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What a Stranded Whale With Scoliosis Can Teach us About Human Idiopathic Scoliosis

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Abstract

Scoliosis is a deformation of the spine that may have several known causes, but humans are the only mammal known to develop scoliosis without any obvious underlying cause. This is called 'idiopathic' and is the most common type. Recent observations showed that human scoliosis, regardless of its cause, has a relatively uniform three-dimensional anatomy. We hypothesize that scoliosis is a universal compensatory mechanism of the spine, independent of cause and/or species. We had the opportunity to study the rare occurrence of scoliosis in a whale (*Balaenoptera acutorostrata*) that stranded in July 2019 in the Netherlands. A multidisciplinary team of biologists, pathologists, veterinarians, taxidermists, radiologists and orthopaedic surgeons conducted necropsy and imaging analysis. (Dorso-)lateral blunt traumatic injury to two vertebrae caused an acute lateral deviation of the spine, which had initiated the development of compensatory curves in regions of the spine without anatomical abnormalities. Three-dimensional analysis of these compensatory curves showed strong resemblance with different types of human scoliosis, amongst which idiopathic. This suggests that any decompensation of spinal equilibrium can lead to a uniform response, regardless of underlying cause or species. The unique biomechanics of the upright human spine, with significantly decreased rotational stability, explains why only in humans this universal mechanism of scoliosis can occur without an obvious cause, and is thus still called 'idiopathic'.

Introduction

Scoliosis is a three-dimensional (3D) deformity of the spine and trunk, in which rotation of the vertebral column in the horizontal plane together with extension in the sagittal plane plays a consistent role, that may be caused by traumatic injury, syndromic conditions, congenital malformations or neuromuscular disease.[1] In mammals, the development of scoliosis without an obvious underlying cause is exclusively observed in humans, this is called 'idiopathic' scoliosis and is the most frequently observed type.[1–3] The condition occurs with a prevalence of 1–4% in otherwise healthy individuals, most commonly adolescent females.[1] Treatment is currently focused on limiting progression of the spinal curve until skeletal maturity, which can necessitate bracing therapy or spinal fusion surgery.[1] Many theories have been brought forward in search of its aetiology.[1,4–12] Upright human spinal biomechanics was shown to play an important role,[13–19] and while the largely uniform shape of the scoliotic spine has been described for over a century,[10,20,21] recent observations have shown that the 3D anatomy is very uniform across different types of human scoliosis, with most of the deformity present as anterior lengthening of the intervertebral disc.[22–26] We hypothesize that scoliosis is a universal compensatory mechanism of the spine, that consists of vertebral rotation into the convexity of the curve, accompanied by anterior lengthening of the intervertebral discs, that can be caused by different primary challenges to spinal equilibrium. One of these challenges, and a possible explanation of idiopathic scoliosis in humans is the unique upright posture with the centre of weight balanced straight above the pelvis, resulting in a unique biomechanical loading of the trunk.[13–17, 27] The unique sagittal profile of the spine in man, is observed to lead to specific areas with decreased rotational stability, making it more prone - under still undetermined circumstances - to start a rotational decompensation than other spines in nature.[18,19] Scoliosis is found rarely in other vertebrates than humans.[28,29] Currently, it is unknown whether scoliotic spines of other mammals share the same 3D anatomy as in different types of human scoliosis. Recently, we had the opportunity to study a whale with scoliosis, a sea mammal that is not known to develop scoliosis spontaneously. There are several reports on cetaceans with scoliosis, however all cases have a clear cause which is mostly of traumatic origin, e.g. following ship collision.[30–32] In the current study we examined a young common minke whale (*Balaenoptera acutorostrata*), which was found stranded in July 2019 in the Netherlands with an obvious spine trauma and subsequent scoliosis (Fig. 1). This (dorso-)lateral blunt trauma to the spine initiated compensatory 3D curves in the initially anatomically unaffected spine. We were interested in these compensatory curves, as they could provide insights into the more general, intrinsic mechanisms that govern alignment of the mammalian spine. A multidisciplinary team of biologists, pathologists, veterinarians, taxidermists, radiologists and orthopaedic surgeons studied the whale and conducted a necropsy and 3D imaging analysis of the spine and compared the findings to non-scoliotic whales. The aim of the study was to assess whether scoliosis is a universal compensatory mechanism that occurs independent of cause and/or species. The hypothesis tested in the current study was that the injured whale would re-align its trunk by creating compensatory curves in the essentially normal spine and that these curves show a similar 3D configuration as is observed in human scoliosis.

Methods

Post mortem examination

Since 2008, cetaceans that stranded dead or died shortly after stranding on the Dutch coast are subjected to post mortem examination, which is conducted at the division of pathology the Faculty of Veterinary Medicine (Utrecht University). The animals described in the current study were not used for scientific or commercial testing. All were free-living whales which died of natural causes or were euthanized on welfare grounds and not for the purpose of this, or other studies. Therefore, since there was no handling of live animals in the current study, according to institutional guidelines, no consent from the Animal Use Committee was required, and animal ethics committee approval was not applicable to this work. On the 8th of July 2019, a young common minke whale washed up on the North Sea beach of Texel, the Netherlands (Fig. 1), and subsequently underwent post mortem investigation aiming to determine its cause of death. A necropsy and tissue sampling procedure was conducted following internationally standardized guidelines.[43] This included the collection of the following measures: total length (measured from the tip of the rostrum to the fluke notch, in a straight line next to the body, in cm), weight (kg) and blubber thickness. The latter was measured immediately anterior to the dorsal fin at three locations (dorsal, lateral and ventral, in mm). Age class was determined based on total length and gross examination of reproductive organs. Tissue samples from various organs, as well as the vertebral bone, were fixed in 4% phosphate-buffered formalin, embedded in paraffin, cut into 4 µm sections, and stained with haematoxylin and eosin. Samples from vertebra were decalcified prior to paraffin imbedding and staining procedures.

Diagnostic Imaging

Upon gross examination of the whale, the spinal malformation was noted. The entire vertebral column was therefore wrapped in plastic sheets and submitted for computed tomography (CT)-scanning. The spine was positioned in ventral recumbency on the table of a 64-slice sliding gantry CT scanner (Somatom Definition AS, Siemens AG, München, Germany).

Control group

As common minke whale strandings infrequently occur on the Dutch coast, a control group of the same species was not possible to acquire. Therefore, a control group was assembled of the harbour porpoise; a smaller member of the cetacean family and the most abundant whale species in the North Sea. Harbour porpoises are regularly subjected to post mortem examination and in a previous study focusing on their anatomy, animals were subjected for full-body CT-scan prior to the necropsies.[44] Ten cases which did not present spinal abnormalities and were positioned straight during CT-scanning were selected from this database and used as a control group in this study.

CT measurements

The orientation of the scanned whales in this study was defined the same way as in humans, with anterior indicating the ventral side and posterior indicating the dorsal side, and furthermore cranial, caudal, left and right as standard. The CT-scans of the whale and control group was measured with dedicated software (ScoliosisAnalysis 4.1; Image Sciences Institute, Utrecht, The Netherlands, developed with MeVisLab, MeVis Medical Solutions AG, Bremen, Germany) to measure the direction and amount of rotation, anterior and posterior height of vertebral bodies and vertebral discs in the exact mid-sagittal plane, corrected for deformity in all three planes. This software is in-house developed and validated with excellent intra- and interobserver reliability.[45] This semi-automated method is used and extensively described in multiple earlier studies.[23,25,45,46] For all upper and lower endplates in the included part of the spine, the observer adjusted the plane of view for coronal and sagittal tilt. In this true transverse plane, the vertebral body and spinal canal were manually segmented by the observer, whereafter the software automatically determined the 3D coordinates of the anterior and posterior point of the endplate, adjusted for rotation and deformity in all planes. The distances between these points were calculated to obtain the anterior and posterior heights of the vertebral bodies and intervertebral discs (Fig. 6). This was done for all the compensatory curves (Cobb-to-Cobb). The corresponding levels of the spine analysed in the whale were also measured in controls. After measurements, the anterior-posterior length discrepancy (AP%) was calculated as [(anterior length – posterior length)/(posterior length)] × 100%, for the total compensatory curved spine, and for the vertebral bodies and the intervertebral discs separately. Endplates severely affected by trauma or wedging were excluded, as proper segmentation was not possible. Positive AP% values indicated that the anterior (ventral) side was longer than the posterior (dorsal) side.

Statistical analysis

The mean AP% results for the total curve, the vertebral bodies and the intervertebral bodies were determined for the minke whale and for the non-scoliotic harbour porpoise control group given with ± standard deviation. The differences in mean AP% between the scoliotic whale and non-scoliotic controls were tested with an independent samples T-test. Statistical analysis was performed in SPSS 25.0 for Windows (IBM, Armonk, NY, USA). The level of statistical significance was set at $p \leq 0.05$.

Results

Post mortem findings

The common minke whale was a 403 cm long, 530 kg female juvenile, with an estimated age between 0.5-4 years.[33] Besides the clear lateral post-traumatic curvature of the spine, other important findings of external examination were multifocal areas of deep haemorrhage and oedema that were present in the subcutis and longissimus dorsi muscle, as well as the presence of blood tinge liquid in the spinal canal and congestion and haemorrhage of the brain. The animal had a poor nutritional condition (blubber layers of 20–25 mm) despite recent feeding.[34] Histology of the fractured vertebrae demonstrated fibrin deposits, some eosinophilic granulocytes, and necrosis, indicative of chronic changes that were still ongoing. The most likely cause of death was considered to be acute recent blunt trauma. In addition, there was clear evidence of earlier trauma that had resulted in the fractures and other deformations of the lumbar vertebrae that had led to a post-traumatic deformity of the spine. Visual inspection showed that the deformity was mostly in the coronal plane with no significant lordosis or kyphosis at that region. This was further investigated after removing all of the soft tissues of the entire vertebral column. Visual inspection showed an epiphysiolysis at the left-side of the lower endplate of vertebra L3, a burst upper endplate at the right-side of vertebra L4, fractured/missing spinous processes of vertebrae L1 to L6, severely wedged vertebrae T11 and T12 and detachment of the transverse processes at multiple levels (Fig. 2). Therefore level L3/L4 was the suspected site of an acute (dorso-)lateral blunt traumatic injury, which subsequently initiated a double compensatory curve cranially and a single compensatory curve caudally (Fig. 3). In areas of the spine that did not show underlying anatomical changes, we analysed the compensatory curves in 3D and compared the morphology with the non-scoliotic spine of 10 control whales. The levels T11/12 (severe wedging) and L3/L4 (traumatic injury) were excluded before CT-scan analysis of the compensatory curvatures.

CT-measurements

The 3D analysis of the compensatory curves showed a rotation of the vertebral bodies in the transverse plane into the convexity of the curve (Fig. 4). The mean anterior-posterior length discrepancy (AP%) of the total compensatory curvature was + 9.4% in the whale. This means that the anterior (ventral) length of the compensatory scoliotic curvature was 9.4% greater than the posterior (dorsal) length, indicating a regional lordosis. This is significantly different from the kyphosis in the same part of the spine in the non-scoliotic control group, with a total AP% of -2.1 ± 0.4%, meaning that the anterior length of the spine was 2.1% shorter than the posterior length ($p < 0.001$). On the contrary, the bony morphology of the vertebral bodies was similar to the controls; the vertebral body AP% of the whale was − 2.5%, which was comparable to the kyphotic shape of the vertebral bodies in controls with − 1.8 ± 0.8% ($p = 0.429$). Almost all anterior lengthening took place in the intervertebral discs, as the disc AP% in the compensatory curvature of the whale was + 99.5%, which meant a lordotic shape of the intervertebral discs with an anterior length almost twice the posterior length. This is in sharp contrast to the kyphosis in the discs of controls with − 4.6 ± 5.0% ($p < 0.001$). The AP% for the separate vertebral bodies and intervertebral discs at every level is shown in Fig. 5.

Discussion

Idiopathic scoliosis is a 3D decompensation of a spine with no anatomical deformities, in an individual without underlying manifest disease. In the search for its aetiology, many theories involving just as many of the body's organ systems have been suggested to play a role.[1,4–12] The usually present lengthening of the anterior side of the thoracic spine in idiopathic scoliosis,[5,6,35] was suggested to be the result of a generalized bony overgrowth disorder (relative anterior spinal overgrowth; RASO), possibly as a compensation for a disturbance of synchronized growth between the neural and the osseous elements.[7–9, 36] Recent observations have shown that the initial deformity in scoliosis presents as anterior lengthening of only the intervertebral disc,[22–25] and changes in vertebral morphology are observed later in severe and progressive scoliosis.[26]

We propose that scoliosis is a very universal compensatory mechanism that can occur as a response to a disturbance of spinal balance. The crucial difference between the human spine and that of other mammals is not in its anatomy, but in the way it is biomechanically loaded, not by the fact that man is bipedal (many species are) but by the fact that humans carry their center of mass more posteriorly than any other species.[13–17, 27] This makes the human spine, in comparison with any other spine in nature, quadrupedal and bipedal alike, a rotationally less stable structure.[18,19] This means that, whereas in other species often draconic measures are necessary to induce a scoliosis,[3] in humans much less is needed to initiate this mechanism. We propose that the possible value of scoliosis research in experimental animals is not in the primary, artificially induced curve, but in the response that follows in the untouched area of the spine, i.e. the compensatory curve.

The objective of the current study was to investigate the mechanism through which a scoliosis develops in the normal area of the spine in an animal that is not known to develop a scoliosis spontaneously, by studying the 3D morphology of the compensatory curves in uninvolved areas of the spine around a traumatically induced coronal plane deformity in a whale. A (dorso-)lateral blunt traumatic injury of two vertebrae caused an acute, predominantly lateral deviation of the spine, that subsequently initiated 3D compensatory scoliotic curves that showed rotation of the vertebral bodies into the convexity of this compensatory curve, and an apical lordosis (+ 9.4%), which differed significantly from the kyphosis in the spine of the control group (-2.1%). The bony anatomy showed no difference with the normal, bony kyphosis in the control group (-1.8%). The lordosis was exclusively located in the intervertebral discs, they showed strong anterior lengthening (+ 99.5%) which was in stark contrast with the kyphotic discs of controls (-4.6%). This 3D morphology is identical to what is found in humans in idiopathic scoliosis, but also in human scoliosis with a known origin, i.e. neuromuscular scoliosis as well as in the compensatory curve that is initiated by a localized congenital malformation.[22–25]

The acute primary scoliosis, following a traumatic accident resulted in the head and tail of the whale being out of line and inhibiting proper locomotion and swimming manoeuvres. As many mammals have a vestibular reflex of self-righting,[37,38] the whale most likely compensated this trunk imbalance by curving other areas of the initially unaffected spine in an attempt to re align its head to its tail, in a mechanism that strongly resembles the 3D morphology of human scoliosis. Whereas most spines in nature require substantial effort to start a permanent rotational deformity due to the stabilizing action of gravity in combination with the trunk's muscles (i.e. the follower load),[39,40] the human spine is much less rotationally stable due to its unique sagittal profile with the body's centre of gravity straight above, rather than in front of the pelvis.[13–17, 27] This reduces the stabilizing anterior shear loading and even induces *posteriorly* directed shear loads that were shown to render the involved spinal segments unstable in the horizontal plane.[18,19]

This rare occurrence of scoliosis in a species that is not known to develop a spinal curvature spontaneously, provided a unique chance to study scoliosis in a completely different model. A limitation of this study was that the common minke whale was not compared to non-scoliotic controls of the exact same species. This is due to the low frequency of stranded common minke whales in the Netherlands, in combination with their large size and weight exceeding the capacity of most CT-scanning facilities. However, the smaller harbour porpoise (*Phocoena phocoena*) share strong commonalities in spinal anatomy and were therefore used as controls in the current study.[41,42] Furthermore, during CT-scan analysis in this study, not all vertebrae of the whale were included. This was mainly due to the extensive morphological changes which, due to trauma and subsequent wedging, impeded the proper study of the spinal anatomy in the traumatized region. However, overview images in the sagittal plane of the CT-scan did not show a significant kyphosis nor lordosis at the site of traumatic injury. Therefore a significant post-traumatic kyphosis, resulting in a compensatory lordotic response could thus be excluded.

The aim of this study was to study whether scoliosis is a more generalized compensatory mechanism that occurs independent of cause and/or species. The results confirm our hypothesis; the compensatory curves in the essentially normal spine of the whale show very strong similarities in 3D configuration with different types of human scoliosis. This suggests that mechanistically, scoliosis is a very universal response to any disturbance of equilibrium. In humans, this mechanism can become operational much easier than in any other species because of an intrinsic lack of rotational stability of the human spine.[18,19]

Declarations

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Author contributions

The necropsy was performed by LLIJ, MJK and AG. Diagnostic imaging was performed by DSW and SV. The software for three-dimensional analysis was developed by MS. CT-scan measurements were performed by SR and analysis of these measurements were performed by SR, JFH, MCK and RMC. SR and LLIJ wrote the main manuscript text and all authors reviewed and edited the manuscript.

Additional information

Animal ethics committee approval: The animals described in the current study were not used for scientific or commercial testing. All were free-living whales which died of natural causes or were euthanized on welfare grounds and not for the purpose of this, or other studies. Therefore, since there was no handling of live animals in the current study, according to institutional guidelines, no consent from the Animal Use Committee was required, and animal ethics committee approval was not applicable to this work.
Competing interests: The authors declare to have no competing interests.
Data availability statement: All generated or analysed data in this study are included in this article or the supplementary information files.

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Figures

Figure 1

Photograph of the common minke whale (Balaenoptera acutorostrata) that washed ashore on the 8th of July 2019 at Texel, the Netherlands. Photograph by Pierre Bonnet (Ecomare, Texel).

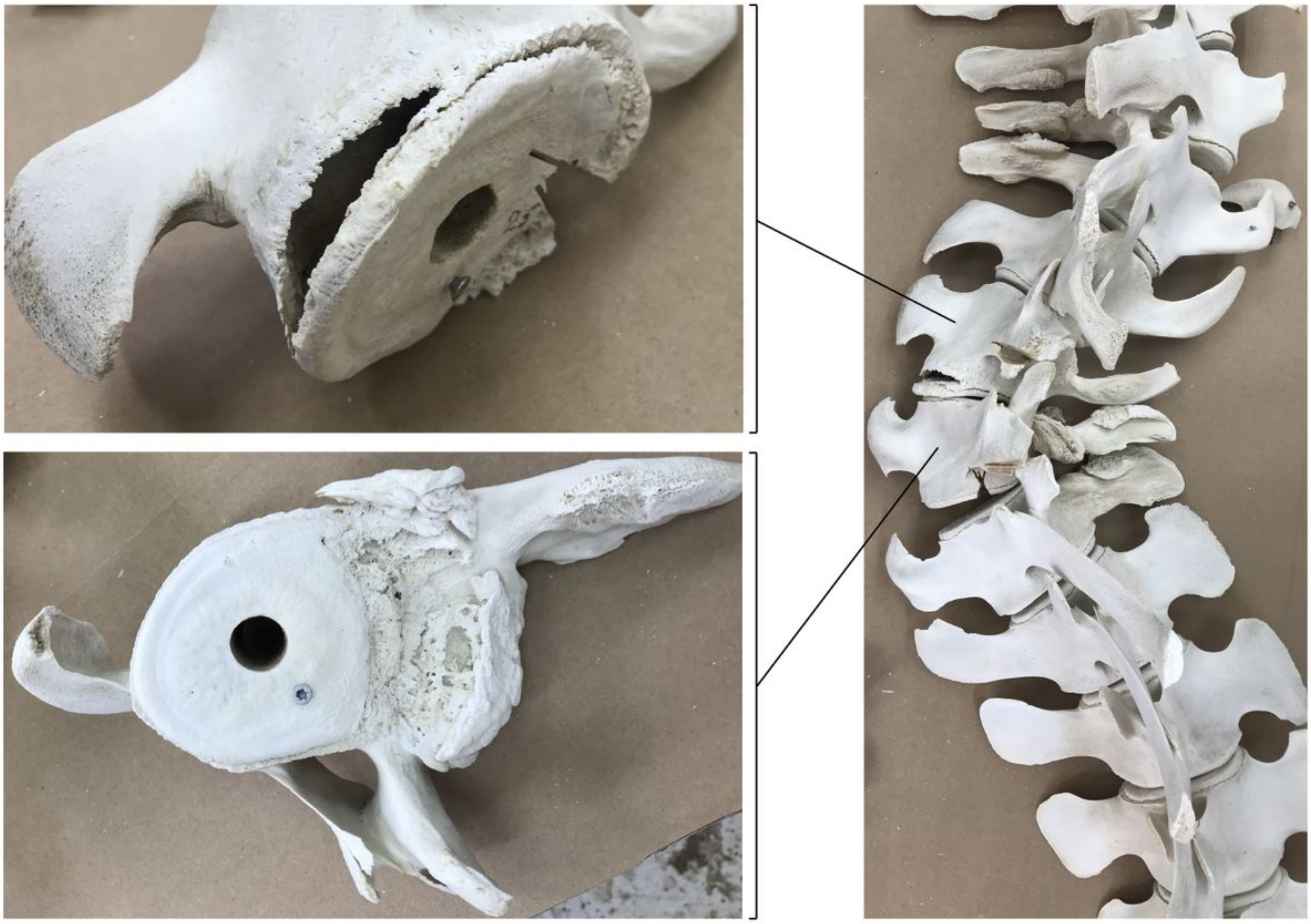

Figure 2

Photographs of the vertebrae after removing the soft tissues. The dorsal overview on the right-hand side shows the post-traumatic primary, abrupt coronal curve at level L3/L4. Close-up inspection reveals an epiphysiolysis at the left-side of the lower endplate of vertebra L3, and a burst upper endplate at the right-side of vertebra L4. Furthermore, fractured spinous processes of vertebrae L1 to L6, severely wedged vertebrae T11 and T12 and detachment of the transverse processes at multiple levels are present. There are multiple post mortem marks following tissue selection for histopathology, and also centre holes and screws that were drilled through the endplates in the process of framing the complete skeleton for museum display. These artefacts did not influence the presented post-traumatic features.

Figure 3

Dorsal view with the cranial side upwards from the CT-scan of level C1 to L7. The suspected site of the (dorso-)lateral blunt traumatic injury at level L3/L4 (indicated with an asterisk) initiated a double compensatory curve cranially and a single compensatory curve caudally.

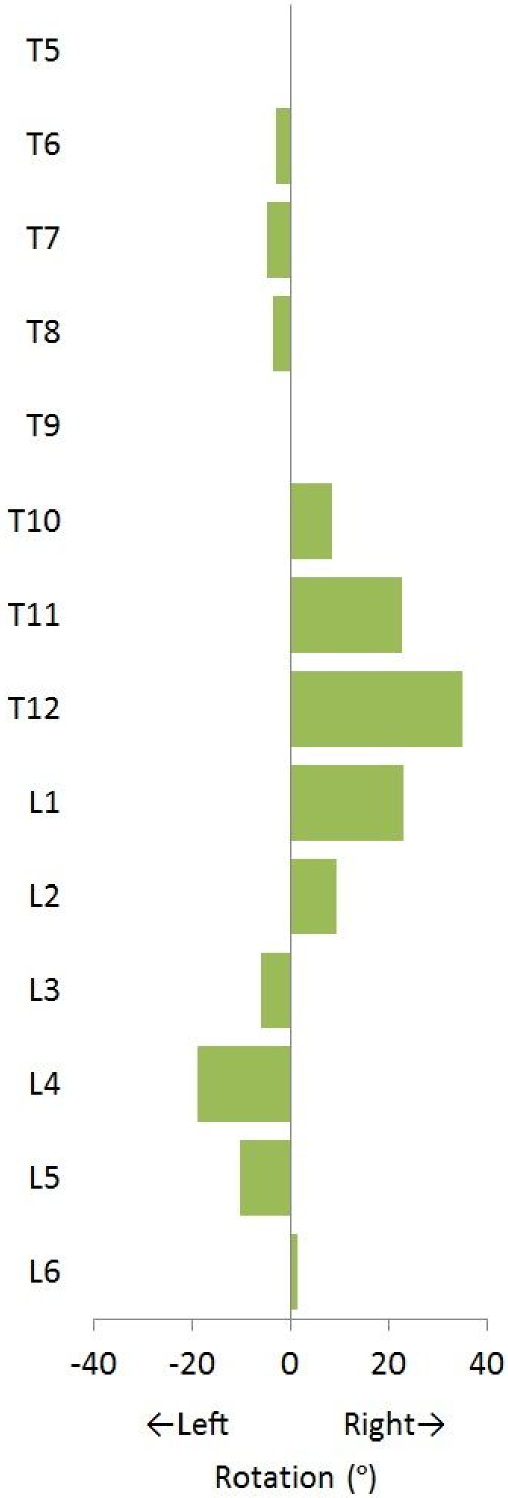

Figure 4

Of the whale with scoliosis, the rotation of the vertebral bodies in the transverse plane is shown in degrees. Positive values indicate that the anterior part of the vertebral body is pointing towards the right. All rotation of the vertebral bodies is into the convexity of the curve.

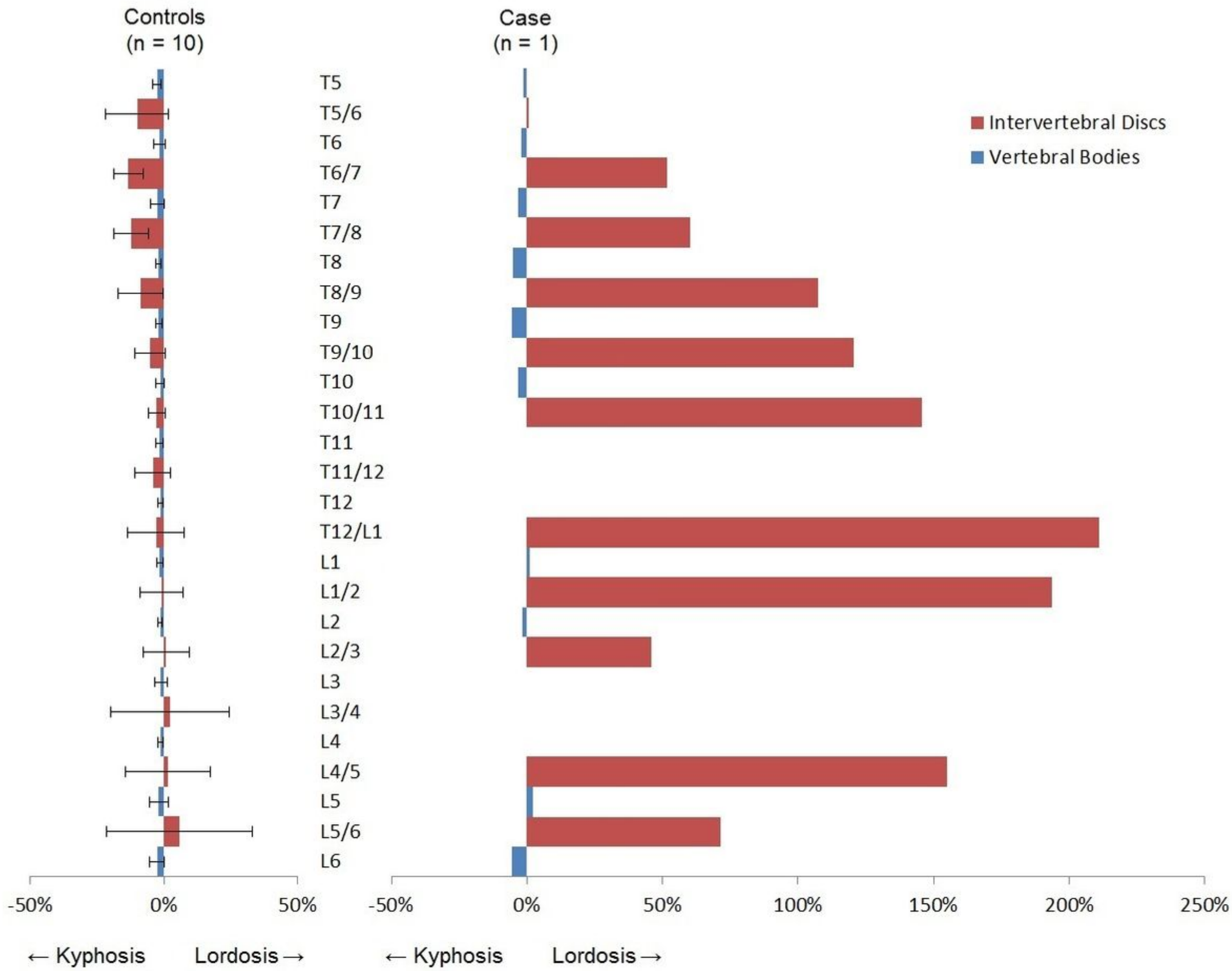

Figure 5

The mean anterior-posterior length discrepancy (AP%) with the standard deviation is shown for the intervertebral discs in red and the vertebral bodies in blue, for both the non-scoliotic controls and the whale with scoliosis. Endplates severely affected by trauma (L3/L4) and wedging (T11/T12) were excluded due to impossibility of proper CT-scan analysis. Positive AP% indicates a larger anterior length than posterior length.

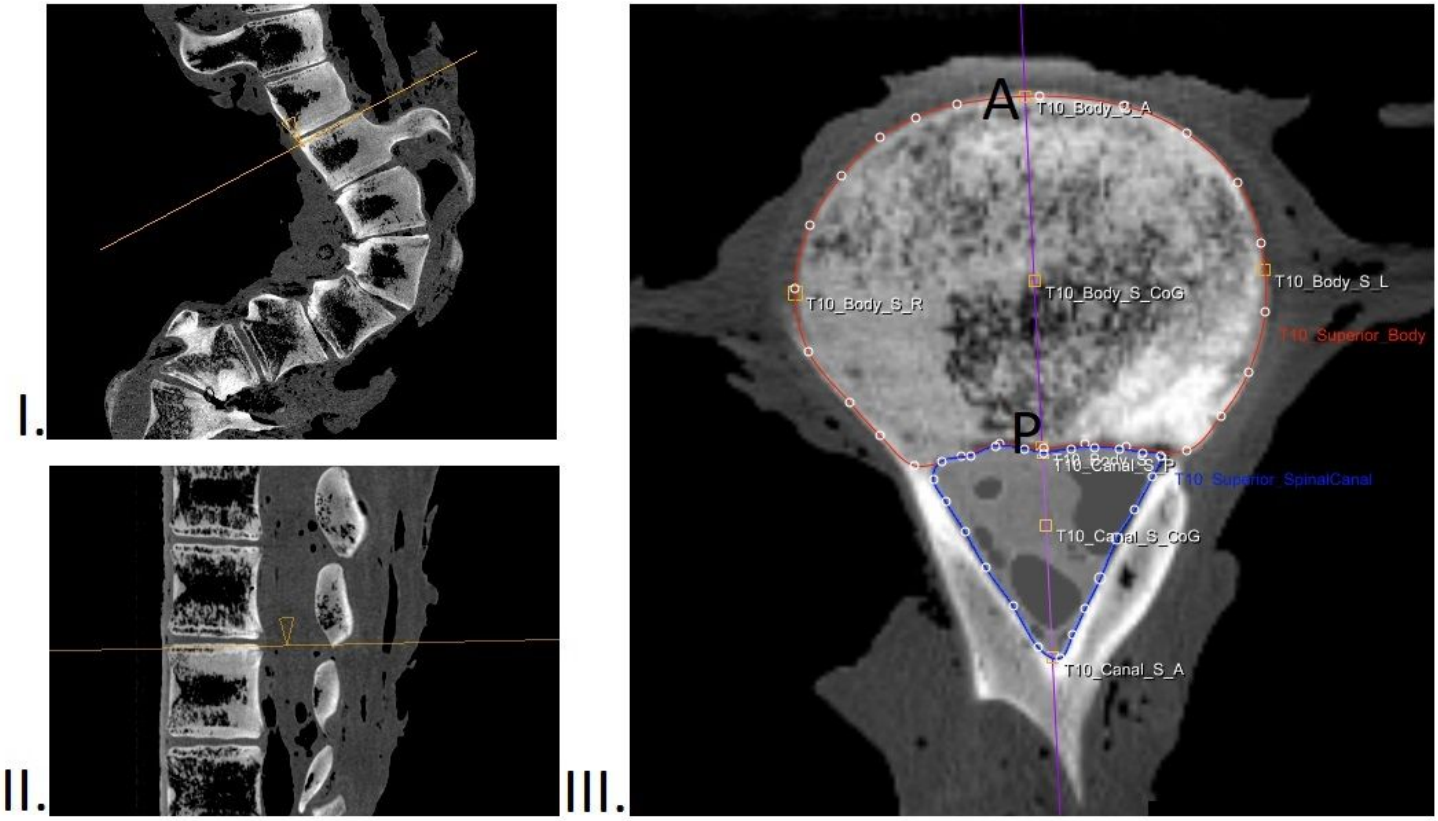

Figure 6

Method of 3D measurements on CT-scans in this study. For all upper and lower endplates, the observer adjusted the plane of view for coronal (I) and sagittal (II) tilt. In the true transverse plane, the vertebral body and spinal canal were manually segmented (III), whereafter the software automatically determined the 3D coordinates of the anterior (A) and posterior (P) point of the endplate, adjusted for rotation and deformity in all planes. The distances between these points were calculated to obtain the anterior and posterior heights of the vertebral bodies and intervertebral discs.

Supplementary Files

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- Supplementaryinformationfiles.xlsx