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Dysbaric osteonecrosis (DON) among the artisanal diving fishermen of Yucatán, Mexico

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Abstract
Popa D, Medak A, Chin W, Huchim-Lara O, Fliszar E, Hughes T, Grover I. Dysbaric osteonecrosis (DON) among the artisanal diving fishermen of Yucatán, Mexico. Diving and Hyperbaric Medicine. 2020 December 20;50(4):391–398. doi: 10.28920/dhm50.4.391-398. PMID: 33325021.)
Introduction: Artisanal diving fishermen in Yucatán, Mexico have high rates of decompression sickness as a result of frequently unsafe diving practices with surface supplied compressed air. In this study, we investigated the prevalence of dysbaric osteonecrosis (DON), a type of avascular necrosis, in the most susceptible joints in a cohort of these fishermen.
Methods: We performed radiographs of bilateral shoulders, hips, and knees of 39 fishermen in Mexico and surveyed them about their medical and diving histories. We performed pairwise correlations to examine if the fishermen's diving behaviours affected the numbers of joints with DON.
Results: The radiographs revealed Grade II or higher DON in 30/39 (76.9%) of the fishermen. Twenty-two of 39 fishermen (56.4%) had at least two affected joints. The number of joints with DON positively correlates with the lifetime maximum diving depth and average bottom time.
Conclusions: These findings represent among the highest prevalence rates of DON in divers and reflect the wide-spread scale of decompression sickness among these fishermen. Through this work, we hope to further educate the fishermen on the sequelae of their diving with the aim of improving their diving safety.

Introduction

Decompression sickness (DCS) among compressed gas divers occurs when dissolved gas absorbed into tissues while at depth (in a hyperbaric environment) comes out of solution and forms bubbles following ascent. While DCS can involve many tissues, bony tissue is frequently affected and can lead to dysbaric osteonecrosis (DON). DON is atraumatic bony infarction, often symptomatic when involving the articular surfaces of long bones, and first described in 1941.[1] However, it was known to affect bridge caisson and tunnel workers, both exposed to hyperbaric environments, in the early 1900s. Severe cases of DON can lead to debilitating bony collapse and impaired function limiting not only employment but also activities of daily living. Osteonecrosis occurs in the bones adjacent to the joints including the head, neck, and shafts of long bones, which from hereon are simply referred to as the joints.

Artisanal diving fishermen of Yucatán state in Mexico typically use surface supplied compressed air, via a 'hookah' system, aboard small open cockpit boats to fish for a variety of marine species including grouper, lobster, and sea cucumber. Hookah systems involve the use of a shipboard air compressor supplying the air to the diver below via a long hose (Figure 1). This technology allows the diving fisherman a virtually unlimited gas supply which permits unsafe diving practices, far exceeding normal recommended safety limits for time while at depth. As a result, an epidemic of DCS plagues this population.[2–4]

Despite frequent DCS, the diving fishermen continue to exceed established safety limits likely due to economic pressures or the drive to earn additional income. In this study, we sought to characterise the prevalence of DON among artisanal diving fishermen with the ultimate goal of informing these divers of the sequelae of DCS and educating them in strategies to reduce their risk of DCS while maintaining their livelihoods as fishermen.

Figure 1

A fisherman from Río Lagartos preparing bait in his fishing boat. A typical Yucatán hookah compressor lies in the bow of the boat with its engine covered by a plastic tarp

Figure 2

A map of the Yucatán Peninsula showing the major fishing communities of the northeast coast. The nearest X-ray facility and hyperbaric chamber to treat DCS are located in the larger community of Tizimín

Methods

The protocol was approved by the institutional review board at the Universidad Marista de Mérida in Yucatán (project # CE_009_2017). Informed consent was obtained from all individual participants included in the study.

Thirty-nine artisanal diving fishermen from Yucatán, Mexico agreed to participate. All subjects work and dive within the fishing communities of Río Lagartos and San Felipe in Yucatán, Mexico (Figure 2). We utilised a non-probabilistic convenience technique to invite subjects to participate in the study and recruited fishermen among the fishing cooperatives of the respective villages. Cooperative officers assisted in recruitment efforts by calling some of the fishermen. Inclusion criteria included membership in a fishing cooperative and a history of fishing the local waters using surface supplied compressed air. Exclusion criteria were age less than 18 years old and no prior diving. Fishermen may become involved in diving fishing as young as 15 years in a supportive role but do not start diving until reaching 18 years.[2] After obtaining consent, we surveyed the fishermen regarding medical and social histories, diving behaviour and experience, and episodes of DCS.

Table 1

The Steinberg modification of the Ficat scoring system for AVN used for DON. Originally, the classification system was developed to grade AVN of the hip but can be used to grade other joints. MRI = magnetic resonance imaging

Grade	Findings
0	Normal X-ray, MRI, or bone scan. Diagnosed histologically
I	Normal X-ray, abnormal MRI or bone scan (but minimal joint pain)
II	Sclerosis and/or cyst formation in the femoral head (or equivalent)
III	Subchondral collapse (crescent sign) without flattening
IV	Flattening of the femoral head (or equivalent) without joint narrowing or acetabular involvement
V	Flattening of the femoral head (or equivalent) with joint narrowing and/or acetabular involvement
VI	Advanced degenerative changes

Nine radiographs were performed per subject: anterior-posterior (AP) and abducted AP views of each shoulder (four total), AP and frog-leg views of each hip (four total), and one AP view of bilateral knees over the course of one week. The radiology technician used a Compagnie Générale de Radiologie (CGR, 1976) analog X-ray machine to obtain the radiographs.

The X-ray films were transported to our facility where two musculoskeletal fellowship-trained, board-certified radiologists independently interpreted the radiographs using the Steinberg modification of the Arlet and Ficat avascular necrosis (AVN) grading system (Table 1).[5–7] The radiologists were blinded to the subjects' demographic information as well as their medical and diving history. The radiologists subsequently resolved conflicting interpretations of the radiographs through consensus.

Furthermore, self-reported diving behaviours were correlated with the number of joints affected by DON. A Shapiro-Wilk test was used to assess normality and distribution of the data set. A parametric and non-parametric Levene's test was used to assess equality of variance. Univariate tabulation across all variables was conducted. A pairwise correlation matrix was conducted across outcome variables: number of affected joints; number of DCS events; lifetime maximum depth;

Table 2
Demographic information and diving behaviour of the artisanal diving fishermen in the study cohort. Of note, the fishermen do not use dive computers or depth gauges. Rather, they estimate depth based on the number of arm's lengths or strokes needed to swim to the bottom. One arm's length approximates to 1.5 msw. BMI = body mass index; DCS = decompression sickness; Max = maximum; msw = metres' seawater; SD = standard deviation

Parameter	Mean (SD)	Range	Median
Age (years)	44.7 (8.5)	26–62	45
BMI (kg·m^{-2})	32.7 (4.7)	23.6–43.2	32.8
Years diving	26.8 (8.5)	10–47	27
Prior DCS episodes	9.0 (7.7)	0–35	8
Dive depth (Arm lengths)	7.8 (5.3)	1–20	8
Dive depth (msw)	11.8 (7.9)	1.5–30	12
Max. depth (Arm lengths)	17.6 (5.6)	7–36	17
Max. depth (msw)	26.4 (8.4)	10.5–54	25.5
Bottom time (minutes)	107.2 (72)	20–360	90

Table 3
Past medical history of the artisanal diving fishermen in the study cohort (n = 39)

Medical history	*n* (%)
Regular alcohol	29 (74.4)
Joint pain over last year	27 (69.2)
Hypercholesterolaemia	17 (43.6)
Skeletal / joint trauma	9 (23.1)
Hypertension	5 (12.8)
Diabetes mellitus	3 (7.7)
Arthritis	2 (5.1)
Lung disease	2 (5.1)

average diving depth; average bottom time; and fishing years. A Chi-square test of independence was conducted among outcome variables, number of affected joints, and number of DCS events and amount of treatment. A user-written algorithm was utilised to create tables and plots. We utilised STATA version 16 for data analysis.

Results

Thirty-nine diving fishermen from the two fishing villages of Río Lagartos and San Felipe completed the study, each undergoing nine radiographs as well as the survey of

Figure 3
Representative radiographs showing Grade II DON in the joints studied. Areas of sclerosis are indicated with red arrows. Top row-AP and abducted AP views of the right shoulder; Middle row-AP and frog leg views of the left hip; Bottom row-AP view of bilateral knees

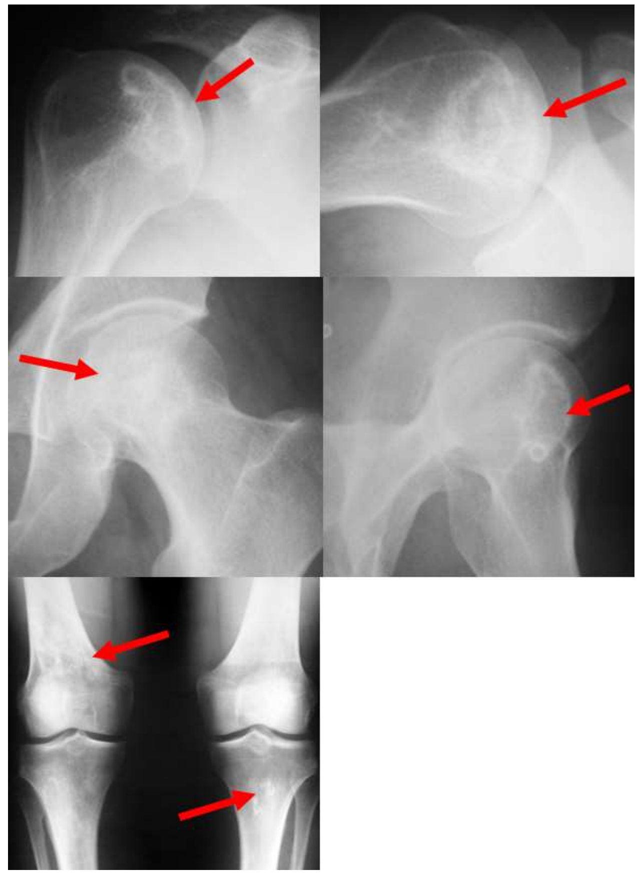
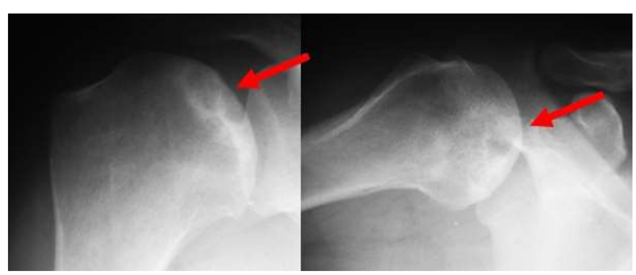


Figure 4
A 51 year-old male fisherman displayed Grade IV DON in both AP and abducted AP views of the right shoulder. Areas of collapse indicated with red arrows

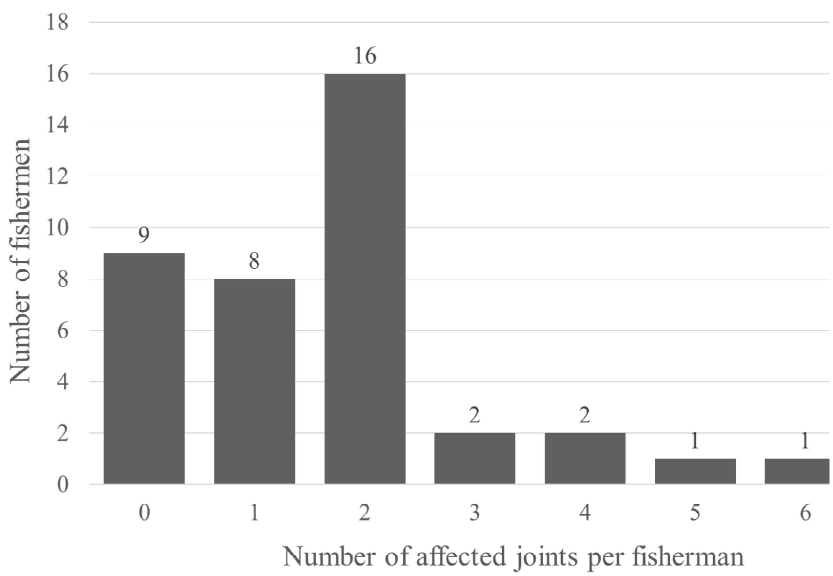


demographic data, diving behaviour, and health data (Tables 2 and 3). All but one of the fishermen (97.4%) reported a history of DCS. Thirty of the 39 fishermen (76.9%) displayed Grade II or higher DON. Of those 30, all subjects had Grade II severity with one exception. Representative images of the radiographic findings of the fishermen can be seen in Figure 3. One fisherman displayed Grade IV DON of the right shoulder seen on both AP and abducted AP

Table 4
Prevalence of DON among the artisanal diving fishermen according to joint type and laterality. As an example, right shoulder entries denote fishermen with DON in the right shoulder but not the left and may have other joints affected as well

Joint(s)	Number of fishermen	% of DON (n = 30)	% of all fishermen (n = 39)
Right shoulder	2	6.7	5.1
Left shoulder	3	10	7.7
Bilateral shoulders	22	73.3	56.4
Right hip	4	13.3	10.3
Left hip	0	0	0
Bilateral hips	3	10	7.7
Right knee	1	3.3	2.6
Left knee	1	3.3	2.6
Bilateral knees	2	6.7	5.1

Figure 5
Histogram of the number of joints with AVN per diving fisherman



radiographs and no other joints affected by DON (Figure 4). The percent agreement between the two radiologists was 94% agreement (220/234 joints). One of the debated joints was a disagreement in the grade of DON, while 6 of the debated joints had one radiologist diagnosing possible or probable DON rather than definite DON. So without these, there was only 3% (7/234 joints) disagreement between the radiologists with all joint findings ultimately achieving a consensus diagnosis.

Within this cohort, DON affected the shoulders to a much greater degree than the hips or knees (Table 4). Both shoulders were typically involved. Of the subjects with DON, 27 of 30 (90%) displayed shoulder involvement with 22 (73.3%) having bilateral involvement. The hips and knees also reflected this trend where bilateral DON prevailed. One subject demonstrated Grade II DON of all six joints and 22 of the 39 fishermen (56.4%) in the entire cohort had two or more joints with DON (Figure 5).

In addition, we attempted to correlate the self-reported diving behaviour of the fishermen with the number of affected joints (Table 5). The number of joints with DON positively correlated with the lifetime maximum depth (correlation coefficient = 0.53) and average bottom time (correlation coefficient = 0.36) but not the number of DCS episodes requiring hyperbaric oxygen treatment (HBOT), average diving depth, or years fishing. Furthermore, the number of

Table 5

Pairwise correlation matrix table of surveyed diving behaviour variables with the number of DON affected joints. Each cell displays the correlation coefficient (r) (top), *P*-value (middle), and number of fishermen included (bottom). * indicates significance at the *P* < 0.05 level. # DON joints = number of joints affected by DON. # DCS = episodes of DCS that required HBOT. Max. depth = lifetime maximum diving depth. Avg. depth = typical depths fished. Bottom time = typical bottom times during fishing. Fishing years = years spent doing surface supplied diving in order to fish

Variable	# DON joints	# DCS	Max. depth	Avg. depth	Bottom time
# DCS	r = 0.280 *P* = 0.08 *n* = 39				
Max. depth	r = 0.527* *P* = 0.006 *n* = 26	r = 0.180 *P* = 0.379 *n* = 26			
Avg. depth	r = -0.054 *P* = 0.747 *n* = 38	r = -0.235 *P* = 0.156 *n* = 38	r = 0.156 *P* = 0.458 *n* = 25		
Bottom time	r = 0.364* *P* = 0.027 *n* = 37	r = 0.270 *P* = 0.106 *n* = 37	r = 0.351 *P* = 0.079 *n* = 26	r = -0.410* *P* = 0.013 *n* = 36	
Fishing years	r = 0.087 *P* = 0.601 *n* = 39	r = 0.320* *P* = 0.047 *n* = 39	r = 0.016 *P* = 0.938 *n* = 26	r = 0.218 *P* = 0.189 *n* = 38	r = 0.070 *P* = 0.681 *n* = 37

years fishing positively correlated with the number of DCS episodes (correlation coefficient = 0.32), while average bottom time negatively correlated with average diving depth (correlation coefficient = -0.41).

Discussion

This cohort of fishermen displays a high prevalence (76.9%) of DON compared to most groups of diving fishermen described in the literature. Indeed, 56.4% of them had multiple joints affected by DON. For a population of diving fishermen, the long-term implications of such radiographic findings on their ability to work in physically demanding jobs and their ability to perform simple activities of daily living is quite concerning.

Previously, DON has been described among caisson (bridge support) workers as well as working divers (fishermen, military divers, etc.). Rates of DON among these populations vary widely, likely a function of the variable risk associated with each diving community's dive profiles, as well as other risk factors. Previous work has established that groups of artisanal fishermen with high rates of DON have dive profiles that allow for significant inert gas loading.

A study of Hawaiian coral divers demonstrated DON in 13 of 20 (65%) subjects.[8] In Honduran lobster divers studied in 1994–1995, 69% had evidence of DON.[9] Interestingly, this cohort displayed a higher percentage of lesions in the distal femur and proximal tibia than in the humeral head. This is contrary to the trend most often seen in divers where the humeral head is predominantly affected. Several groups of Japanese shellfish divers have been studied and the rate of DON ranged from 28% to 100%.[10–13] The study that detected DON in 100% of its divers used MRI imaging to make the diagnosis. Korean shellfish divers had a 67% prevalence of DON while Turkish sponge divers had a rate of 70–85% DON.[14,15] In contrast to these artisanal diving fishermen, the rates of DON in recreational SCUBA divers is considerably lower.[16,17] Also, commercial divers not engaged in fishing have been shown to have a low prevalence of DON (4.2% in British North Sea commercial divers) although a more recent study among Turkish diving instructors showed a 25% prevalence of DON.[18,19] A comprehensive review of DON among professional divers describes rates of DON among these different populations.[7]

The mechanism by which DON develops remains unclear. DON occurs in bones with yellow (fatty) marrow and not in bones with red (hematopoietic) marrow. Nitrogen absorption and release occurs slowly in fatty marrow as a result of relatively low blood flow. Current evidence shows that DON can develop after a significant hyperbaric exposure causes gas loading of the fatty tissue. Subsequently, inadequate decompression can lead to bubble formation that could disrupt the tissue. These bubbles also could cause an ischaemia-reperfusion injury, platelet aggregation, fibrinogen deposition, disseminated intravascular coagulation (DIC), lipid emboli and the release of vasoactive substances.[20] The ultimate result of this cascade of events caused by the bubbles could be the ischaemic bone infarction of DON. Analyses have shown a very high correlation between divers that have suffered DCS (the 'bends') and the development of DON. A prevalence of 4/21 (19%) DON was reported among French recreational divers who presented to hyperbaric facilities with musculoskeletal DCS and underwent MRIs immediately after their injury and 3–4 months later.[17] According to others, "*more than 60% of divers with DON have experienced one or more symptomatic episodes of limb bends in spite of a certain degree of under-reporting and self-treatment of mild DCS symptoms*."[20]

The patient with early DON is usually asymptomatic and has no physical findings (Grade 0-II). Once bony collapse occurs (Grade III and IV), the patient typically becomes symptomatic. Symptoms include joint pain, reduced range of motion, and muscle spasms. Late findings include joint deformity, crepitus, and contractures. Rarely, DON in its early stages is reversible. Once DON has developed, it is usually progressive and carries a poor prognosis. Most patients will eventually develop Grade IV DON equivalent to secondary degenerative arthritis.

Examining the correlation between fishing behaviours and the number of joints affected, we found that the diving fishermen who reported deeper lifetime maximum depths and longer average bottom times tended to have more joints affected, although this correlation was not terribly strong and was based on self-reported data. Among this population, the deeper diving fishermen are likely subjected to more bubble stress which in turn leads to more DON. Similarly, longer bottom times also likely lead to more bubble stress and in turn more DON in the same way. However, deeper lifetime maximum depths or longer average bottom times may simply be markers for other causative factors such as riskier diving, increased overall workload (such as more effort obtaining the catch, increased lifting power required to offload the catch, or both), or some other unknown factors.

We found no correlation between episodes of DCS or years fishing with the number of joints affected. The fishermen identify episodes of DCS as times they have required treatment in the nearest hyperbaric chamber in Tizimín, not necessarily episodes of joint pain. Retrospectively quantifying pain-only DCS episodes is essentially impossible among this working population due to the frequency of joint pain, self-treating with medication (predominantly non-steroidal anti-inflammatory medication such as diclofenac) and alcohol, and confounders such as muscle strain or soreness from heavy fishing activity. Hence, the number of DCS episodes the fishermen reported is surely an underestimate and fails to capture episodes of Type I (i.e., pain only) DCS. Previous work by members of our group has shown that among a group of 105 diving fishermen, 97 (92.4%) had at least one episode of DCS that required HBOT over a 25-year span during which medical records were available.[21] Unsurprisingly, more years fishing correlated with more episodes of DCS requiring HBOT, while deeper average depths correlated with shorter average bottom times. Of note, the fishermen undergo HBOT when they cannot tolerate or manage their joint pain with self-treatment or have significant symptoms they or the community recognise as DCS. The fishermen do not generally use in-water recompression techniques in part due to the shallow depths near shore because of the gentle sloping Yucatán Shelf. HBOT is provided at the nearest chamber in Tizimín and most often consists of U S Navy Treatment Table 5 as the first line treatment with U S Navy Treatment Table 9 used for subsequent treatments if needed.[2]

Currently, no specific treatment for DON exists. Once a patient has progressed to Grade IV DON and symptoms are no longer responding to conservative measures, then total joint replacement is often the treatment of choice. Early treatment for decompression sickness with HBOT has been shown to reduce the risk of DON in sheep.[22] At the present time, no formal HBOT protocol for DON has been developed in the United States. Italian hyperbaric physicians have created a protocol, however, with promising early data for the treatment of femoral head necrosis.[23] This protocol consists of daily treatments, 5–6 days per week, with at least 60 minutes of 100% O_2 between 223–253 kPa, for a total of 60–90 treatments. Nevertheless, this protocol has not been examined in treating DON or other affected joints beyond the hip. Given the lack of clearly effective therapies for DON, the high prevalence of DON among the artisanal diving fishermen in Yucatán will likely lead to significant morbidity in the future, especially if diving behaviour and safety practices do not improve.

With our current understanding of the pathophysiology of DON, including ischaemia-reperfusion injury and the release of vasoactive substances, HBOT could plausibly prove an effective treatment modality for these diving injuries. A prospective human study is warranted to test this hypothesis, however.

LIMITATIONS

This study's primary limitations centre around its observational design without age-matched controls. AVN, regardless of etiology, is a known, relatively uncommon disease with 20,000–30,000 new cases per year in the United States (approximately 0.01% among the total population).[24] Alcohol use, a risk factor for AVN, was high among the fishermen (Table 3). However a study examining 1,157 patients undergoing treatment for excessive alcohol consumption showed 5.3% of their cohort had AVN, higher than the regular population but far lower than the prevalence we found among the fishermen.[25] Compared to the present cohort with such a well-known risk factor as DCS and the staggering amount of DCS in this population, it is considered likely that the radiographic findings within this cohort are secondary to DON and not some other AVN aetiology.

The subject recruitment methodology may have also added some selection bias as it was a convenience sample of volunteers. However, previous work has shown a rate of DCS requiring HBO_2 therapy of over 92% in these fishermen, so the current rate of 38/39 divers (97.4%) with a history DCS is sufficiently similar to suggest that selection bias was unlikely to play a significant role in the high rates of

DON described here.[21] In addition, the analog radiographs were not of the highest quality in comparison to the latest technology; however, they represent the most feasible option available in that region of Mexico. Furthermore, the subjects were surveyed about their diving behaviour and history of DCS which is subject to recall bias in addition to the fact that true depth gauges are not used during their diving. Rather, they estimate depth based on the number of arm's lengths or strokes needed to swim to the bottom. Not all fishermen answered all the survey questions. Our analysis sought to correlate the surveyed diving behaviour results with the radiographic findings but should not be taken to imply causation.

Conclusions

The artisanal diving fishermen in Yucatán, Mexico exhibit some of the highest rates of DON described in the literature. Likely, their extremely high rates of decompression sickness have contributed to these findings. In the future we hope to repeat this study with a larger cohort, incorporating MRI exams, and ultimately providing these radiographic findings to the fishermen. Through this feedback, we hope that they will modify their diving behaviour and institute a diving 'safety stop', as is common among recreational divers. This routine practice allows the diver to more safely unload nitrogen from tissues and reduce the likelihood of bubble formation, in turn reducing the risk of DCS, and therefore of DON.

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Conflicts of interest and funding

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