

A "New" Diaphragm Following Prosthetic Repair of Experimental Hemidiaphragmatic Defects in the Pup

ROBERT J. TOULOUKIAN, M.D.

Hemidiaphragmatic defects were created in ten pups and repaired with Silastic Sheeting (Dow-Corning .007u) to determine the long-term results of prosthetic replacement of the diaphragm in growing subjects. All animals survived the operation without complication and grew and developed normally, weighing between 35 and 40 pounds at one year of age. The prosthesis gradually became located in the extreme left anterolateral costophrenic sulcus in nine of the ten dogs studied by sequential chest x-rays. Slight paradoxical movement was visible by fluoroscopy without eventration of the diaphragm. One animal developed an asymptomatic posterolateral diaphragmatic hernia nine months following operation. The other animals were sacrificed at one year of age. The liver, spleen and stomach were partially covered by a "new" muscle containing diaphragm having a mean area of 176 ± 8.3 cm, compared to the original defect which measured 15.89 ± 1.2 cm. Skeletal muscle cells extended into the margins of the prosthesis. The dorsolateral location of the "new" diaphragm is evidence that postnatal skeletal muscle growth may be derived from persisting precursor tissues.

OCCASIONALLY, THE DIAPHRAGMATIC defect in a newborn with a congenital diaphragmatic hernia is too large for primary closure to be achieved. The actual incidence of an extremely large foramen of Bochdalek defect or congenital agenesis of the entire hemidiaphragm based on review of several large series is extremely low.^{2,4,6,10,11,18} A family incidence of agenesis of the hemidiaphragm has been reported⁸ suggesting that this disorder is inherited as an autosomal recessive trait.

The size of the anatomic defect in an infant with the typical clinical and radiographic features of a posterolateral diaphragmatic hernia is never known prior to operation. A fluid and air-tight barrier which has the

From the Department of Surgery, Section of Pediatric Surgery, Yale University School of Medicine and the Yale-New Haven Hospital, New Haven, Connecticut

strength to prevent recurrence yet is flexible enough to move with respiration must be provided if the baby with a hemidiaphragmatic defect is to survive. We believe that a prosthesis is preferable to a variety of tissue reconstructive techniques involving the chest wall or adjacent viscera.

The following study using Silastic Sheeting for prosthetic repair of experimental hemidiaphragmatic defects in the pup was undertaken to determine: 1) the long-term clinical result of prosthetic substitutes of the diaphragm in the growing experimental animal, 2) the possibility that persisting marginal skeletal muscle may grow and reconstitute a portion of the diaphragmatic defect and 3) whether or not nonporous prostheses such as Silastic Sheeting are suitable for diaphragmatic replacement.

Materials and Methods

Ten pups weighing four to six pounds at six to eight weeks of age underwent excision and replacement of the entire left hemidiaphragm with Silastic Sheeting (Dow-Corning .007 u) through a midline abdominal incision under endotracheal halothane anesthesia. The prosthesis, an inert silicone rubber material, was anchored laterally to the ribs with encircling 4-0 Tevdek sutures and medially to the septum transversum and made identifiable for serial x-rays by affixing silver clips to its margins (Fig. 1).

Submitted for publication: March 3, 1977.

0003-4932-78-0100-0047-0075 © J.B. Lippincott Company

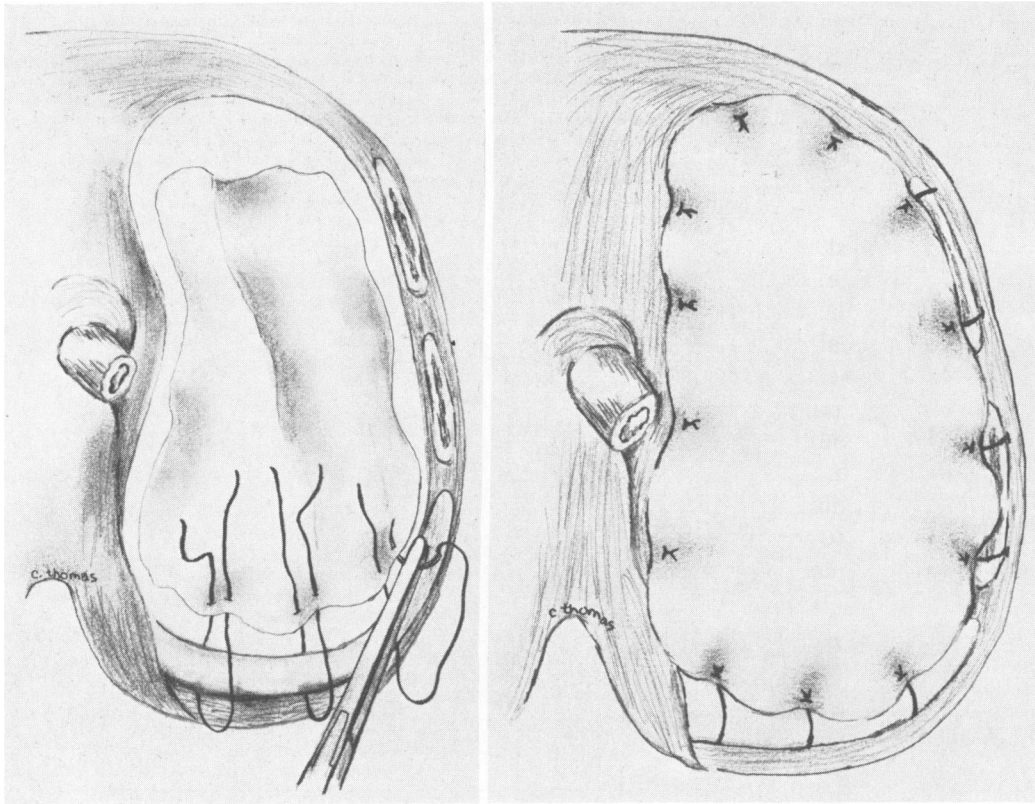
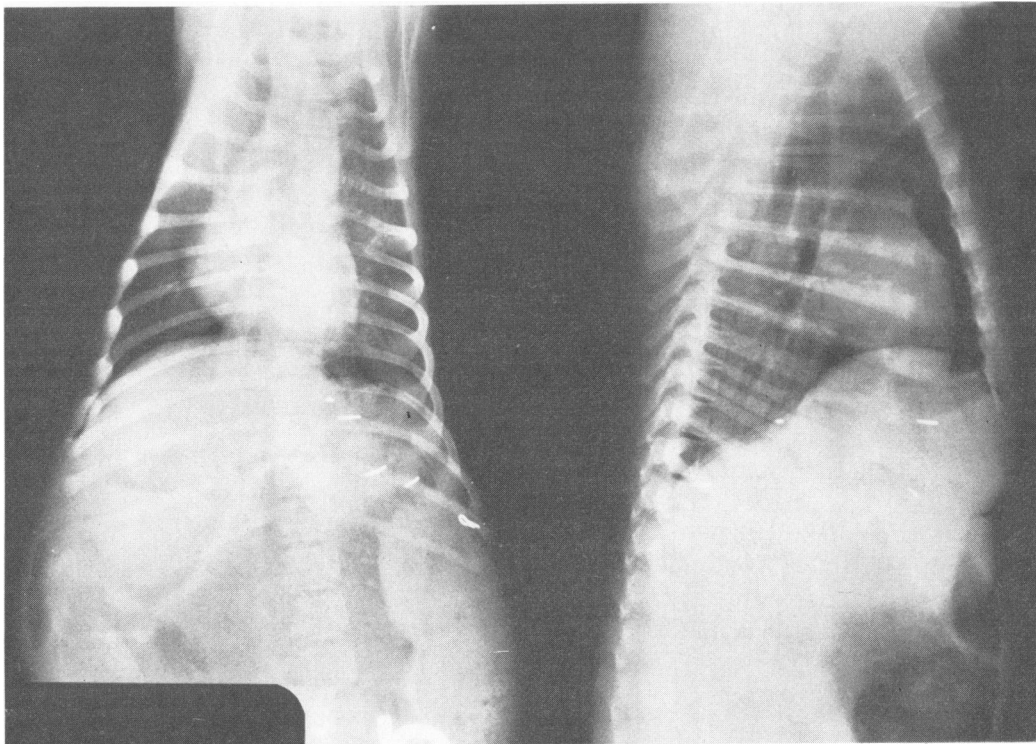


FIG. 1a and b. Technique of prosthetic repair of a left-sided hemidiaphragmatic defect through a transabdominal incision. Material is first anchored to the ribs (a, left) with encircling Tevdek sutures and the repair completed with interrupted sutures to the medial margin of the diaphragm and crus (b, right).

Results

All animals survived the operation without complication and grew and developed normally, weighing be-

tween 35 and 40 pounds at one year of age. Their activity level was identical to the control animals. The prosthesis gradually became located in the extreme left anterolateral costophrenic sulcus in nine of the ten dogs



FIGS. 2a-c. P-A and lateral view of the chest of a four week old pup immediately following hemidiaphragmatic replacement with Silastic sheeting (a) and ten months later (b and c). Note that the silver clips at the margin of the prosthesis are gradually displaced into the left costophrenic sulcus.

studied by sequential chest x-rays (Fig. 2). Slight paradoxical movement was visible by fluoroscopy performed at six, nine and 12 months under light intravenous pentothal anesthesia, but eventration of the diaphragm did not occur. One animal developed an asymptomatic posterolateral diaphragmatic hernia between the prosthetic and the marginal tissues six months after operation and was sacrificed. The other animals were sacrificed at one year of age. The liver, spleen and stomach were adherent to the undersurface of a membrane comprised of normal appearing diaphragmatic muscle and fibrous adhesions. The "new" diaphragm was intimately incorporated into the margins of the prosthesis, and could not be separated with opposing manual traction. The mean area of the Silastic patch used in the nine animals studied was 15.89 ± 1.2 cm compared to 176 ± 8.3 cm for the entire area of the "new" diaphragm at

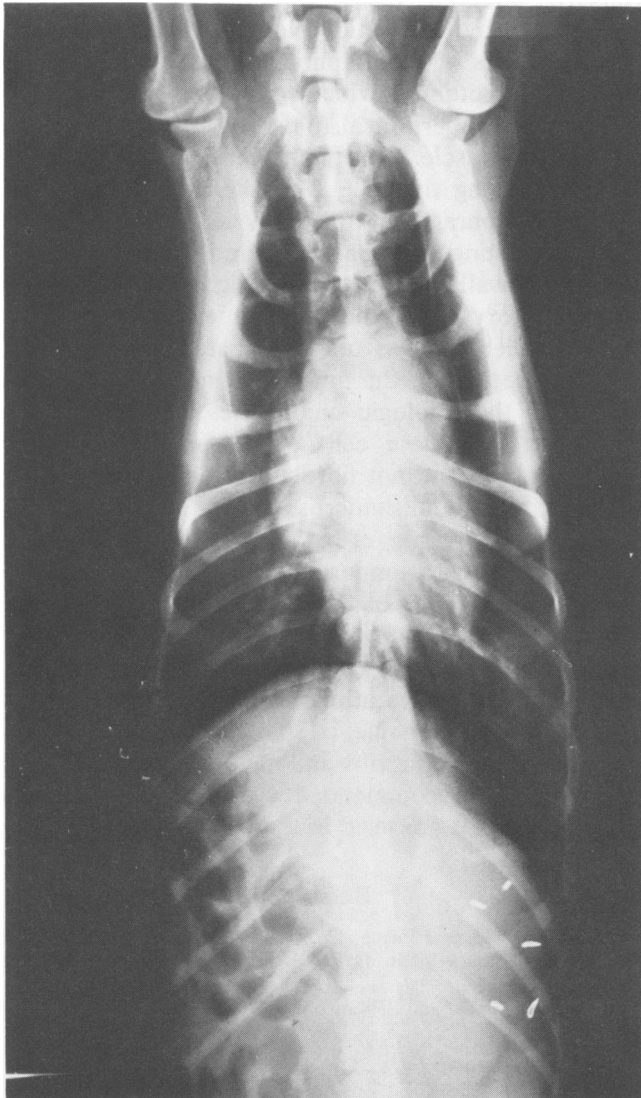


FIG. 2b. See legend for Figures 2a-c.

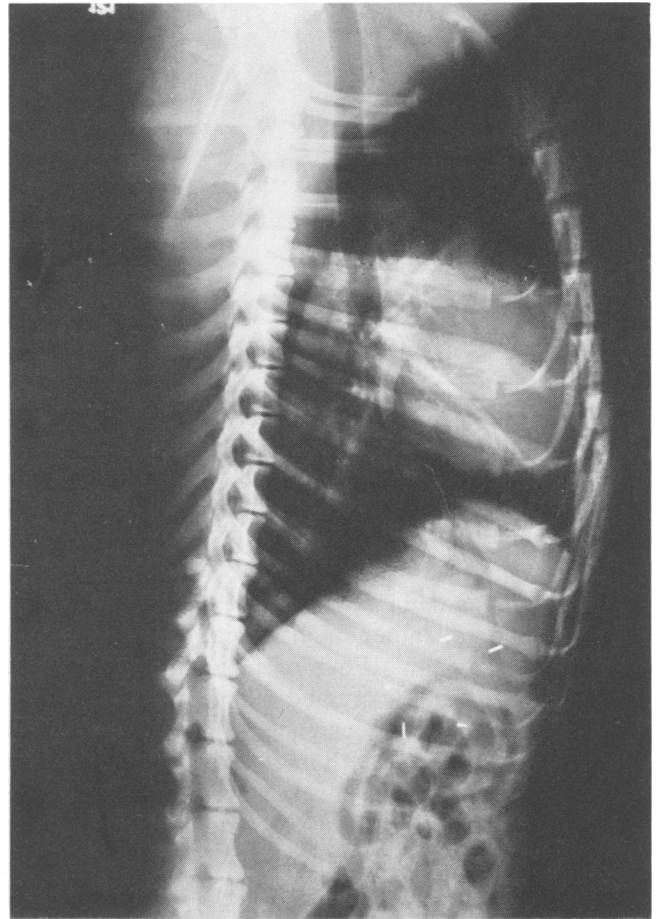


FIG. 2c. See legend for Figures 2a-c.

one year of age. The grossly normal appearing diaphragmatic muscle accounted for approximately 50% of the entire hemidiaphragmatic space.

Light microscopy using both routine Hematoxylin and Eosin as well as Mallory trichrome stains revealed bundles of normal skeletal muscle in the "new" diaphragm and immature cells containing large amounts of cytoplasm within the margins of the Silastic patch. These cells stained pinkish-red and had the histologic appearance of immature rhabdomyocytes (Fig. 3).

Discussion

Primary repair of a congenital diaphragmatic defect can be accomplished without difficulty in over 90% of cases. Occasionally, the posterolateral margin of remaining diaphragm is curled on itself and must be dissected free to gain adequate length. Defects too large to close primarily have been repaired by a number of different reconstructive procedures often with a high risk of recurrence, morbidity and mortality. Some of these are of historical importance. Neville and Clowes¹⁵ used the left lobe of the liver to repair

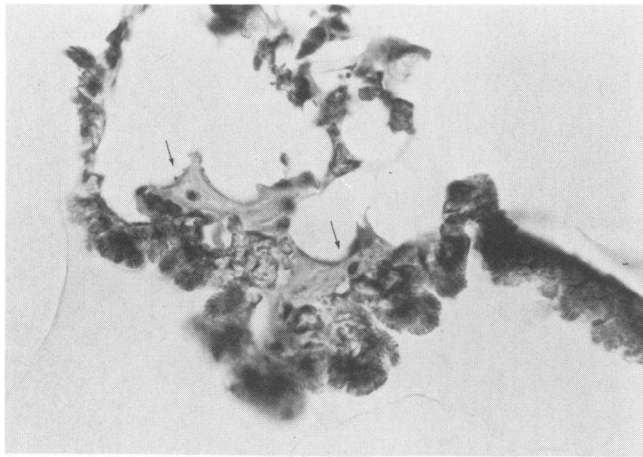


FIG. 3. Immature skeletal muscle cells (arrows) with abundant cytoplasm which stain pinkish-red with Mallory trichrome solution are found within the margins of the Silastic sheeting.

a hemidiaphragmatic defect in a newborn but reported a "subsequent rather high position of the newly created diaphragm." Four of 16 adult mongrel dogs with experimentally created hemidiaphragmatic defects had prompt recurrence of the hernia using this technique. Other authors recommended occluding the opening with renal fascia,¹⁹ free cutis grafts⁹ and "stomach, spleen, colon and omentum."¹² Gradual eventration if not prompt recurrence is certain to occur if the tissue partition does not have adequate tensile strength.

More extensive procedures using fascia and muscle have also been attempted. Rosenkrantz and Cotton¹⁶ created abdominal muscle pedicle flaps while Holcomb¹³ turned flaps of tissue "consisting of ribs, cartilages, intercostal muscles, nerves and blood vessels which could be fashioned from the lower anterior and lateral rib cage—and hinged anteriorly might serve as a diaphragm." Meeker and Snyder¹⁴ slid the anterior subcostal layer of muscle and fascia posteriorly much in the same way as the "doors of some garages are unfolded." Operations such as these are technically difficult and lose more time and blood in a sick newborn than is safe if an equally good alternative were available.

The ideal prosthetic material for replacement of a large hemidiaphragmatic defect in a growing baby must be tough, durable, inert and pliable. Mesh substances such as Marlex (polypropylene) or Mersilene (dacron) have been used as fascial substitutes in the repair of large ventral and inguinal hernias and with apparent success in the repair of large hemidiaphragmatic defects in the neonate.^{4,6,11,13,17} Nylon

tulle, a widely porous, pliable synthetic net and a closely woven calendered nylon fabric were tested as diaphragmatic substitutes in adult dogs.¹ Each functioned satisfactorily but evidence of new muscle growth was not reported. These mesh prostheses were eventually incorporated into the regional tissues by the host fibroblasts during the organization phase of wound healing but controlled studies of their fate as a diaphragmatic substitute in growing experimental subjects has never been conducted.

Silastic sheeting is a nonreinforced medical grade silicone rubber material which is nonporous and because of its inertness,⁵ could be displaced by active growth forces. This property of Silastic was demonstrated by the movement of the prosthesis into the anterolateral chest in growing dogs. The presence of skeletal muscle in the posterolateral chest is taken as evidence that the "new" diaphragm may be derived from persisting precursor tissues which have an anteriorly directed vector force. This hypothesis is supported by the embryologic evidence that the muscle fibers of the diaphragm are derived from the cervical myotomes which grow between the preformed pleuroperitoneal folds and by subsequent ingrowth of muscle from undifferentiated mesenchyme in surrounding tissues.²⁰ The skeletal muscle in the "new" diaphragm of the pups is probably derived from persisting secondary muscle ingrowth arising in the chest wall. Finding cells which stain pinkish-red with Mallory trichrome solution extending into the Silastic sheeting was not anticipated since previous experience using this material sutured to fascia⁷ indicates that biologic union does not occur. Final evidence that these cells are definitely of skeletal muscle origin and not fibroblasts requires the finding of myofibrils on ultramicroscopy.

Prosthetic repair of experimental hemidiaphragmatic defects with nonporous silon sheets was achieved without recurrence of the hernia in nine of ten pups studied. These findings provide an option to mesh materials as diaphragmatic substitutes, particularly when prompt restoration of intrathoracic negative pressure must be achieved. The technique of anchoring the patch to the ribs and medially to the crura, as described, is recommended whether or not silon or mesh materials are used in the repair.

References

1. Adler, R. H. and Firme, C. N.: The Use of Nylon Prostheses for Diaphragmatic Defects. *Surg. Gynecol. Obstet.* 104: 669, 1957.
2. Allen, M. S. and Thomson, S. A.: Congenital Diaphragm-

- matic Hernia in Children Under One Year of Age: a 24 Year Review. *J. Pediatr. Surg.* 1:157, 1966.
3. Benjamin, H. B.: Agenesis of the Left Hemidiaphragm. *J. Thorac. Cardiovasc. Surg.* 46:265, 1963.
 4. Boles, E. T., Jr., Schuller, M. and Weinberger, M.: Improved Management of Neonates with Congenital Diaphragmatic Hernias. *Arch. Surg.* 103:344, 1971.
 5. Brown, J. B., Ohlwiler, D. A. and Fryer, M. P.: Investigation of and Use of Dimethyl Siloxanes, Halogenated Carbons and Polyvinyl Alcohol as Subcutaneous Prostheses. *Ann. Surg.* 152:534, 1960.
 6. Cerille, G. J.: Foramen of Bochdalek Hernia: A Review of the Experience at Childrens Hospital of Denver, Colorado. *Ann. Surg.* 159:385, 1964.
 7. Cordero, L., Touloukian, R. J. and Pickett, L. K.: Staged Repair of Gastroschisis with Silastic Sheeting. *Surgery* 65:676, 1969.
 8. Feingold, M.: Aplasia of the Diaphragm. *Pediatrics* 47:601, 1971.
 9. Geever, E. D. and Merendino, K. A.: The Repair of Diaphragmatic defects with Cutis Grafts. *Surg., Gynecol. Obstet.* 95:308, 1952.
 10. Graivier, L., Dorman, G. W. and Votteler, T. P.: Congenital Diaphragmatic Hernia in Children. *Surg., Gynecol. Obstet.* 132: 408, 1971.
 11. Harberg, F. J., Meagher, D., Wetchler, S., et al.: Congenital Anomalies of the Diaphragm. Personal Experience with Thirty-five Consecutive Cases. *Am. J. Surg.* 132:747, 1976.
 12. Hedblom, C. A.: Diaphragmatic Hernia. *JAMA* 85:947, 1925.
 13. Holcomb, G. W., Jr.: A New Technique for Repair of Congenital Diaphragmatic Hernia with Absence of the Left Hemidiaphragm. *Surgery* 51:534, 1962.
 14. Meeker, I. A., Jr. and Snyder, W. H. Jr.: Management of Diaphragmatic Defects in Infants. *Am. J. Surg.* 104:196, 1962.
 15. Neville, W. E. and Clowes, G. H. A. Jr.: Congenital Absence of Hemidiaphragm and Use of a Lobe of Liver in its Surgical Correction. *Arch. Surg.* 69:282, 1954.
 16. Rosenkrantz, J. G. and Cotton, E. K.: Replacement of Left Hemidiaphragm by a Pedicled Abdominal Muscular Flap. *J. Thorac. Cardiovasc. Surg.* 48:912, 1964.
 17. Shaffer, J. O.: Prosthesis for Agenesis of the Diaphragm. *JAMA* 188:1000, 1964.
 18. Snyder, W. J. Jr. and Greaney, E. R. Jr.: Congenital Diaphragmatic Hernia: 77 Consecutive Cases. *Surgery* 57: 576, 1964.
 19. Weinberg, J.: Diaphragmatic Hernia in Infants: Surgical Treatment with the Use of Renal Fascia. *Surgery* 3:78, 1938.
 20. Wells, L. J.: Development of the Human Diaphragm and Pleural Sacs. (Carnegie) Contributions. *Embryology* 35:107, 1954.