aggressiveness of mandibular osteomyelitis can be equal to or even exceed that of neoplastic changes.

The need for surgery and the extent of surgical resection are typically guided by biopsy results and the patient's response to initial medical management, such as antimicrobial therapy. Advanced imaging modalities such as CT can improve biopsy accuracy by identifying the most representative areas for sampling. However, biopsy results may still be misleading due to non-representative sampling, particularly in heterogeneous or necrotized bone lesions.

Furthermore, initial medical therapy is often ineffective in osteomyelitis due to antimicrobial resistance or poor drug penetration caused by local ischemia and the presence of a "blood-bone" barrier (22). In this case, although preoperative biopsy was considered, surgical resection was prioritized because of the rapid progression of the lesion over one month and the risk of TMJ involvement. Recognizing the potential for sarcoma, wide resection margins were intentionally obtained, including complete removal of the mandibular canal, to mitigate the risk of incomplete excision. In feline patients, achieving margins greater than 1 cm is challenging due to their small craniofacial dimensions (6). Since wide resection was attainable without exposure of the TMJ, surgery was considered a better option than waiting for a definitive diagnosis. Additionally, no remission was observed with previous long-term administration of antibiotics and NSAIDs prescribed for presumed FORL, further supporting the need for surgical management. Thus, while preoperative biopsy remains the gold standard, in extensive bone lesions where wide resection is achievable without significant morbidity, direct surgical excision may be justified to preserve anatomical function and optimize outcomes.

Several classification systems exist; however, osteomyelitis of the jaw is generally classified according to its chronicity and etiology (4, 5, 12). In humans, the arbitrary standard between acute/subacute and chronic is 4 weeks (4, 5, 12, 23). Although no set standard exists in veterinary medicine, the long-term recognition of mandibular swelling and the histological findings of bony reconstruction in this patient fits the criteria for chronic osteomyelitis.

The underlying cause of osteomyelitis in this case was unclear. Although no microorganisms were observed on histopathology, the exclusion of infectious agents was difficult because culture and sensitivity testing were not performed. However, cultures are often unrewarding (23, 24) because oral microbiota is detected in most cases of mandibular osteomyelitis, indicating the possibility of sample contamination. In addition, negative culture results may result from inadequate techniques (4, 5, 12, 23). Osteomyelitis of the jaw may occur as a secondary consequence of periodontal disease, endodontic disease, or smoldering regional bacterial infections following extraction, trauma, or systemic inflammatory/metabolic diseases. In cats, osteomyelitis in the form of alveolar bone expansion commonly occurs with periodontitis and/or FORL (4). This phenomenon has not been extensively investigated and can be expressed in various ways such as "peripheral osteitis, " "alveolar osteitis, " and "peripheral buttressing" (4). Although the causal relationship between osteomyelitis, periodontitis, and FORL has not been fully elucidated, some studies suggest that FORL plays a significant role in causing this phenomenon (4, 25).

The patient's history, as reported by the owner, indicated a prior diagnosis of FORL, though no diagnostic imaging or clinical records were available. Histopathology confirmed periodontitis, suggesting a possible association with the osteomyelitis. However, without definitive diagnostic confirmation, the contribution of FORL remains speculative. Given the absence of other comorbidities, a potential link cannot be entirely excluded.

The possibility of medication-related osteonecrosis of the jaw (MRONJ) associated with long-term bisphosphonate therapy should also be considered. This patient had several risk factors consistent with MRONJ, including a history of prolonged alendronate administration, mandibular involvement, and chronic osteomyelitis associated with periodontitis. However, in the absence of necrotic bone or clinical signs of bone exposure, the likelihood of MRONJ in this case is reduced, although it cannot be entirely excluded. (26)

There were minor complications following extended subtotal mandibulectomy; a ranulalike lesion and increased drooling. A ranula-like lesion can develop from formation of hematoma and/or trauma to the salivary ducts. It is speculated that the ranula-like lesion was only transient because careful dissection was made avoiding inadvertent trauma to the mandibular duct and the major sublingual duct that open on the sublingual caruncle. Drooling was likely a consequence of altered jaw conformation following mandibulectomy. No major complications were identified.

The clinical implications of this case report can be summarized as follows. First, to the best of our knowledge, this is the first report suggesting that mandibular osteomyelitis in cats can present with an extensive, spiculated periosteal reaction ("sunburst" subtype), a pattern typically associated with malignant bone tumors. Second, even in patients with benign mandibular lesions, surgical resection plays an important role in improving masticatory function and alleviating oral pain. Particularly, in fast-growing masses that have not yet invaded the TMJ, timely surgical intervention may be critical to preserve joint function and optimize postoperative outcome. Third, extended subtotal mandibulectomy may offer an effective surgical strategy for managing extensive mandibular lesions associated with osteomyelitis, providing both disease control and functional preservation.

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Razširjena subtotalna mandibulektoemija pri mački z alveolarnim osteomielitisom, za katerega je značilna spikularna periostalna reakcija

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Izvleček: 10-letna sterilizirana škotska mačka z ravnimi ušesi je imela trdno, nepremično maso na desni spodnji čeljusti. V zadnjem mesecu se je masa postopoma večala, kar je povzročilo težave pri prehranjevanju in posledično izgubo telesne mase. Računalniška tomografija (CT) je pokazala, da je bila masa omejena na telo spodnje čeljusti, z izrazito periostalno reakcijo, za katero je bil značilen špičast vzorec. Obsežna in nepravilna morfologija periostalne reakcije je nakazovala malignost. Pri kliničnem pregledu ali diagnostičnem slikanju ni bilo ugotovljenih znakov, ki bi nakazovali na metastaze. Zaradi hitre rasti mase in s tem povezanih kliničnih znakov je bila kirurška resekcija prednostna pred pridobitvijo dokončne diagnoze. Za diagnostične in terapevtske namene je bila opravljena razširjena subtotalna mandibulektomija, ki je razkrila alveolarni osteomielitis. Po operaciji je imela mačka boljšo žvečilno funkcijo brez očitnih znakov bolečine v ustih, kar kaže na dober kirurški izid. V 16-mesečnem obdobju spremljanja je mačka ostala v dobrem splošnem in fizičnem stanju brez ponovitve tvorbe. Po našem vedenju je to prvo poročilo, ki kaže, da se osteomielitis lahko kaže z obsežno, nepravilno periostalno reakcijo z radiacijskimi črtami (podtip »sunburst«), ki je običajno povezan z malignimi tumorji. Poleg tega je kirurška resekcija lahko ključnega pomena za povrnitev žvečilne funkcije in lajšanje bolečin v ustih, tudi v primerih benignih lezij mandibule.

Ključne besede: mandibularna masa; alveolarni osteomielitis; periostalna reakcija; mandibulektomija; mačka