

A Case of Polyostotic Fibrous Dysplasia in a Spider Monkey (*Ateles paniscus*)^[1]

Gulbin SENNAZLI^{1,a} Ozge ERDOGAN-BAMAC^{1,b} Gulay YUZBASIOGLU-OZTURK^{1,c} Lora KOENHEMSI^{2,d}

^[1] Presented as an oral presentation at the 7th National Veterinary Pathology Congress, 08-10 September 2014, Kars, Turkey

¹ Istanbul University-Cerrahpasa, Department of Pathology, Faculty of Veterinary Medicine, TR-34320 Istanbul - TURKEY

² Istanbul University-Cerrahpasa, Department of Internal Medicine, Faculty of Veterinary Medicine, TR-34320 Istanbul - TURKEY

ORCIDs: ^a 0000-0002-8419-2833; ^b 0000-0002-0352-4841; ^c 0000-0002-1761-0409; ^d 0000-0002-0352-4841

Article ID: KVFD-2019-23798 Received: 17.12.2019 Accepted: 30.04.2020 Published Online: 04.05.2020

How to Cite This Article

Sennazli G, Erdogan-Bamac O, Yuzbasioglu-Ozturk G, Koenhemsli 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

Abstract

This paper presents the gross and histopathologic findings of polyostotic fibrous dysplasia which is rarely seen in non-human primates. A 3-year-old female spider monkey (*Ateles paniscus*) was referred to the clinics of our faculty with the complaints of severe dyspnea, lethargy and cough and was diagnosed with dilated cardiomyopathy following the radiographic examination. However, the monkey died due to depression of respiration before treatment was done. Necropsy was performed on the request of the owner. Dilated cardiomyopathy and irregularly shaped tumoral masses with diameters of 0.5-1.0 cm in the right and left ribs were detected. Following the histopathological examination, the lesions were diagnosed as fibrous dysplasia.

Keywords: Fibrous dysplasia, Histopathologic findings, Spider monkey

Bir Örümcek Maymununda (*Ateles paniscus*) Poliostotik Fibröz Displazi Olgusu

Öz

Bu bildiri, insan olmayan primatlarda nadir olarak görülen polyostotik fibröz displazinin makroskopik ve histopatolojik bulgularını sunmaktadır. Üç yaşındaki dişi örümcek maymunu (*Ateles paniscus*) şiddetli dispne, letarji ve öksürük şikayetleri ile fakültemiz kliniklerine getirilmiştir. Yapılan radyografik muayene neticesinde dilate kardiyomiopati teşhisi konulmuştur. Ancak, herhangi bir tedavi uygulanmadan önce, solunum yetmezliği sonucunda ölüm gerçekleşmiştir. Hasta sahibinin isteği üzerine uygulanan nekropside, dilate kardiyomiopatinin yanısıra, sağ ve sol kostalarda çapları 0.5-1.0 cm arasında değişen, düzensiz şekilli tümoral kitleler tespit edilmiştir. Histopatolojik inceleme sonucunda lezyonlar fibröz displazi olarak teşhis edilmiştir.

Anhtar sözcükler: Fibröz displazi, Histopatolojik bulgular, Örümcek maymunu

INTRODUCTION

Fibrous dysplasia (FD) is a tumor-like intramedullary fibro-osseous lesion of any bone in humans and animals^[1-3]. FD is commonly described as the replacement of normal bone elements with benign cellular fibrous connective tissue within which irregular trabeculae of woven bone are haphazardly distributed^[4]. The lesions of FD develop during skeletal formation and growth and have a variable natural evolution^[2]. FD occurs throughout the skeleton with predilection for the long bones of extremities,

ribs, pelvis and craniofacial bones^[2,5], which leads to severe consequences including pathological fractures, impairment of limb function, facial and limb deformities, and compressive damage of sensory nerves resulting in blindness or deafness^[4,6]. It has three clinical patterns namely; monostotic, polyostotic, and the McCune-Albright syndrome. In monostotic fibrous dysplasia, there is single bone involvement. Ribs and craniofacial bones are the most common sites. Polyostotic fibrous dysplasia involves two or more of the long bones, pelvis bones and ribs. In McCune- Albright syndrome polyostotic form is



Correspondence



+90 212 8663700/43660



ayyildiz@istanbul.edu.tr

accompanied by endocrine disturbances and cutaneous hyperpigmentation [3,7].

Although it has been documented in several animal species, including horse, pig, dog, and domestic cat, there are still few reports of FD in animals [1,8-10]. Only three cases have been reported in non-human primates up to date [11-13]. This paper presents a case of polyostotic fibrous dysplasia which was detected in the ribs of a spider monkey.

CASE HISTORY

A 3-year-old, 4.2-kg, female spider monkey which was housed in a private zoo was referred to the Clinics of Veterinary Faculty of Istanbul University- Cerrahpasa with the complaints of severe dyspnea, lethargy and cough. She was diagnosed with dilated cardiomyopathy following the radiographic examination. There were no distinct abnormalities that could be attributed to FD in the thoracic radiograph. The monkey died due to depression of respiration before treatment was done. Necropsy was performed at the Pathology Department of Veterinary Faculty of Istanbul University- Cerrahpasa on the request of the owner. Informed consent form was obtained from the owner for the procedures to be applied to this case.

On postmortem examination, severe edema, congestion and areas of consolidation in the lungs and dilated cardiomyopathy were observed. Irregularly shaped, nine

tumoral masses with diameters of 0.5- 1 cm were detected in the left 3rd, 6th and 7th ribs and in the right 8th rib. They were moderately dense, gritty and whitish in colour. No gross lesions were noted in the skull or long bones.

Several sections of the tumoral masses and various organs were fixed in 10% neutral buffered formalin. Following decalcification in ethylenediamine tetraacetic acid (EDTA) solution the tissue samples were routinely processed, embedded in paraffin, cut at 5 μ m, stained with hematoxylin and eosin (H&E), and evaluated under light microscopy.

Histopathological examination revealed severe areas of collateral hyperemia, edema and atelectasis in the lungs. The tumoral masses of the ribs consisted of thin, curved trabeculae of woven bone embedded in a loose fibrous connective tissue stroma. There was a variably cellular fibroblastic proliferation in storiform pattern with osteoid or bony trabeculae which was poorly formed (Fig. 1-A). Collagen fibers in the stroma were generally delicate. Osteoblasts were not rimmed on trabecular surfaces (Fig. 1-B). The hook-like, long crooked spicules of bone were typically seen in specimens (Fig. 1-C). The degree of calcification of trabeculae was variable. Degeneration including hyalinization of stroma was seen in the primary lesion. There was no sign of atypia in the stroma. Clusters of giant cells resembling osteoclasts were often found in close proximity to the trabecule (Fig. 1-D). Hemorrhagic

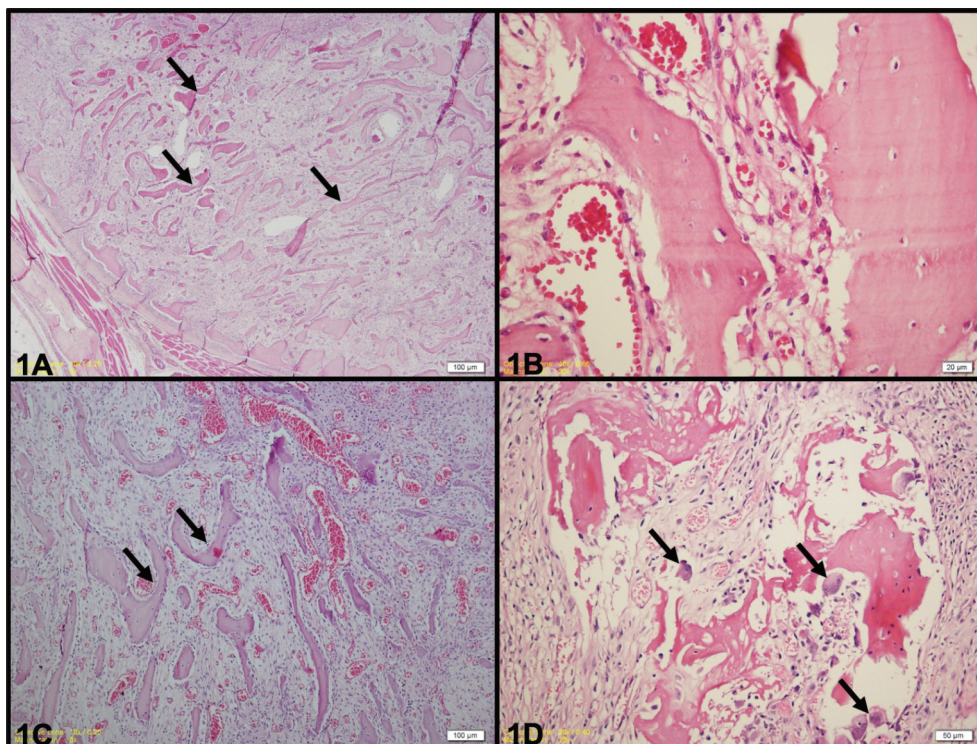


Fig 1. Osteodysplastic lesions, ribs; A: Cellular fibroblastic proliferation in storiform pattern with osteoid or bony trabeculae which was poorly formed (arrow), B: Osteoblasts were not rimmed on trabecular surfaces, C: The hook-like, long crooked spicules of bone (arrow), D: Clusters of giant cells resembling osteoclasts (arrow), rib, (H&E)

scattered areas and cartilaginous foci were present, but cysts or necrotic foci were not seen in the lesions. The number of inflammatory cells and mitotic figures were rare. No sign of malignancy was observed in any of the specimens. The lesions were diagnosed as polyostotic fibrous dysplasia according to the histopathologic findings.

DISCUSSION

Tumor-like lesions of bone are uncommon in domestic animals and even less in non-human primates [11-14]. Bauer et al. [11] reported FD in the long bones of extremities (tibia, fibula and femur) in a cynomolgus macaque and Duncan et al. [12], reported FD in the maxilla and mandibula of a 4.5-year-old male spider monkey [11,12]. In veterinary literature, in comparison with the elder ones, young animals are reported more likely to be affected by FD [15]. The spider monkey of this report was a young individual like the ones described in previous reports [11-13]. Patients with FD are generally reported to have swellings, skeletal deformities and fracture symptoms in the localization sites. Generally, lesions are difficult to distinguish from malignancies on gross and radiologic examinations because of their infrequent clinical occurrence resulting in few specimens available for study by veterinary radiologists and pathologists [1]. In this case, those findings were not observed during physical examination. In routine radiographs, global cardiac enlargement was prominent, but no distinct abnormalities attributed to FD were noticed.

Common sites of skeletal involvement of FD in humans are long bones, ribs, craniofacial bones, vertebra and pelvis [2]. In domestic animals lesions have been found in sinuses, mandible and long bones [15]. In the case reports of non-human primates with FD, lesions were present in the maxilla and in the long bones [11-13]. In this case polyostotic lesions were observed only in the ribs. Although localization of FD lesions in the ribs have been reported in humans [16,17], it has not been reported in animals according to the authors knowledge.

The pathogenesis of FD suggests that it arises from a somatic mutation of the gene associated with cell differentiation from mesenchymal stem cells into mature osteocytes [18]. At necropsy, cytogenetic analysis on the monkey was not performed as the lesions were evaluated as neoplasia. As there were no endocrine disturbances involved such as cutaneous hyperpigmentation, hyperthyroidism, hyperparathyroidism, acromegaly and no pathologic lesions in thyroid and parathyroid gland, McCune-Albright syndrome was not considered as a potential diagnosis.

Fibrous dysplasia, fibro-osseous lesion of bone, is thought to be a developmental abnormality of bone-forming mesenchyme rather than a neoplasm. Benign fibro-osseous proliferations of bone in veterinary medicine include ossifying fibroma, osteoma, osteofibrous dysplasia [14].

Sometimes the tumor-like lesions can be confused with even malign neoplasia. Thus, FD can be distinguished histopathologically from ossifying fibroma, osteoma, osteosarcoma, and perhaps even from fibrous osteodystrophy [4,18]. FD consists of bone spicules that are usually more uniform and are not rimmed by osteoblasts, which is also a feature that helps to distinguish this lesion from other fibro-osseous lesions [18]. In the present case, collagen fibers in the stroma were generally delicate and the hook-like, long crooked spicules of bone were present in specimens and there were no osteoblasts on trabecular surfaces of immature woven bone. Thus, possibility of other fibro-osseous lesions for diagnosis were eliminated and the case was evaluated as polyostotic fibrous dysplasia. Since polyostotic fibrous dysplasia of ribs in a non-human primate is rarely seen, the authors believe that this case report is going to contribute valuable information to the veterinary literature.

REFERENCES

- 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
- DiCaprio MR, Enneking WF:** Fibrous dysplasia. Pathophysiology, evaluation, and treatment. *J Bone Joint Surg Am*, 87 (8): 1848-1864, 2005. DOI: 10.2106/JBJS.D.02942
- Hartley I, Zhadina M, Collins MT, Boyce AM:** Fibrous dysplasia of bone and McCune-Albright syndrome: A bench to bedside review. *Calcif Tissue Int*, 104, 517-529, 2019. DOI: 10.1007/s00223-019-00550-z
- Riminucci M, Liu B, Corsi A, Shenker A, Spiegel AM, Robey PG, Bianco P:** The histopathology of fibrous dysplasia of bone in patients with activating mutations of the Gsa gene: Site-specific patterns and recurrent histological hallmarks. *J Pathol*, 187, 249-258, 1999. DOI: 10.1002/(SICI)1096-9896(199901)187:2<249::AID-PATH222>3.0.CO;2-J
- Dorfman HD:** New knowledge of fibro-osseous lesions of bone. *Int J Surg Pathol*, 18 (Suppl. 3): 62S-65S, 2010. DOI: 10.1177/1066896910369924
- Robinson C, Collins MT, Boyce AM:** Fibrous dysplasia/McCune-Albright syndrome: Clinical and translational perspectives. *Curr Osteoporos Rep*, 14, 178-186, 2016. DOI: 10.1007/s11914-016-0317-0
- Charpurlat RD, Orcel P:** Fibrous dysplasia of bone and McCune-Albright syndrome. *Best Pract Res Clin Rheumatol*, 22 (1): 55-69, 2008. DOI: 10.1016/j.berh.2007.11.004
- Wilson RB:** Monostotic fibrous dysplasia in a dog. *Vet Pathol*, 26, 449-450, 1989. DOI: 10.1177/030098588902600513
- 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
- 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
- Bauer C, Dunn BG, Brothman AR, Dick Jr EJ, Christensen C, Voges A, Moore CM:** Polyostotic fibrous dysplasia in a cynomolgus macaque (*Macaca fascicularis*). *Comp Med*, 62 (2): 142-148, 2012.
- Duncan JR, Lederer HA, Ramsey FK, Tyler DE:** Fibrous dysplasia in a monkey. *Iowa State Univ Vet*, 25, 81-82, 1962.
- Williams WM, Lombard LS, Firfer HS:** Fibrous dysplasia in a monkey and a kudu. *J Am Vet Med Assoc*, 147, 1049-1052, 1965.
- Slyter MV, Boosinger TR, Pool RR, Dammrich K, Misdorp W, Larsen S:** Histological classification of bone and joint tumors of

domestic animals. **In**, World Health Organization. 2nd Series, Vol. 1, 9-11, Armed Forces Institute of Pathology, American Registry of Pathology, Washington DC, 1994.

15. Thompson KG, Dittmer KE: Tumors of bones. **In**, Meuten DJ (Ed): Tumors in Domestic Animals. 5th ed., 356-424, John Wiley&Sons, Inc., 2017.

16. Mahadevappa A, Patel S, Ravishankar S, Manjunath GV: Monostotic fibrous dysplasia of the rib: A case report. *Case Rep Orthop Res*, 1-5, 2012. DOI: 10.1155/2012/690914

17. Traibi A, El Oueriachi F, El Hammoumi M, Al Bouzidi A, Kabiri EH: Monostotic fibrous dysplasia of the ribs. *Interact Cardiovasc Thorac Surg*, 14, 41-43, 2012. DOI: 10.1093/icvts/ivr048

18. Maki M, Saitoh K, Horiuchi H, Morohoshi T, Fukayama M, Machinami R: Comparative study of fibrous dysplasia and osteofibrous dysplasia: Histopathological, immunohistochemical, argyrophilic nucleolar organizer region and DNA ploidy analysis. *Pathol Int*, 51, 603-611, 2001. DOI: 10.1046/j.1440-1827.2001.01252.x