Aquatic Mammals 2009, *35*(1), 32-35, DOI 10.1578/AM.35.1.2009.32

A Case Study of Congenital Diaphragmatic Hernia in a Juvenile Striped Dolphin (*Stenella coeruleoalba*)

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Abstract

Congenital Diaphragmatic Hernia (CDH) was observed in a stranded juvenile male striped dolphin. The 2- to 3-y-old animal had survived with its stomachs and intestines in the thoracic cavity, which had caused a large size difference between its two lungs. The animal also had a relatively small penis. The animal's combination of anomalies was either due to a genetic syndrome or caused by maternal exposure to toxic agents.

Key Words: striped dolphin, *Stenella coeruleoalba*, congenital, CDH, *hernia diaphragmatica*, PCB

Introduction

Many odontocetes strand annually on the Dutch coast, most of which are dead. However, some animals strand alive and, if found in time, are transported to a veterinary treatment center (Kastelein et al., 1997). On 5 November 1999, a male striped dolphin (*Stenella coeruleoalba*) stranded alive on the Dutch coast. The animal died soon after arrival at the treatment center. Necropsy revealed congenital anatomical abnormalities such as a diaphragmatic hernia, which may have been caused by toxins in the environment. If other researchers find similar anatomical abnormalities in striped dolphins, efforts to study the environmental factors might need to be intensified.

Materials and Methods

The study animal had a standard length of 159 cm, a girth at the eyes of 72 cm, a girth in front of the pectoral fins of 85 cm, an axillary girth of 90 cm, a girth in front of the dorsal fin of 88 cm, a girth at the anus of 47 cm, and weighed 50.2 kg. No external abnormalities were observed. Based on body weight, body length, and growth layer groups of the teeth, the animal was estimated to be a juvenile of 2 to 3 y old (Di-Méglio et al., 1996; Willy Dabin, pers. comm.).

The dolphin was taken to a marine park in the Netherlands for veterinary treatment and rehabilitation. During transport, he remained very stressed (indicated by his high skin temperature and high respiration rate) despite intramuscular administration of Diazepam (Valium). Blood analysis showed no signs of dehydration (Hematocrit: 0.54 L/L, Hemoglobin: 11.5 mmol/L, Erythrocytes: 5.1 × 1,012/L) or infection (Leucocytes: 7.3 × 109/L). After arriving at the treatment center and being placed in a pool, the animal displayed many muscular cramps followed by strong, labored respirations.

The animal died 6 h after arrival at the treatment center. The carcass was frozen immediately after death but was later thawed so that an autopsy could be performed.

Results

Both the stomachs and the intestines were found to be in the thoracic cavity (Figures 1 & 2). The intestines were 22 m long and unusually varied in color over their length. The forestomach and fundic stomach showed no gross abnormality. The dislocation of the abdominal organs into the thoracic cavity organs was judged to be congenital because the left lung was less than half the volume of the right lung (left: 700 ml vs right: 1,600 ml; measured by water displacement in a measuring beaker while both lungs were deflated) (Figure 3). The lungs showed signs of pneumonia. Also, the dorsal opening in the left side of the diaphragm was due to the absence of a considerable part of the diaphragm (Figure 4). The left side of the diaphragm was approximately a third of the surface area of the right side, and the dorsal edge of the left side of the diaphragm was smooth, rounded, and thick (Figure 5), not a jagged edge which would have indicated a recent rupture.

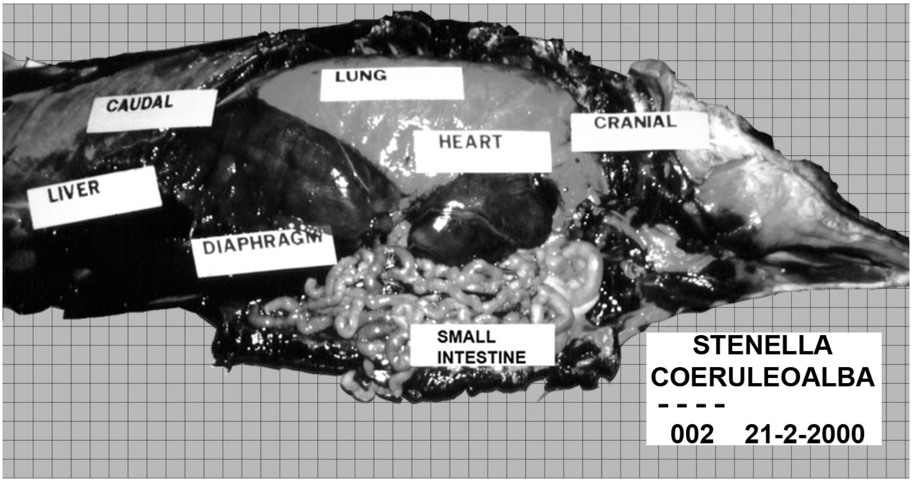

Figure 1. The right side of the striped dolphin (right rib cage removed) showing the intestines in the thoracic cavity; background grid: 2 × 2 cm.

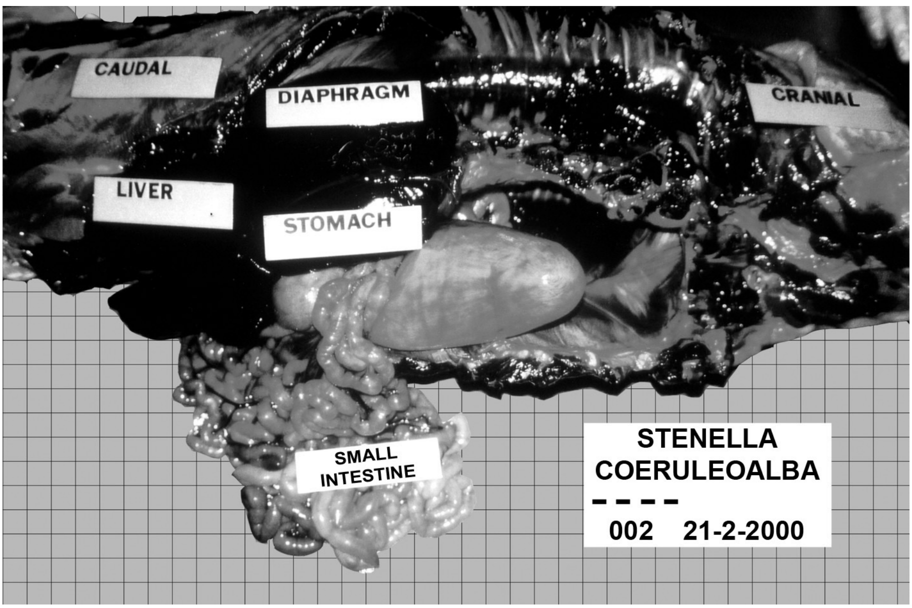

Figure 2. The right side of the striped dolphin, showing the forestomach in the thoracic cavity after the intestines were removed; background grid: 2 × 2 cm.

The animal also had hypoplasia of the penis (micropenis; the penis was less then 10% the size of the penis of a normal animal of this age and size). The heart (435 g) and kidneys (left: 250 g; right: 240 g) showed no gross abnormalities. The blubber thickness mid-dorsally was 13 mm, mid-laterally, 12 mm, and mid-ventrally, 11 mm; this is within the range found in healthy striped dolphins.

Based on the normal blood parameters, the muscular cramps and labored respirations observed during the last hours of the animal's life, the varied color of the intestines, and the limited space for the stomach and intestines, the most likely cause of death was determined to be Congenital Diaphragmatic Hernia (CDH, *hernia diaphragmatica*), the protrusion of one or more abdominal organs through the diaphragm into the thoracic cavity, with left lung hypoplasia and congestion of the intestines. The animal also had pneumonia, but it is difficult to determine whether this was the cause of death or a result of cramps

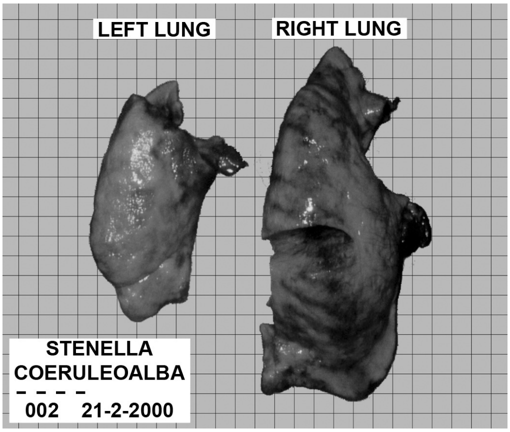

Figure 3. The left and right lungs of the striped dolphin; note the clear size difference. Background grid: 2 × 2 cm.

Figure 4. The congenital hole in the left side of the diaphragm (indicated by the fore and middle fingers of the researcher) through which the stomachs and intestines protruded into the thoracic cavity.

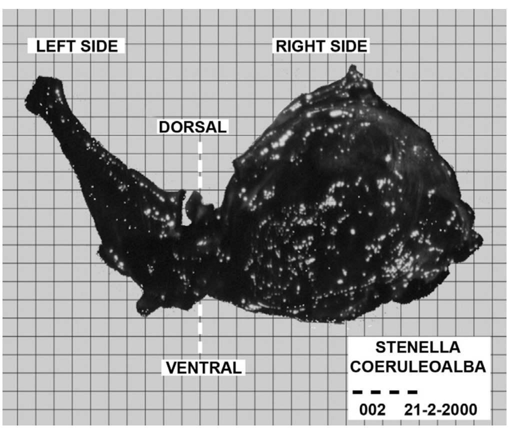

Figure 5. The left and right sides of the diaphragm of the striped dolphin; note the clear asymmetry. Background grid: 2 × 2 cm.

caused by congestion of the digestive tract, which may have made the animal inhale water. Most stranded odontocetes have pneumonia, but this is often a secondary effect of another illness (Kastelein et al., 1990, 1997).

Discussion

It is remarkable that this animal was in such good condition when it stranded. It must have fed shortly before stranding, despite the fact that its left lung was half the normal size and that after eating a meal the full forestomach must have reduced the volume of the left lung further. Also, it is curious that congestion of the digestive tract did not occur sooner. Perhaps the congestion that led to the animal's death was due to growth of the internal organs. In humans with CDH, the time of intestinal circulatory failure is also quite variable.

CDH is a severe birth defect in humans, characterized by a combination of failure in diaphragm formation and development, unilateral or bilateral lung hypoplasia, and postnatal pulmonary hypertension. The prevalence of CDH is about 0.3 per 1,000 births. Mortality rates vary depending on case selection, particularly in cases with associated malformations (Torfs et al., 1992). Although efforts have been made to understand the pathophysiology of CDH, current understanding of the etiology in humans remains sparse. The most common anatomical type of CDH is the posterolateral defect or Bochdalek hernia, which represents about 96% of the cases. In humans, the defect in the diaphragm is predominantly unilateral and on the left side as was the case in the striped dolphin in the present study.

In animals, spontaneous occurrence of CDH has been reported, mostly in case reports. Diaphragmatic defects are found in albino rats (Andersen, 1941), rabbits (Pasquet, 1974), dogs and cats (Wilson et al., 1971), and golden lion tamarins (*Leontopithecus rosalia*). In the latter, there is familial occurrence of ventromedial to lateral diaphragmatic hernias (Bush et al., 1980). However, there is also a teratogen that induces diaphragmatic hernia in rats. The herbicide nitrofen (2,4-dichlorophenyl-p-nitrophenyl ether) induces diaphragmatic defects of the Bochdalek type and lung hypoplasia in the offspring when administered to a pregnant rat between the 9th and 11th d of gestation (Costlow & Manson, 1981; Kluth et al., 1990; Tenbrinck et al., 1990). Thus, both environmental and genetic factors are considered to be important in the etiology of CDH (Beurskens et al., 2007).

Unfortunately, the tissues of the study animal were destroyed before they could be analyzed for the presence of toxicological agents such as polychlorinated biphenyls (PCBs) in serum or plasma or in adipose tissue. PCBs resemble nitrofen in their mechanism of action and general formula. Exposure to PCBs may have a similar effect on diaphragm development as exposure to nitrofen. Marine animals may be exposed to these toxic agents and are therefore at risk of developing birth defects.

Knowledge of the chromosomal pattern of the dolphin would be interesting because the combination of CDH and a micropenis could have been caused by a genetic syndrome. This combination has also been observed in humans, sometimes in association with a deletion of the terminal part of the long arm of chromosome 15 (Klaassens et al., 2005) or with other chromosomal deletion syndromes. Although a certain degree of homology exists between human and dolphin chromosomes (Bielec et al., 1998), it would be difficult to find small deletions.

In conclusion, the physical abnormalities—CDH and a micropenis—of this striped dolphin are unique. The combination of abnormalities was either due to a genetic syndrome or caused by maternal exposure to toxic agents.

Acknowledgments

We thank Willy Dabin (Centre de Recherche sur les Mammifères Marins, La Rochelle) for estimating the age of the dolphin based on growth layer counts in the teeth, and Nancy Jennings (Dotmoth.co.uk) for providing constructive comments on this manuscript. Rob Triesscheijn improved the labeling of the organs on the photographs by using *Photoshop*.

Literature Cited

Andersen, D. H. (1941). Incidence of congenital diaphragmatic hernia in the young of rats bred on a diet deficient in vitamin A. *American Journal of Diseases of Children*, *62*, 888-889.

Beurskens, N., Klaassens, M., Rottier, R., De Klein, A., & Tibboel, D. (2007). Linking animal models to human congenital diaphragmatic hernia. *Birth Defects Research (Part A)*, *79*, 565-572.

Bielec, P. E., Gallagher, D. S., Womack, J. E., & Busbee, D. L. (1998). Homologies between human and dolphin chromosomes detected by heterologous chromosome painting. *Cytogenetics and Cell Genetics*, *81*, 18-25.

Bush, M., Montali, R. J., Kleiman, D. G., Randolph, J., Abramowitz, D., & Evans, R. F. (1980). Diagnosis and repair of familial diaphragmatic defects in golden lion tamarins. *Journal of the American Veterinary Medicine Association*, *177*, 858-862.

Costlow, R. D., & Manson, J. M. (1981). The heart and diaphragm: Target organs in the neonatal death induced by nitrofen (2,4-dichlorophenyl-p-nitrophenyl ether). *Toxicology*, *20*, 209-227.

Di-Méglio, N., Romero-Alvarez, R., & Collet, A. (1996). Growth comparison in striped dolphins, *Stenella coeruleoalba*, from the Atlantic and Mediterranean coasts of France. *Aquatic Mammals*, *22*, 11-21.

Kastelein, R. A., Bakker, M., & Dokter, T. (1990). The medical treatment of three stranded harbor porpoises (*Phocoena phocoena*). *Aquatic Mammals*, *15*, 181-202.

Kastelein, R. A., Bakker, M. J., & Staal, C. (1997). The rehabilitation and release of stranded harbour porpoises (*Phocoena phocoena*). In A. J. Read, P. R. Wiepkema, & P. E. Nachtigall (Eds.), *The biology of the harbour porpoise* (pp. 9-61). Woerden, The Netherlands: De Spil Publishers.

Klaassens, M., van Dooren, M., Eussen, H. J., Douben, H., den Dekker, A., Lee, C., et al. (2005). Congenital diaphragmatic hernia and chromosome 15q26: Determination of a candidate region by use of fluorescent in situ hybridization and array-based comparative genomic hybridization. *American Journal of Human Genetics*, *76*, 877-882.

Kluth, D., Kangah, R., Reich, P., Tenbrinck, R., Tibboel, D., & Lambrecht, W. (1990). Nitrofen-induced diaphragmatic hernias in rats: An animal model. *Journal of Pediatric Surgery*, *25*, 850-854.

Pasquet, J. (1974). The fawn rabbit of Burgundy in teratology. Spontaneous malformations and malformations provoked by thalidomide. *Biological Medicine (Paris)*, *3*, 149-177.

Tenbrinck, R., Tibboel, D., Gaillard, J. L., Kluth, D., Bos, A. P., Lachmann, B., et al. (1990). Experimentally induced congenital diaphragmatic hernia in rats. *Journal of Pediatric Surgery*, *25*, 426-429.

Torfs, C. P., Curry, C. J., Bateson, T. F., & Honore, L. H. (1992). A population-based study of congenital diaphragmatic hernia. *Teratology*, *46*, 555-565.

Wilson, G. P., Newton, C. D., & Burt, J. K. (1971). A review of 116 diaphragmatic hernias in dogs and cats. *Journal of the American Veterinary Medicine Association*, *159*, 1142-1145.