

NOTE

# Osteochondromatosis (multiple cartilaginous exostoses) in an immature killer whale *Orcinus orca*

Maíra Laeta<sup>1,2,3,\*</sup>, Erwin J. O. Kompanje<sup>4</sup>, Alastair Watson<sup>5</sup>, Sheila M. F. M. Souza<sup>6</sup>, Katharina Dittmar<sup>7</sup>, Sandra C. Cuenca<sup>8,9</sup>, Lucas B. Hassel<sup>10</sup>, Salvatore Siciliano<sup>3,11</sup>

<sup>1</sup>Programa de Pós-graduação em Biodiversidade e Biologia Evolutiva, Universidade Federal do Rio de Janeiro (UFRJ), Cidade Universitária, Rio de Janeiro, RJ 21941-902, Brazil

<sup>2</sup>Setor de Mastozoologia, Departamento de Vertebrados, Museu Nacional da Universidade Federal do Rio de Janeiro (MN/UFRJ), São Cristóvão, Rio de Janeiro, RJ 20940-040, Brazil

<sup>3</sup>Grupo de Estudos de Mamíferos Marinhos da Região dos Lagos (GEMM-Lagos), Rua São José, 1.260, Praia Seca, Araruama 28970-000, Brazil

<sup>4</sup>Natural History Museum Rotterdam, Westzeedijk 345, 3015AA Rotterdam, Netherlands

<sup>5</sup>35 Torquay Place, Bryndwr, Christchurch 8053, New Zealand

<sup>6</sup>Departamento de Endemias Samuel Pessoa, Escola Nacional de Saúde Pública Sergio Arouca, Fundação Oswaldo Cruz, Rio de Janeiro, RJ 21040-900, Brazil

<sup>7</sup>Department of Integrative Biology, 401 WIDB, Brigham Young University, Provo, UT 84602, USA

<sup>8</sup>Centro Universitário Monte Serrat (UNIMONTE), Rua Comendador Martins, 52, Santos, SP 11015-530 Brazil

<sup>9</sup>Universidade Metodista de São Paulo (UMESP), Rua Alfeu Tavares, 112, São Bernardo do Campo, SP 09641-000 Brazil

<sup>10</sup>Shell Brasil Petróleo Ltda, Barra da Tijuca, Rio de Janeiro, RJ 22640-102, Brazil

<sup>11</sup>Laboratório de Enterobactérias, Instituto Oswaldo Cruz/Fiocruz, Pav. Rocha Lima, 3º. Andar, Av. Brasil 4.365, Manguinhos, Rio de Janeiro, RJ 21041-210, Brazil

**ABSTRACT:** An immature killer whale *Orcinus orca* found dead on the southeastern Brazilian coast had multiple bone proliferations: on the skull, vertebrae, hemal arches, and ribs. The bony formations were characterized as multiple osteochondromas, as defined by osteochondromatosis. The diagnosis was based on macroscopic and radiographic observations. These benign osseocartilaginous tumors affect young individuals and grow until skeletal maturity is achieved. Case reports of this condition, besides humans, include other mammals, with most reports for pets and domestic mammals such as cattle, and a report in a fossil canid (*Hesperocyon*) from the Oligocene. The etiology, diagnosis, developmental characteristics, and occurrence of osteochondromas are distinct among different species. This report describes the first case of multiple osteochondromas in a wild cetacean.

**KEY WORDS:** Osteochondroma · Osteochondromatosis · Benign tumors · Endochondral ossification · Killer whale · Cetacea

Resale or republication not permitted without written consent of the publisher

## 1. INTRODUCTION

The killer whale *Orcinus orca* (Linnaeus, 1758) is a large-bodied cosmopolitan odontocete cetacean, most commonly observed in temperate and polar waters, especially in high-productivity areas (Ford 2009). Throughout their distribution, killer whales may occur seasonally or as year-round residents;

however, these habitat usage patterns are not fully known (Ford 2009, Pitman & Ensor 2003). Ecologically distinct groups, or ecotypes, are recognized, e.g. the distinct populations in the North Pacific and the Antarctic (de Bruyn et al. 2013).

In the Southern Atlantic Ocean, killer whales occur commonly in coastal waters of the temperate region, mainly off Argentina and southeastern Brazil (Pit-

man & Ensor 2003, Ford 2009). Information on killer whales in southeastern Brazilian waters is known from individual strandings and opportunistic coastal sightings (Dalla Rosa & Secchi 2007, Ott et al. 2017), predominantly during the austral spring and summer (Siciliano et al. 1999, Dalla Rosa & Secchi 2007). In offshore waters, sightings are known from winter and spring, often in interactions with longline fisheries (Dalla Rosa & Secchi 2007).

The few reports on skeletal pathology in the killer whale are all related to lesions of minor severity, most likely subsequent to infection of distinct parts of the skeleton including the vertebrae, rib, scapula, radius, and dental alveoli (Colyer 1938, Kompanje 1991, 1995).

Here we describe skeletal abnormalities found in an immature female killer whale stranded on the Brazilian coast. This is the first report of osteochondromatosis in a cetacean.

## 2. MATERIALS AND METHODS

A female killer whale *Orcinus orca* was found dead in a decomposed condition on Praia Grande beach (22° 57' 57" S, 42° 00' 51" W), Arraial do Cabo, Rio de Janeiro state, Brazil, on 23 July 2001. Estimated total body length (TBL) (Norris 1961) was 4.69 m.

After necropsy on the beach, the disarticulated skeleton was laid on 1/4 inch (6.35 mm) mesh hardware cloth and buried under approximately 1 m of sand, to be exhumed 6 mo later. After cleaning and drying of the recovered bones, the skeleton was articulated and mounted for exhibition at the Museu da Vida/Fundação Oswaldo Cruz, Rio de Janeiro, RJ, Brazil of the Ministério da Saúde.

The skeleton was almost complete: the cranium including 3 hyoid bones, vertebral column consisting of C1-2 (fused)-7 (cervical), T12 (thoracic), L11 (lumbar), Cd21+ (caudal) with 8 hemal arches = 51+ total vertebrae; 11 pairs of vertebral ribs, 6 pairs of sternal ribs, 3 separate sternbrae; and the ossa coxarum. The terminal few caudal vertebrae were not recovered during preparation. The 12<sup>th</sup> thoracic vertebra had distinct costal articular surfaces at the end of both transverse processes, but the associated small last pair of ribs was not salvaged. The skeleton of both flippers contained the scapula, humerus, radius, ulna, and some metacarpal and phalangeal osseous elements. The only ossified epiphysis in the limb was the proximal humeral epiphysis, and bilaterally this was well developed but incompletely fused to the body of

the humerus. In the vertebral column, the epiphyses of the vertebral bodies were at an early stage of ossification and only those of the most cranial cervical and the terminal caudal vertebrae showed any degree of coalescing with their respective vertebral bodies. The unfused or incompletely fused vertebral body epiphyses indicated a skeleton from a physically immature odontocete. The dental formula was 12 teeth in each jaw on both sides, upper and lower, total 48.

The cleaned skeleton was examined grossly by eye, and the 3<sup>rd</sup> left rib was also examined radiographically.

Standard anatomical nomenclature and directional terminology was used based on the Nomina Anatomica Veterinaria (ICVGAN 2012).

## 3. RESULTS

### 3.1. Macroscopic examination

#### 3.1.1. Skull

Two dense pathological proliferative bony masses extended caudally from the lateral aspects of the occipital plate. These cauliflower-like lesions were bilaterally symmetrical and located immediately caudal to the nuchal crest, measuring 26 × 15 × 6 cm (Fig. 1), and with other small lesions, continuous or not with the main ones, were present on the frontal, maxillary, and squamosal bones.

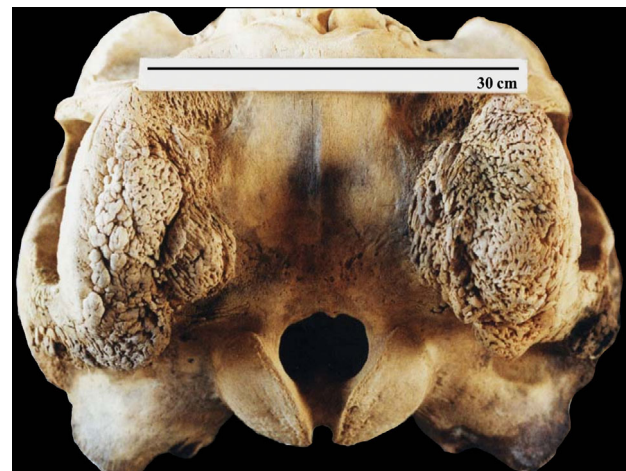


Fig. 1. Caudal view of the skull from an immature female *Orcinus orca* (total body length: 4.69 m), with symmetrical cauliflower-like bony lesions of the occipital plate extending caudally and including proliferations from the lateral edges of the frontal and squamosal bones

### 3.1.2. Vertebrae

On the 6th lumbar vertebra, there was a small irregular proliferative mass on the pedicle of its right vertebral arch. The spinous processes between the 9th lumbar and 3rd caudal vertebrae showed some bone remodeling. Separate proliferative lesions were found on 2 hemal arches (Fig. 2): a small mass measuring  $3.5 \times 5 \times 2$  cm was located on the left lateral aspect of the 7th hemal arch; and a larger proliferation measuring  $10.5 \times 14.5 \times 6$  cm on the 8th hemal arch extended from the right side and caudally to near the 9th hemal arch.

### 3.1.3. Ribs

The 3rd and 6th left ribs each had a circumferential bony proliferation near their sternal ends. The 3rd rib was more severely affected, with a prominent ovate calcified mass measuring  $17 \times 12 \times 7$  cm, which had many vascular channels and embraced the original contours of the rib (Fig. 3). The general contour of the bony mass was regular, well defined, and bulged outward mainly laterally. At the proximal and distal ends of the tumor, the superficial cortical surface of the rib was smooth and thick, with the diameter of the entire rib increasing from the normal portion to the tumor portion due to the periosteal apposition. In the cleaned bones, overlying parts of the swellings growing along the surface of the normal bone were clearly visible, leaving a space formerly filled by soft structures. The bone structure of the rib deep to the lesion appeared grossly normal. The 6th rib showed a smaller lesion at the sternal end measuring about 4 cm in diameter.

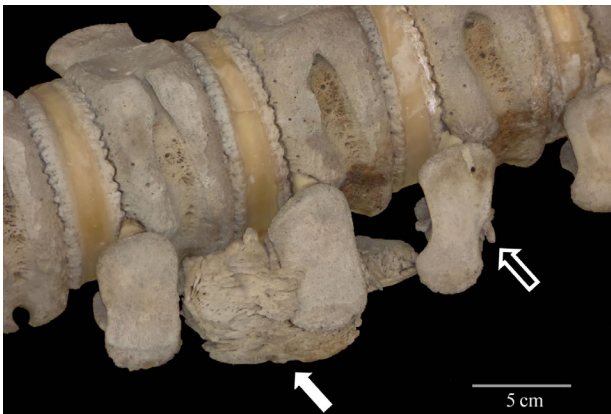


Fig. 2. Hemal arches no. 7 (minor exostosis on left side, unfilled arrow) and no. 8 (extensive exostosis, filled white arrow) from an immature female *Orcinus orca* (total body length: 4.69 m). Right lateral view; cranium is to the right



Fig. 3. Third left rib from an immature female *Orcinus orca* (total body length: 4.69 m), showing a dense, proliferative bony mass that surrounds most of the distal end of this rib (arrow)



Fig. 4. Radiograph of the distal end of the 3rd left rib from an immature female *Orcinus orca* (total body length: 4.69 m), showing the proliferative bony mass seen in Fig. 3

## 3.2. Radiographic examination

Radiographic images showed an anomalous proliferative bony mass with well-defined external margins encompassing the left 3rd (Fig. 4) and 6th ribs distally. There were no cavities except those of the vascular channels. Dense masses of appositional bone in irregular pattern were centered around the original shaft of each rib, which corroborated with the cauliflower-like appearance seen macroscopically (Fig. 3).

## 4. DISCUSSION

This is the first reported case of osteochondromatosis (multiple osteochondromas) in a cetacean. Our diagnosis in this immature female killer whale was supported by macroscopic and radiographic observations, in which the anatomic location, cranial symmetry, growing pattern, and associated periosteal reaction were comparable to that reported

in humans and other mammals (Bovée & Hogendoorn 2002).

Osteochondromatosis is represented by the development of multiple osteochondromas, which are benign osseocartilaginous tumors arising from the cartilage-capped bone surface formed by endochondral ossification (Bovée et al. 2002, Jim et al. 2012). The axial distribution of lesions in this killer whale contrasts with that more commonly seen in other mammals in which the affinity for multiple osteochondromas is in long bones. Osteochondromatosis is also known as diaphyseal aclasis, multiple cartilaginous exostoses, and hereditary multiple exostoses. The lesions often appear as pedunculated or sessile protuberances from their bony base, and sometimes have a cauliflower-like appearance (Solomon 1963, Bovée et al. 2002, Jim et al. 2012), as was well demonstrated in the killer whale in the present report, especially on the occipital plate (Fig. 1). Multiple osteochondromas only affect young individuals, with tumor growth ceasing when skeletal maturity is achieved (Bovée et al. 2002, Thompson & Pool 2002).

In most cases, osteochondromas, which have an affinity for long bones, are asymptomatic, although these slow-growing tumors may cause complications and discomfort depending on the location and size of the lesions (Thompson & Pool 2002, North & Banks 2009, Jim et al. 2012). Complications include fractures, bone deformity, vascular injury (arterial or venous stenosis), pseudoaneurysms, and neurological compromise, such as from compression of the spinal cord or nerve roots in domestic mammal pets and farm mammals (Prata et al. 1975, Thompson & Pool 2002, de Brot et al. 2013). As benign lesions, these osteochondromas usually show no propensity for metastasis, and only occasionally may undergo malignant transformation. The most frequent malignant transformation of osteochondroma is into chondrosarcoma (Bovée et al. 2002, North & Banks 2009, Jim et al. 2012), and less frequently into fibrosarcoma or osteosarcoma (Bovée et al. 2002, North & Banks 2009, Jim et al. 2012).

Multiple osteochondromas is a known autosomal dominant hereditary disorder in humans (Solomon 1964, Bovée & Hogendoorn 2002) and in some domestic mammals (Thompson & Pool 2002, Rosa & Kirberger 2012). In humans and mice, the heterogeneous disorder has 2 genes extensively related to its manifestation, *EXT1* and *EXT2*, and the involvement of a third gene, *EXT3*, is strongly suspected (Bovée & Hogendoorn 2002, Jones et al. 2010). The *EXT1* and *EXT2* genes encode, respectively, exostosin glycosyltransferase 1 and 2. These enzymes participate in the

biosynthesis of the proteoglycan heparan sulfate (HS), which regulates a range of cellular metabolic activities, especially increasing the efficiency of fibroblast growth factor (Lind et al. 1998).

There are reports of multiple osteochondromas in humans (Bovée et al. 2002) and in various domestic mammals, including dogs (Prata et al. 1975), cats (Pool & Carrig 1972), horses (Leone et al. 1987), pigs (de Brot et al. 2013), and laboratory rats (Ernst et al. 1992). Reports in captive or free-ranging wild mammals are rare, but include free-ranging white-tail deer (Kierdorf et al. 2017), rhesus macaque (Matthews et al. 2012), white rhinoceros (Smit et al. 2016), and an extremely rare case in a fossil canid *Hesperocyon* (Wang & Rothschild 1992). To this short list of cases in wild mammals, we now add the first cetacean.

In domestic cats, osteochondromas are strongly linked with the feline leukemia virus (Rosa & Kirberger 2012). Unlike 'classic' osteochondromas in other domestic mammals, where lesions are primarily in the long bones, those in cats develop in flat bones and other bones derived from intramembranous ossification, and thus will grow uninterruptedly, progressively enlarging in physically mature individuals (North & Banks 2009). Furthermore, these are relatively fast-growing tumors, with high conversion rates to malignancy, up to 20% (Thompson & Pool 2002, Rosa & Kirberger 2012).

In conclusion, the distinctive skeletal tumors described in an immature female killer whale, stranded dead on the Brazil coast, were, based on our gross anatomical and radiological observations, diagnosed as multiple osteochondromas, or osteochondromatosis. This case report documents the first example of this rare pathology in a wild cetacean.

*Acknowledgements.* We thank Bianca de Sanctis, Fagner A. de Magalhães, Andréa Carneiro, Aniela Manço, and Patrícia Golodne for their assistance during necropsy and recovery of the skeleton. Field work was supported by Cetacean Society International and Yaqu Pacha.

#### LITERATURE CITED

- Bovée JVMG, Hogendoorn PCW (2002) Multiple osteochondromas. In: Fletcher CDM, Unni KK, Mertens F (eds) World Health Organization classification of tumours. Pathology and genetics of tumours of soft tissue and bone. IARC Press, Lyon, p 360–362
- ✦ Bovée JVMG, Sakkars RJB, Geirnaerd MJA, Taminiau AHM, Hogendoorn PCW (2002) Intermediate grade osteosarcoma and chondrosarcoma arising in an osteochondroma. A case report of a patient with hereditary multiple exostoses. *J Clin Pathol* 55:226–229



- Colyer JF (1938) Dento-alveolar abscess in a grampus (*Orca gladiator* Bonn.). *Scott Nat* 230:53–55
- ✦ Dalla Rosa L, Secchi ER (2007) Killer whale (*Orcinus orca*) interactions with the tuna and swordfish longline fishery off southern and southeastern Brazil: a comparison with shark interactions. *J Mar Biol Assoc UK* 87:135–140
- ✦ de Brot S, Grau-Roma L, Vidal E, Segalés J (2013) Occurrence of osteochondromatosis (multiple cartilaginous exostoses) in a domestic pig (*Sus scrofa domesticus*). *J Vet Diagn Invest* 25:599–602
- ✦ de Bruyn PN, Tosh CA, Terauds A (2013) Killer whale ecotypes: is there a global model? *Biol Rev Camb Philos Soc* 88:62–80
- ✦ Ernst H, Karbe E, Mohr U, Nolte T, Sander E (1992) Osteochondroma in laboratory rats: a report of 3 cases in a Fischer-344, a Sprague-Dawley, and a Wistar rat. *Toxicol Pathol* 20:264–267
- Ford JKB (2009) Killer whale *Orcinus orca*. In: Perrin WF, Würsig B, Thewissen, JGM (eds) *Encyclopedia of marine mammals*. Academic Press, San Diego, CA, p 650–657
- ✦ ICVGAN (International Committee on Veterinary Gross Anatomical Nomenclature) (2012) *Nomina Anatomica Veterinaria*, 5th edn (rev.). Editorial Committee of ICVGAN, Hannover. [www.wava-amav.org/downloads/nav\\_2012.pdf](http://www.wava-amav.org/downloads/nav_2012.pdf)
- Jim S, Wu MD, Mary G, Hochman MD (2012) *Bone tumors — a practical guide to imaging*. Springer, New York, NY
- ✦ Jones KB, Piombo V, Searby C, Kurriger G and others (2010) A mouse model of osteochondromagenesis from clonal inactivation of *Ext1* in chondrocytes. *Proc Natl Acad Sci USA* 107:2054–2059
- ✦ Kierdorf U, Miller KV, Flohr S, Gomez S, Kierdorf H (2017) Multiple osteochondromas of the antlers and cranium in a free-ranging white-tailed deer (*Odocoileus virginianus*). *PLOS ONE* 12:e0173775
- Kompanje EJO (1991) Een oud geval van osteomyelitis bij een orka *Orcinus orca*. *Lutra* 34:71–76
- Kompanje EJO (1995) Strandings of killer whales *Orcinus orca* in the Netherlands between 1783 and 1995 with some remarks on skeletal and dental pathology (Mammalia, Cetacea, Odontoceti). *Deinsea* 2:67–82
- ✦ Leone NC, Shupe JL, Gardner EJ, Millar EA, Olson AE, Phillips EC (1987) Hereditary multiple exostosis. A comparative human-equine-epidemiologic study. *J Hered* 78:171–177
- ✦ Lind T, Tufaro F, McCormick C, Lindahl U, Lidholt K (1998) The putative tumor suppressors EXT1 and EXT2 are glycosyltransferases required for the biosynthesis of heparan sulfate. *J Biol Chem* 273:26265–26268
- ✦ Matthews KA, Strait K, Connor-Stroud F, Courtney CL (2012) Osteochondromatosis in a rhesus macaque (*Macaca mulatta*). *Comp Med* 62:149–152
- ✦ Norris KS (1961) Standardized methods for measuring and recording data on the smaller cetaceans. *J Mammal* 42:471–476
- North S, Banks T (2009) *Small animal oncology: an introduction*. Elsevier, Philadelphia, PA
- Ott PH, Sucunza F, Wickert J, Danilewicz D, Tavares M (2017) Evidences of attack of killer whale on a calf southern right whale in Southern Brazil. *Mastozool Neotrop* 24:235–240
- Pitman RL, Ensor P (2003) Three forms of killer whales (*Orcinus orca*) in Antarctic waters. *J Cetacean Res Manag* 5:131–139
- ✦ Pool RR, Carrig CB (1972) Multiple cartilaginous exostosis in a cat. *Vet Pathol* 9:350–359
- ✦ Prata RG, Stoll SG, Zaki FA (1975) Spinal cord compression caused by osteocartilaginous exostoses of the spine in two dogs. *J Am Vet Med Assoc* 166:371–375
- ✦ Rosa C, Kirberger RM (2012) Extraskeletal osteochondroma on a cat's elbow. *J S Afr Vet Assoc* 83:104
- Siciliano S, Lailson-Brito A Jr, Azevedo AF (1999) Seasonal occurrence of killer whales. *Z Säugetierkd (Mamm Biol)* 64:251–255
- ✦ Smit Y, Steyl J, Marais J (2016) Solitary osteochondroma of the distal third metacarpal bone in a two-year-old white rhinoceros (*Ceratotherium simum*). *J Zoo Wildl Med* 47:1086–1089
- ✦ Solomon L (1963) Hereditary multiple exostosis. *J Bone Joint Surg Br* 45B:292–304
- ✦ Solomon L (1964) Hereditary multiple exostosis. *Am J Hum Genet* 16:351–363
- Thompson KG, Pool RR (2002) Tumors of bones. In: Meuten DJ (ed) *Tumors of domestic animals*. Iowa State University Press, Ames, IA, p 245–317
- ✦ Wang X, Rothschild BM (1992) Multiple hereditary osteochondroma in Oligocene *Hesperocyon* (Carnivora: Canidae). *J Vertebr Paleontol* 12:387–394

Editorial responsibility: Michael Moore,  
Woods Hole, Massachusetts, USA

Submitted: August 6, 2018; Accepted: March 13, 2019  
Proofs received from author(s): May 5, 2019