

CASE REPORT

Concurrent Occurrence of Aneurysmal Bone Cyst and Fibrous Dysplasia in the Mandibular Bone of a Neonatal Calf

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INTRODUCTION

Fibrous dysplasia (FD) of the bone is a congenital disorder characterized by replacing typical cortical and medullary bone with disorganized fibrous bone tissue in humans and animal species. This disorder disrupts the mechanisms of bone remodeling, leading to a developmental, non-neoplastic abnormality in bone growth^[1]. The principal cause of FD in humans is associated with a somatic mutation in the GNAS gene (Guanine Nucleotide binding protein, Alpha Stimulating activity polypeptide)^[2]. Significantly, FD is characterized by abnormal bone formations embedded within the fibrous tissues^[1]. The incidence of FD in the animal population is reportedly rare, with documentation confined mainly to case reports including three canines^[3-5], a single equine^[6], multiple primate species^[7,8], as well as a single instance in a kudu^[8]. While it mainly impacts the craniofacial skeleton, there are recorded cases affecting the femur, tibia, and ribs in animals.

Aneurysmal bone cysts (ABCs) represent uncommon intraosseous lesions, predominantly impacting the axial and appendicular skeletons of both young animals and

Abstract

This report documents the simultaneous occurrence of aneurysmal bone cysts and fibrous dysplasia in a neonatal calf's mandibular bone. The mandibular bones exhibited significant protrusion, cystic cavities, and thin woven bone spicules. Additionally, the bone trabeculae were underdeveloped with necrotic changes, and there was an absence of bone marrow and osteoblastic rimming in the woven bone. Aneurysmal bone cysts were characterized by blood-filled cystic spaces lacking epithelial or endothelial cells, separated by fibroblastic septa. In conclusion, a unique case of mandibular fibrous dysplasia displaying certain similarities to cherubism and ABC has been recorded in a calf.

Keywords: Aneurysmal bone cyst, Bovine, Congenital fibrous dysplasia, Mandibular bone

humans^[9]. ABCs have been observed in a range of species, noted in canines^[4], felines^[10], equines^[11], and bovines^[12]. Morphologically, ABCs are distinguished by their expansive, osteolytic features, composed of intraosseous cavities of diverse sizes occupied by either blood or serosanguineous fluid^[10].

The purpose of this report is to describe the morphological changes observed in a calf affected with both fibrous dysplasia and aneurysmal bone cyst of the mandibular bone in a calf.

CASE HISTORY

Ethical Consideration

Information about necropsy procedures was provided to the animal owner before the necropsy, and a consent form was obtained from him (Protocol number: 5802).

Case Description

A neonatal male calf of mixed breed was presented to the Pathology Department, Veterinary Faculty of Firat University for necropsy at two hours of birth. The calf experienced complications during birth, requiring the



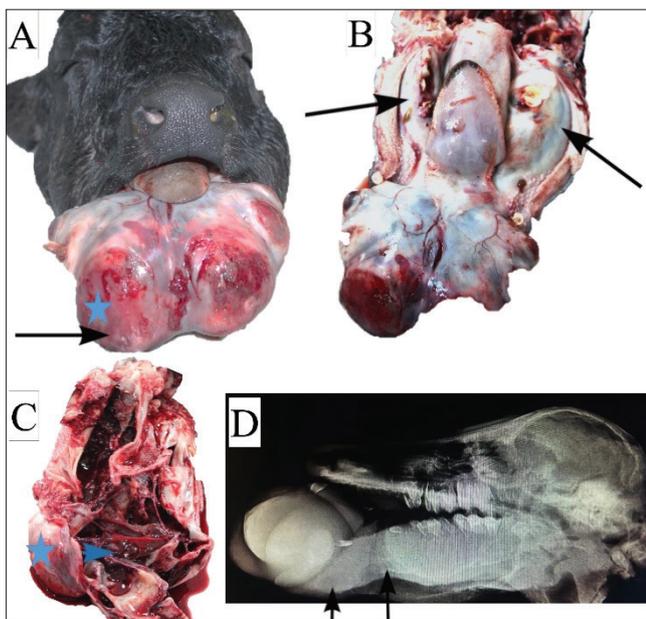


Fig 1. Gross and radiological images of the mandibular changes. **A.** Anterior view of the head, highlighting bilateral expansion and bulging of the mandible (arrow). **B.** Anterior view of oral cavity, there was a noticeable enlargement and swelling (arrows) of the mandible. **C.** In the longitudinal section of the right hemimandible, multiple cystic spaces are evident (arrowhead), as well as stars marking their presence. **D.** The cystic spaces in the mandible, radiological examination, Lateral view

intervention of veterinary services. The dam had been purchased in her third trimester of gestation and did not exhibit any previous health problems. Prophylactic measures included vaccination against foot-and-mouth disease and administering antiparasitic agents roughly four months before parturition. The farm, maintaining a bovine population of 30 cattle, reported no prior occurrences of congenital anomalies in calves.

Gross Lesions

The entire mandible, extending bilaterally from the symphysis to the condyles, was swollen and filled with many cystic cavities (Fig. 1-A,B,C). The swelling protruding outward from the mandibular symphysis measured 12x10x10 cm (Fig. 1A). The bone's thickness ranged from 2 to 3 mm, and the cyst-like spaces were filled with blood-tinged fluid (Fig. 1-C). The mandibular bone was so delicate that it could be cut with a scalpel. The affected areas exhibited a fragile and porous texture, attributable to the deterioration and enlargement. Consequently, this pathological process led to the extensive degradation of osseous tissue, forming numerous cystic cavities. Furthermore, this deterioration displaced the lower incisors and premolars from their respective alveolar lumens.

Radiological Findings

Radiographic examination revealed a mass encompassing the mandible, predominantly situated in the area typically

reserved for the incisor teeth. Notably, the mandible exhibited cystic formations, particularly around the incisor zone (Fig. 1-D). The lesion was multilocular, with well-defined boundaries consisting of multiple cysts or chambers.

Histopathologic Changes

Tissue samples from mandibula, kidney, liver, spleen, brain, lungs were fixed in a 10% formalin solution before being routinely processed and stained with hematoxylin and eosin (H&E) for light microscopic examination.

Microscopically, the most common lesion was numerous thin spicules of woven bone. They had a curved configuration, often forming circular patterns reminiscent of Chinese letters (Fig. 2-A), although occasionally they appeared linear. The woven bones were embedded in fibrous tissue containing fibroblastic cells. These trabeculae lack osteoblastic rimming. Bone trabeculae showed necrosis and remodeling and were surrounded by numerous multinucleated cells. (Fig. 2-B) Throughout the stroma, multinucleated giant cells were scattered throughout the stroma (Fig. 2-C). The foci of multifocal cystic degeneration were present focally (Fig. 2-D). There was no bone marrow nor rimming of osteoblasts present. Focal calcified foci were present. Lymphoid cells were infiltrated focally at the mass, and siderophages (iron-filled macrophages) were detected in the surrounding tissue. The subepithelial oral mucosa showed hemorrhage and hyalinization of the connective tissue.

Aneurysmal bone cysts were identified in a multifocal distribution in dysplastic bone tissue. These cysts are histologically characterized by the presence of blood-filled

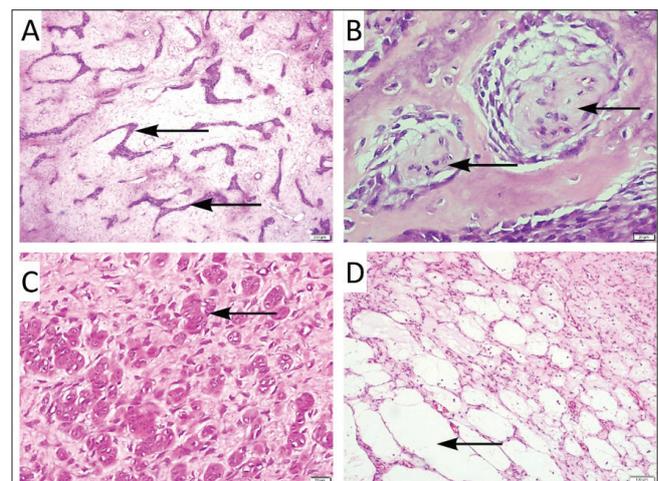


Fig 2. Histologic lesions of the fibrous dysplasia, x4, H&E. **A.** Branching and interconnecting immature bone spicules, both circular and irregular in shape (resembling Chinese characters) were embedded throughout the fibrous tissue, x40 H&E. **B.** Multinucleated giant cells (arrows) were situated in Howship's lacunae, x40 H&E. **C.** Diffusely distributed multinucleated cells (arrow), x20, H&E. **D.** Stromal cystic degeneration (arrow), x10, H&E

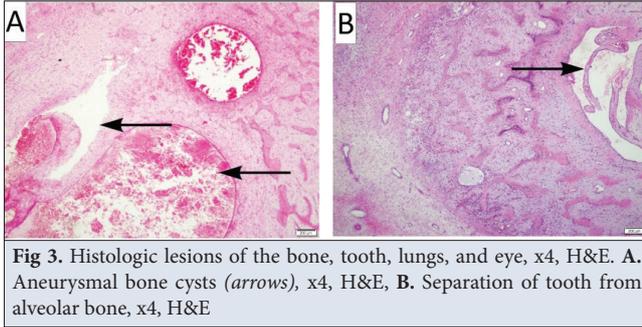


Fig 3. Histologic lesions of the bone, tooth, lungs, and eye, x4, H&E. **A.** Aneurysmal bone cysts (arrows), x4, H&E. **B.** Separation of tooth from alveolar bone, x4, H&E

cystic spaces devoid of epithelial or endothelial cells, which are separated by fibroblastic septa (*Fig. 3-A*). The incisive teeth have become detached from the alveolar bone, with the periodontal ligament remaining in position (*Fig. 3-B*). Some alveolar spaces showed deposition of meconium, hyperemia and interstitial edema. The optic nerve in the eyes showed mild papillary edema on the optic disc.

Examination of the lungs revealed marked emphysema in some lobes, atelectasis and interlobar edema.

DISCUSSION

Fibrous Dysplasia and Aneurysmal Bone Cyst are two distinct pathological entities, each characterized by its unique pathogenetic mechanisms. A review of veterinary literature has shown no record of the simultaneous occurrence of these two conditions. Nevertheless, in humans, 36 reported cases of simultaneous fibrous dysplasia and aneurysmal bone cyst, which include four instances of mandibular involvement, as identified in literature ^[13] up to 2019. The simultaneous occurrence of these conditions is attributed to the hemodynamic disturbances arising from FD ^[14]. However, this concurrent presence is not specific to FD; ABCs may also occur secondarily within or adjacent to other tumours or malignant processes, like osteoblastoma. Therefore, any factor weakening the bone structure could potentially lead to the formation of ABCs ^[13]. An alternative explanation may be attributed to alterations in bone pressure or the redistribution of mechanical stress, stemming from structural changes within the bones. Contrary to what the name “ABC” might suggest, the term is somewhat misleading since the lesion does not present the typical features of an aneurysm or a cyst, particularly due to the lack of an endothelial lining ^[13]. At present, ABCs are recognized as benign osseous lesions that may undergo rapid enlargement and cause significant compromise to the integrity of the bones they occupy ^[13]. The differentials for ABC included hemangioma, hemangiosarcoma and osteosarcoma in this report. ABC is characterized by blood-filled spaces, delineated by connective tissue housing bone trabeculae or osteoid tissue. It also has irregular vascular channels devoid of endothelial lining, lacking malignant cells and endothelial cell. However,

osteosarcoma contains malignant osteoid or bone forming tissue and hemangiosarcoma is characterized by malignant endothelial cells forming vascular structures

FD is characterized by the presence of underdeveloped bone segments, fibroblasts, hypertrophic osteoblasts, diverse trabecular bone patterns, multinucleated osteoclastic giant cells, and the absence of rimming osteoblasts in the surfaces of the trabeculae ^[6,7]. In differential diagnoses, the following lesions should be considered: non-ossifying fibroma, osteoma, adamantinoma, low-grade osteosarcoma, and Paget's disease ^[10]. The differentiation depended on the evaluation of clinical, radiological, and histopathological features in this report. In the clinical perspective, osteoma and ossifying fibroma present as palpable bony masses. Radiologically, fibrous dysplasia exhibits a distinctive ground-glass appearance. Conversely, ossifying fibroma displays a combination of radiolucent and radiopaque areas, often featuring a central calcified region. At the histopathological level, fibrous dysplasia manifests fibrous tissue with irregular trabeculae reminiscent of woven bone. In contrast, osteoma is characterized by the presence of lamellar bone ^[1,4]. Non-ossifying Fibroma has sheets of histiocytes, foam cells, and giant cells within a fibrous stroma. It does not show osteoblastic rimming, distinguishing it from fibrous dysplasia. Adamantinoma is characterized by epithelial-like nests and strands within fibrous stroma and is distinguished by the presence of epithelial components ^[10].

The current case, through its gross and histological findings, also exhibits characteristics similar to cherubism. This uncommon autoinflammatory bone disorder is characterized by the symmetrical expansion of fibro-osseous tissue in the mandible and/or maxilla, most commonly observed in children aged 2 to 5. Key features of the case -including bilateral mandibular involvement, bone tissue degradation, teeth dislodging from their alveoli, and the detection of osteoclast-like cells - indicate characteristics and are reportedly found in human cherubism ^[14]. Despite these similarities, the terminology “chimerism” remains unrecognized in veterinary literature. Nonetheless, a genetically developed animal model, specifically in mice, has been established ^[15].

In the present case, the diagnosis was based on the presence of woven bone spicules and the absence of osteoblastic rimming. The concurrent presence of fibrous dysplasia and cherubism presented here supports the theory that cherubism represents a hereditary form of fibrous dysplasia in humans ^[14]. The pulmonary lesions, including emphysema, atelectasis, and meconium aspiration, may have arisen due to the posterior displacement of the tongue caused by the effect of the mandibular mass. Similarly; separation of teeth from alveolar bone and edema of optic

nerve head were probably secondary lesions of fibrous dysplasia.

In conclusion, a unique case of mandibular fibrous dysplasia displaying certain similarities to cherubism and ABC has been recorded in a calf.

DECLARATIONS

Availability of Data and Materials: The data that support the findings of this case report are available from the corresponding author (Y. Eröksüz) upon reasonable request.

Conflict of Interest Statement: The author declares no conflict of interest

Author Contributions: Conceptualization, YE and CAI; Acquisition, analysis, and interpretation of data, YE and HE, CAI and MT; Writing - original draft preparation, Y.E.; Review and editing, H.E., CAI, MT. All authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part are appropriately investigated and resolved.

REFERENCES

1. Kim HY, Shim JH, Heo CY: A rare skeletal disorder, fibrous dysplasia: A review of its pathogenesis and therapeutic prospects. *Int J Mol Sci*, 24:15591, 2023. DOI: 10.3390/ijms242115591
2. Kim DY: Current concepts of craniofacial fibrous dysplasia: Pathophysiology and treatment. *Arch Craniofac Surg*, 24 (2): 41-51, 2023. DOI: 10.7181/acfs.2023.00101
3. Fitzgerald W, Slocombe R, Caiafa A: Fibrous dysplasia of mandibular bone in a dog. *J Vet Dent*, 19(2):77-81, 2002. DOI:10.1177/089875640201900203
4. Soltero-Rivera M, Engiles JB, Reiter AM, Reetz J, Lewis JR, Sánchez MD: Benign and malignant proliferative fibro-osseous and osseous lesions of the oral cavity of dogs. *Vet Pathol*, 52 (5): 894-902, 2015. DOI: 10.1177/0300985815583096
5. Wilson RB: Monostotic fibrous dysplasia in a dog. *Vet Pathol*, 26:449-450, 1989. DOI: 10.1177/030098588902600513
6. Raval SH, Joshi DV, Patel BJ, Sutariya P, Patel JG, Parmar SN: Monostotic fibrous dysplasia in the nasal sinus of Marwari mare: A case report. *Indian J Vet Pathol*, 39 (4): 358-359, 2015. DOI: 10.5958/0973-970X.2015.00088
7. Sennazli G, Erdogan-Bamac O, Yuzbasioglu-Ozturk G, Koenhemi L: A case of polyostotic fibrous dysplasia in a spider monkey (*Ateles paniscus*). *Kafkas Univ Vet Fak Derg*, 26 (4): 563-566, 2020. DOI: 10.9775/kvfd.2019.23798
8. Williams WM, Lombard LS, Firfer HS: Fibrous dysplasia in a monkey and a kudu. *J Am Vet Med Assoc*, 147, 1049-1052, 1965.
9. Stewart HL: Aneurysmal bone cysts as a diagnostic consideration in juvenile patients: Considerations from humans and animals. *Equine Vet Educ*, 35, 189-193, 2023. DOI: 10.1111/eve.13717
10. Biller DS, Johnson GC, Birchard SJ, Finland RB: Aneurysmal bone cyst in a rib of a cat. *J Am Vet Med Assoc*, 190, 1193-1195, 1987.
11. Bryant U, Fallon L, Lee Ma, Pool RB: Congenital aneurysmal bone cyst in a foal. *J Equine Vet Sci*, 32 (6): 320-323, 2012. DOI: 10.1016/j.jevs.2011.12.002
12. Belknap EB, Brodie S, Lowry J, Getzelman R: Aneurysmal bone cyst in a Holstein bull. *J Am Vet Med Assoc*, 201, 1413-1415, 1992. DOI: 10.2460/javma.1992.201.09.1413
13. Rau LH, Reinheimer A, Meurer MI, Marodin AL, Espezim CS, Klüppel LE, Dos Santos Vaz Fernandes PC, Rivero ERC: Fibrous dysplasia with secondary aneurysmal bone cyst: A rare case report and literature review. *Oral Maxillofac Surg*, 23 (1): 101-107, 2019.
14. Bhattacharya S, Mishra RK: Fibrous dysplasia and cherubism. *Indian J Plast Surg*, 48 (3): 236-248, 2015. DOI: 10.4103/0970-0358.173101
15. Sharma T, Cotney J, Singh V, Sanjay A, Reichenberger EJ, Ueki Y, Maye P: Investigating global gene expression changes in a murine model of cherubism. *Bone*, 176:115135, 2020. DOI: 10.1016/j.bone.2020.115315