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DIFFUSE IDIOPATHIC SKELETAL HYPEROSTOSIS IN AN ORANGUTAN (*PONGO P. PYGMAEUS*) IN THE AMSTERDAM ZOO
(A case report)

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Summary

In this paper the alterations of the skeleton of a 42 year old, Bornean Orangutan (*Pongo pygmaeus pygmaeus*, Linnaeus, 1760) are described. The skeleton showed typical hyperostosis and calcification on the right side of the thoracic vertebrae, consistent with the skeletal disease known in human pathology under the name of Diffuse Idiopathic Skeletal Hyperostosis (DISH, ankylosing hyperostosis). This disease is well known in man, but to our knowledge this is the first description of a case of DISH in an Orangutan.

Zusammenfassung

Die vorliegende Arbeit beschreibt die Veränderungen am Skelett eines 42-jahrigen Borneo-Orang-Utans (*Pongo pygmaeus pygmaeus*, Linnaeus, 1760). Das Skelett zeigte charakteristische Hyperostosen und Kalkablagerungen an der rechten Seite der Brustwirbel, die mit einer aus der Humanpathologie bekannten Krankheit des Skeletts übereinstimmten, nämlich der diffusen idiopathischen Hyperostosis des Skeletts (DISH, ankylosierende Hyperostose). Diese Krankheit ist vom Menschen gut bekannt, dievorliegende Arbeit ist aber unseres Wissens eine Erstbeschreibung beim Orang Utan.

Résumé

Cet article décrit les altérations du squelette d'un orang utan de Bornéo (*Pongo pygmaeus pygmaeus*, Linnaeus, 1760) âgé de 42 ans. Le squelette présenta des hyperostoses et des dépôts de calcium caractéristiques sur le côté droit des vertèbres thoracales. Les altérations sont identiques avec celles d'une maladie du squelette humain, à savoir l'hyperostose idiopathique du squelette (DISH, hyperostose ankylosante). Cette maladie est bien connue chez l'homme, ceci en est par contre, à notre connaissance, la première description chez un orang utan.

Keywords
Primates, Orangutan, *Pongo pygmaeus pygmaeus*, vertebra, pathology, DISH, ankylosing hyperostosis.

Introduction

Diffuse Idiopathic skeletal Hyperostosis (DISH) or ankylosing hyperostosis is a common skeletal disease of unknown etiology in man. Features consistent with DISH have been described in literature under different names (Table 1).

Table 1
Synonyms for DISH

- Moniliform hyperostosis (13)
- Spondylitis ossificans ligamentosa (14)
- Senile ankylosing hyperostosis of the spine (7)
- Ankylosing hyperostosis (10)
- Hyperostosis of the spine (8)
- Diffuse idiopathic skeletal hyperostosis (17)

The disease occurs after middle age, and more often in males than in females. The principal manifestations are ligamentous calcification and ossification of the anterolateral (anti-cardiac) part of the spinal column, sometimes leading to ankylosis. The appearance of the osteophytes has been described as 'dripping candle wax', involving three or four vertebral bodies. DISH is distinguished from ordinary spondylosis deformans by the absence of disc degeneration. The distal thoracic spine is the part of predilection. There is, in contrast with spondylosis deformans, an absence of erosions and sclerosis of the vertebral epiphysis.

The appearance of the vertebral pathology has often led to confusion with ankylosing spondylitis and with spondylosis deformans. DISH and spondylosis deformans are often found together in one individual. In some cases there may be extraspinal manifestations in the form of bony spurs around the patellae, olecranon and calcaneus (16, 25). In humans this occurs in about 35 % of all cases (25).

Patients often complain of dysphagia due to compression of the oesophagus by the osteophytes. The compression is severe enough to make eating solid food a strenuous chore. Hyperglycaemia may occur and the incidence of diabetes mellitus is about 40%. The therapy for DISH is symptomatic. As the name already indicates, the etiology and pathogenesis of DISH is unknown.

Research so far has focused on possible metabolic-endocrine and toxic factors. The relation between diabetes mellitus and DISH may provide some insight. Diabetes is frequently associated with rheumatic conditions proliferation of fibrous tissue (6).

Results

Clinical history

On 27 January 1992, a male Orangutan (*Pongo pygmaeus pygmaeus*, Linnaeus, 1760), named 'Sam', died of old age in the Zoological Gardens of Amsterdam (Artis), the Netherlands. The animal was born in 1950 on Borneo, Indonesia, and arrived in the zoo on 12 December 1952. The skin and skeleton of the animal were prepared at the Zoological Museum, University of Amsterdam. Forty-two years is extremely old for a Bornean Orangutan (2, 3).

Necropsy of Orangutan "Sam" was performed one day after death at the Veterinary Faculty, Utrecht, The Netherlands. A mild splenomegaly and some chronic ulceration of the colon were found. A few mesenterial lymph nodes were enlarged, and had a purulent content. The unusual pathology of the vertebrae became visible after cleaning the skeleton by maceration.

Pathology

The atlas showed a small osteophyte on the inner side of the lateral mass. The second cervical vertebra showed severe pathological changes (Fig.1), particular the left inferior articular process. The whole area of contact was covered with perforations and there were marginal osteophytes. The spinous process was curved to the right. The body of this vertebra also showed pathological changes with marginal osteophytes. These features were consistent with cervical spondylosis deformans. The third and the fourth cervical vertebrae also showed marginal osteophytes. The following three cervical vertebrae (5-7) were fused together by grotesque new bone formation.

The intervertebral spaces, however, were normal and showed no perforations and/or sclerosis of the epiphysis. The first thoracic vertebra showed normal epiphysis, but severe marginal osteophytes. The following four thoracic vertebrae were also fused together by a grotesque ankylosis (Fig. 2,3 and 4).

Again, the intervertebral spaces and epiphyses were normal. The following thoracic and lumbar vertebrae showed only degenerative changes consistent with the age of the animal. The other extraspinal bones of the skeleton and the skull showed no gross pathology. The teeth were worn and the jaws showed the results of peridontitis, which is normal for a primate that has been living in a zoo for nearly forty years.

Discussion

There is limited literature on osteopathology in primates. Especially Rothschild and Woods did extensive work on spondyloarthropathy in Great Apes, Lesser Apes and Old World Monkeys (19, 20, 21, 22, 23). Records of osteopathology in Orangutans are rare. The pathology seen in Orangutans is different from the pathology seen in Gorillas, Chimpanzees and Lower Primates.

Ottow (15) studied several skeletons of Gorilla (*Gorilla gorilla*), Chimpanzee (*Pan troglodytes*) and Orangutan. Changes indicative of spondylosis deformans were not found in the 22 studied skeletons of Orangutan. Ottow (15) cited (9), who examined 41 skeletons of Orangutan, two of which showed vertebral deformations consistent with degenerative disc disease. Sokoloff et al. (24) described the pathologic findings in two cases of spinal ankylosis in Rhesus Monkeys (*Macaca mulatta*). One resembled spondylitis ankylopoetica. The other was diagnosed as hyperostotic spondylosis, which is an obsolete name for DISH. Hyperglycaemia was also found in this case, suggesting DISH. Clevenger et al. (5) speculated on the possible role of *Mycoplasma* infections in the genito-urinary and upper respiratory tract, and its possible role in the etiology of rheumatoid arthritis in primates. Hime et al. (11) described a case of hypertrophic pulmonary osteoarthropathy in an 14 year old Orangutan. Adams et al. (1) described a case of spondylitis ankylopoetica in a 30 year old female Gorilla. Rothschild and Woods (20) studied 99 skeletons of adult Lowland Gorillas (*Gorilla g. gorilla*) for the evidence of monoarticular or osseous pathology. In eight skeletons they found moderate to severe osteoarthritis. In 20 individuals they found sacroiliac and reactive erosive pathology (spondyloarthropathy) suggesting reactive spondyloarthropathy. Rothschild and Woods (22) studied furthermore no less than 1699 skeletons of Old World primates including 54 Orangutans. They found a high frequency of reactive spondyloarthropathy in Great Apes (*Gorilla* and *Pan*) and Lower Primates, but not in Orangutans. The bone-pathology they found in the five Orangutans was quite different from those they observed in other Great Apes. The Orangutan is, as Rothschild and Woods (22) also mention, an 'odd' primate with probably species specific arthritis.

The literature on the occurrence of DISH in man is extensive. However records of DISH in other mammals than man, or other animals are very rare and the existing records are not very convincing. Woodard et al. (26) gave a description of the radiographic and gross pathological findings in a skeleton of a 6 year old domestic dog (Labrador retriever) and concluded that the animal had suffered from DISH. They named it 'Canine diffuse idiopathic skeletal hyperostosis'. The features in this case were however not convincingly consistent with the diagnostic criteria for the diagnosis of DISH used for epidemiologic purposes in human populations (25). Bjorkengren (4) studied skeletal remains of the prehistoric Saber-toothed Cat (*Smilodon californicus*). They found 48 fused thoracic and lumbar vertebrae, which showed, according to the authors, characteristics of three major pathologic processes: trauma, DISH and reactive arthritis similar to ankylosing spondylitis. Rothschild (18) studied over 200.000 bones of Dinosaurs and early mammals for pathological changes. The author accepted the presence of vertebral bridging without zygoapophyseal joint erosion and fusion, spinal syndesmophyte formation, sacroiliac disease or vertebral endplate destruction as evidence for DISH. He defined bridging as osseous overgrowth uniting or transgressing vertebral disc space(s) extrinsic to the site of attachments of the annulus fibrosus. The author concluded that DISH was present in a large number of dinosaurs according to his criteria. Again the criteria used here are different from the diagnostic criteria for DISH in man (25). Rothschild (19) reports briefly, without detailed description, about 'Sinbad', a Gorilla which manifested classic changes of DISH.

The features in the described case of the Orangutan "Sam" are inconsistent with discarthrosis (spondylosis deformans), ankylosing spondylitis or spondyloarthropathy. The intervertebral spaces of the affected part of the spinal column were normal and the epiphysis showed no sign of degeneration. The bizarre osteophytes are atypical for discarthrosis. The typical 'dripping candle wax' osteophytes on the anticardiac side are diagnostic for DISH. Furthermore there was a continuous calcification and ossification along the anterolateral aspect of at least four contiguous vertebral bodies. In the involved areas the disk space was essentially normal and there was absence of apophyseal joint ankylosis in the affected areas and absence of erosions and sclerosis of the sacroiliac joints. These features are diagnostic for DISH in man.

Unfortunately, glucose levels in the blood of 'Sam' were not measured, but no clinical signs of diabetes mellitus were observed. The last two month of his life he only ate soft food. This could be due to the dysphagia which is diagnostic for DISH in humans. The last six years of his life 'Sam' showed stiffness, especially during winter. Almost 40% of human patients with DISH complain about stiffness aggravated by cold and wet weather (25).

Maybe this is not the first case of DISH in an other mammal than man, as we reported earlier (12). However, it is the first case in an Orangutan. Of the reported cases so far, the features in this case are most similar with those which are diagnostic for DISH in man.

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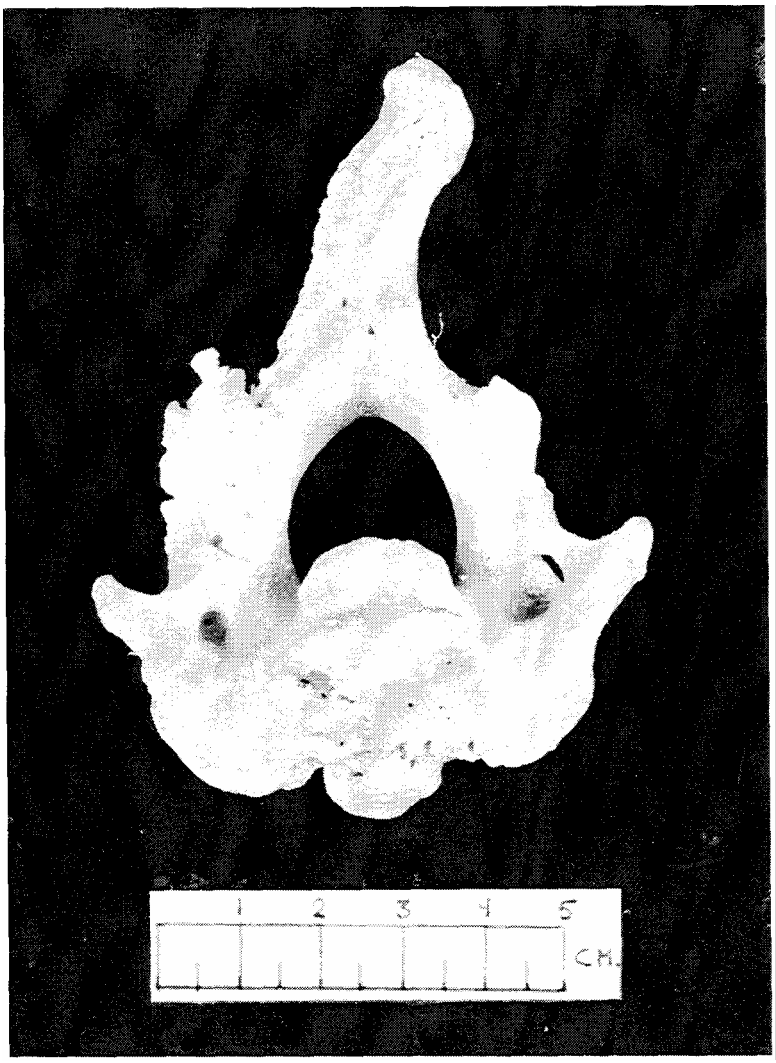

Fig. 1 *Pongo p.pygmeus*. Second cervical vertebra; note the severe bone change on the left inferior process. [Photogr. Rob 't Hart]

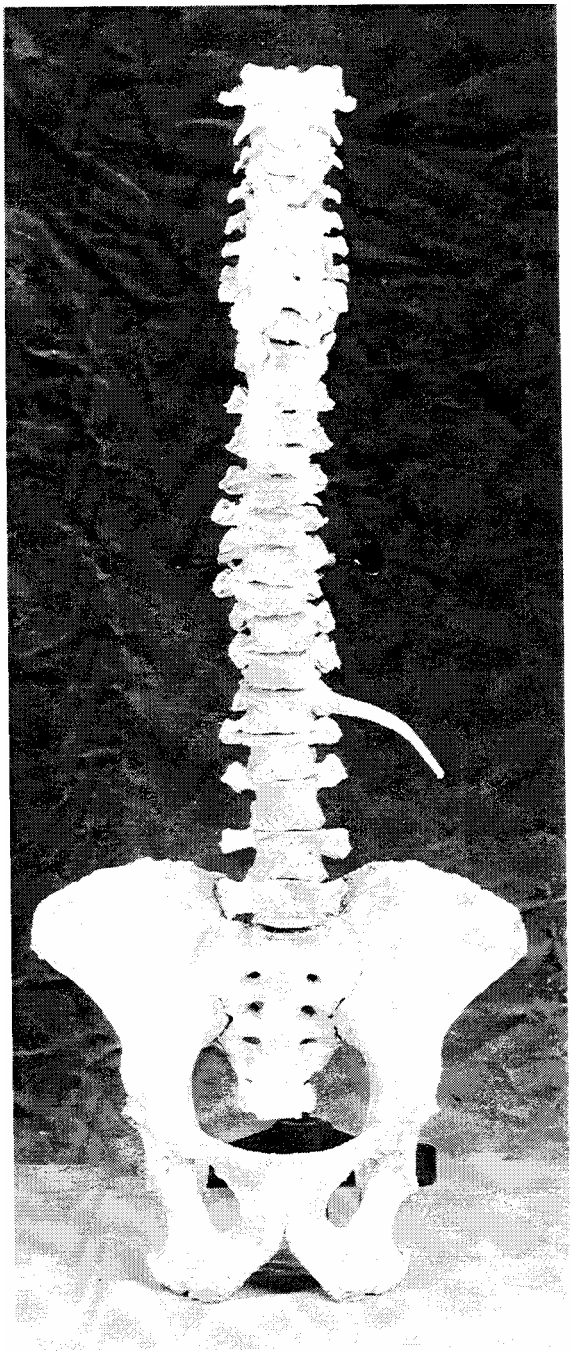

Fig. 2 *Pongo p.pygmeus.* Mounted vertebral column and pelvis showing the characteristic 'dripping candle wax' osteophytes. [Photogr. Rob 't Hart]

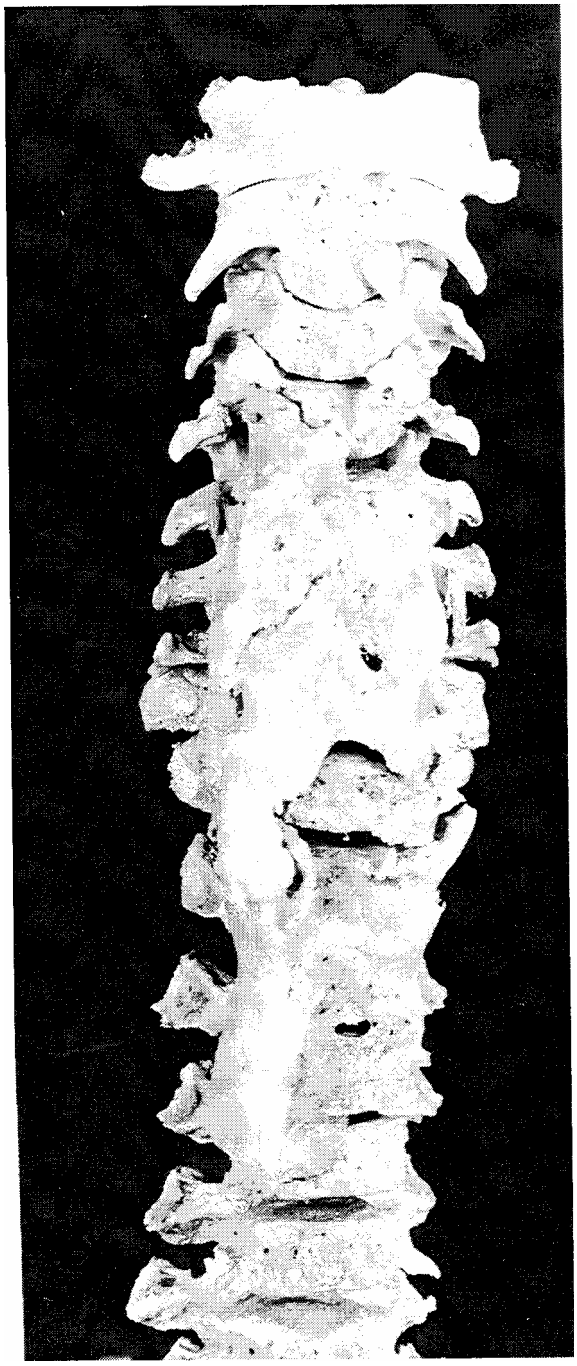

Fig. 3 *Pongo p.pygmeus*. Detail of the cervical and upper thoracic vertebrae. [Photogr. Rob 't Hart]

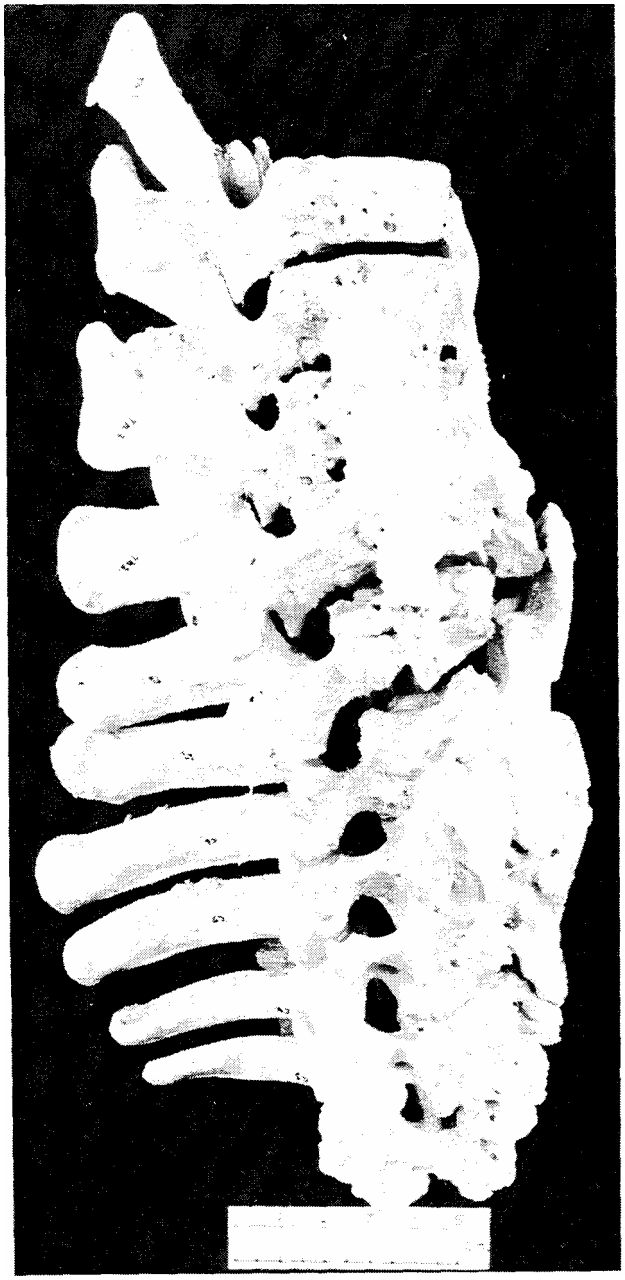

Fig. 4 *Pongo p.pygmeus*. Lateral view of the third to fifth thoracic vertebra. [Photogr. Rob 't Hart]