Case report: Congenital aneurysmal bone cyst in a Holstein calf

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SUMMARY
Aneurysmal bone cysts (ABCs) are rare, intraosseous, tumour-like lesions that can affect the axial and appendicular skeletal system primarily of young animals and humans. A 3-days old male Holstein-Frisian calf exhibited a large and solid swelling involving the most rostral portion of the left hemi-mandible. The mass was composed of variably sized cavernous spaces containing clotted blood and serofibrinous exudate, separated by fibrous and fibro-osseous septa. Histologically, the spaces were lined by flattened to plump spindle cells and contained haemorrhages, fibrin, and multinucleated osteoclast-like cells. Curettage of the cavity was performed to remove the cyst lining and break down the bony septa. Three months later, the mandibular swelling had resolved with radiographic evidence of remodelling and resolution of the cyst.

Keywords: Aneurysmal bone cyst, calf, congenital lesion, surgery.

Introduction

Aneurysmal bone cysts (ABCs) are rare, intraosseous, tumour-like lesions that can affect the axial and appendicular skeletal system primarily of young animals and humans [14, 18, 19]. ABCs usually present as solitary lesions with benign behaviour, although cases of multiple lesions in a single animal and malignant transformation after surgical manipulation have been reported [1, 10, 11, 15, 20]. ABCs have been reported in llamas [1], dogs [19], cats [4], horses [14, 17, 21-24], psittacines [9], and cattle [3]. In horses, the mandible is the most common location for ABCs unlike in humans where the metaphysis of long bones, pelvis, and vertebral column are more commonly affected [2, 17, 19, 25]. Grossly, ABCs are expansive, lytic bone lesions that are composed of variably sized intraosseous cavernous spaces containing serosanguinous fluid and/or blood. Hemangiosarcomas and telangiectatic osteosarcomas resemble ABCs grossly and should be considered as primary differentials [14, 20, 25]. Histologically, blood-filled cavernous spaces of ABCs are separated by variably thickened fibrous and fibro-osseous septa composed of spindle cells and multinucleated osteoclast-like cells. Additional elements that can be randomly dispersed within the septal stroma include regions containing haemorrhages, hemosiderophages, mineral deposits, and irregular spicules and trabeculae of reactive and poorly mineralized bone. Cavernous spaces are lined by flattened to plump spindle cells and often contain multinucleated cells as observed within surrounding stroma [14, 16, 18].

To our knowledge, this is the first report that describes a rare congenital bone anomaly of unclear aetiopathogenesis, involving the mandible of a young calf.

Case Report

A 3-days old male Holstein-Frisian calf was discovered to have a large, solid swelling involving the most rostral portion of the left hemi-mandible (figure 1). Initially, the
mandibular mass did not hinder nursing and was treated conservatively and monitored for progressive enlargement. Within 3 days after birth, the mandibular mass increased in size. The referring veterinarian recommended euthanasia of the calf because of a poor prognosis, as the location, size, and extent of involvement of the mass made surgical removal unrewarding because it would cause major disfigurement and nursing complications.

On physical examination, all mandibular incisors were present (figure 1). Lateral radiographs showed a multi-lobular cystic lesion extending caudally from the incisor teeth (figure 2). There was expansion to the cortical bone, with multiple fine incomplete septa contained within the cystic structure. The cyst contained sanguineous fluid and a discrete lining that was removed along with loose trabecular bone.

Surgical exploration and curettage were recommended. Antibiotic therapy (procaine / penicillin 20 mg/kg twice daily IM) and administration of flunixin-meglumine (1.1 mg/kg once daily IV) was commenced and continued for 5 days postoperatively. The calf was anaesthetised and maintained with an intravenous 100 pg/kg b.w. of detomidine (Domosedan, Pfizer, Turkey), 10 mg/kg b.w. of ketamine (Alfamyne; Egevet, Turkey), and 0.5 mg/kg of midazolam (Demizolam; Dem, Turkey) combination [12] in dorsal recumbency. Two 3 cm ventral incisions were made either side of midline and continued into the cyst cavity without penetrating the buccal mucosa. The cyst was filled with sanguineous fluid and a discrete lining was identified. Curettage of the cavity was performed to remove the cyst lining and break down the bony septa. Polyvidone iodine soaked gauze was packed into the cavity and a silastic through-and-through drain was placed. The gauze packing was removed after 3 days and the cavity washed twice daily with saline. The calf was discharged 5 days postoperatively with instructions to continue wound lavage.

Histopathological examination of the cyst lining revealed fibrous connective tissue, hemosiderophages and osteoid cells and a diagnosis of aneurysmal bone cyst was established.

Three months later, the mandibular swelling had resolved with radiographic evidence of remodelling and resolution of the cyst.

Discussion

The pathogenesis for the development of these lesions is still unclear, although it has been proposed that possible malformation of intraosseous vasculature secondary to trauma, bleeding disorders, fibrous dysplasia, hematomas, or underlying neoplasia may be responsible [3, 23, 24]. LEITHNER et al. [15] theorized that some human ABC lesions are indeed neoplasms with suspected genetic predispositions based on immunohistochemical and genetic studies and recommend retrieving more data from patients about possible familial associations.

Grossly, primary differential diagnoses for firm, mandibular and maxillary lesions of young animals include unicameral bone cyst, dentigerous cyst, ameloblastoma, odontoma, ossifying fibroma, and fibrous dysplasia [17, 20, 24]. Of the aforementioned differentials, unicameral bone cysts not only possess similar gross features but also have similar histological features (e.g. lytic lesion, cystic cavities,
multinucleated cells and hemosiderophages) as observed in ABCs, although they have solitary cystic cavities with clear to serosanguinous fluid compared with the variably sized blood-filled cysts consistently found in ABCs [25]. Dentigerous cysts, ameloblastomas, and odontomas contain variable amounts of odontogenic epithelial components with and without the presence of well-formed teeth, denticles, or tooth-producing elements (e.g. dentin, enamel, cementum, etc.) [7]. Histologically, ossifying fibromas and fibrous dysplasia are easily differentiated from ABCs because of the lack of cyst formation as well as the predominant accumulations of fibroblasts and loose fibrous tissue, respectively [7].

Spontaneous resolution of ABCs in animals and humans as well as responses to intra-lesional injection of steroids in humans has been reported [3]. Surgical curettage with bone grafting has been the recommended treatment of choice for these lesions [2]. Adjuvant treatment to surgical curettage and bone grafting such as cryosurgery and phenol infusion has proven to decrease the recurrence rates of ABCs in humans [10, 14, 24]. Treatment recommendations for aneurysmal bone cysts include surgical drainage and curettage and removal of the cystic lining [2, 10, 20, 25]; however, recurrence rates following curettage alone may be higher [2]. Recurrence rates in the human literature reportedly range from 30% with en bloc resection to 50% with curettage alone [5, 16]. Recurrence may be due to incomplete removal of the cyst lining or failure to resolve underlying pathology. Cancellous or corticocancellous bone grafting has also been recommended for its osteoinductive and osteoconductive properties [6, 7, 13] however, adjuvant treatments may not be necessary for resolution of uncomplicated aneurysmal bone cysts. There have been a limited number of reports on the success of treatment of aneurysmal bone cysts in animals, but where there is no apparent underlying disease process, the prognosis for surgical therapy appears to be good [3, 8].

As a conclusion, the reported case is the first description of an aneurysmal bone cyst in a calf and patient signalment, history, clinical presentation, radiographic and histopathological features are equally important when making a diagnosis of an aneurysmal bone cyst. Surgical drainage and aggressive curettage of uncomplicated mandibular aneurysmal bone cysts appeared to offer a good prognosis for resolution in this calf.

References


