CASE REPORTS

Three-Dimensional Reconstruction of the Anorectal Continence Organ in a 14-Week-Old Fetus

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Background/Purpose: The fetal development and anatomy of the muscular structures of the anorectal continence system are unclear. To the pediatric surgeon, these structures are of clinical relevance in reconstructive surgery. The aim of this study was to investigate the fetal development of the anorectal continence organ.

Methods: A male fetus (14 weeks postconceptionem) of 114-mm crown-rump length was sectioned serially at 18-μm intervals. The sections were stained, and relevant contours of the sections were transferred onto paper using a Zeiss Axioskop drawing apparatus. The drawings then were scanned and digitized.

Results: Three-dimensional images were created (and animated in a video). These have permitted the demonstration of isolated anatomic structures, the disassembling and reassembling of compound structures, as well as the visualization of structures from different angles.

Conclusions: Further studies are now undertaken of older fetal stages through to birth, as well as during postnatal stages. Comparative studies in animals and animations of isolated muscles also are required to show functional capacities. Such studies may lead eventually to an improvement of contemporary surgical techniques.
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INDEX WORDS: Fetal anorectal musculature, 3-dimensional reconstruction.

THE GOAL of surgery for anorectal malformations is fecal continence. Successful surgery requires knowledge of the anatomy. In the absence of normal anatomy, the knowledge of the phylogenetic and fetal development of the continence organ may help in the search for alternatives.

Our understanding of the anatomy and the function of anorectal muscles still is limited. Surgery is macroscopic. Fine anatomic structures are difficult to appreciate in the newborn.

The goal of this study was to establish an initial step in studying the development of anorectal muscles in the human with the aim of later comparing it with other species and set it into perspective with the regular development in the human until birth and later. The complexity of the organ may be better understood if its fetal development is known. The ultimate aim is to search for modifications of contemporary surgery for fecal incontinence.

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MATERIALS AND METHODS

In a 14-week p.c. male fetus the pelvis was prepared, decalcified, embedded, serially sectioned at 18-μm intervals, and Azan stained. One hundred twenty relevant contours of the sections were transferred onto paper using a Zeiss Axioskop drawing apparatus. The drawings then were scanned and digitized using Alias/Wavefront software. Three-dimensional pictures (3-D) were created and surface structures generated. The technique has been published previously.[1]

RESULTS

As early as in the 14th week of development, the anal canal can be observed in the 3-D picture as an S-shaped cylinder surrounded by a complex muscle system. Even details of the muscle complex can be seen. There are funnel-like and circular muscles (Fig 1).

Most of the funnel mass consists of 2 symmetric sections of the levator muscle. The ventral part of the levator muscle is much stronger. It originates, at 2 symmetric areas, from the posterior aspects of the pubic bones and in addition, more laterally, in a circular fashion from the fascias covering the internal obturator muscles. This circular insertion can be observed in the 3-D image as the curved uppermost margin of the levator muscle (Fig 2).

This section of the levator muscle is called the pubococcygeus muscle by anatomists. It is a thin plate that surrounds the rectum dorsally. It does not join with muscle fibers from the other side. A far smaller dorso-

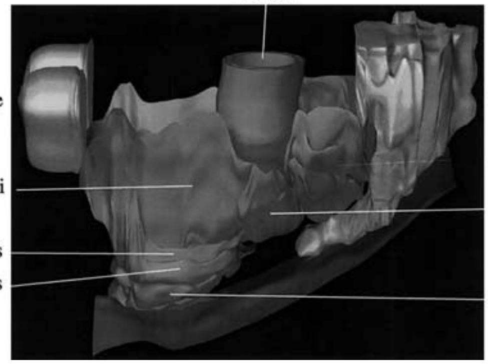

Fig 1. Overview of all muscles.

lateral segment of the levator muscle is called the iliococcygeus by anatomists. It originates from the area of the arcus tendineus all the way around to the spina ischiadica of the pelvis, and thus almost completes the funnel (Fig 3).

The back part of the funnel is closed by the coccygeus, a rather voluminous muscle clearly separated from the other levator segments. The caudal part forms a U-shaped sling (the puborectalis sling), which is open anteriorly (Fig 4).

The inferior tip of the funnel marks the transition of the rectum into the anus. There, the rectum leaves the funnel through the urogenital hiatus. the anus is surrounded by a circular system. The external sphincter is a 3-part circular system. Its deep, subcutaneous, and superficial segments are clearly discernible (Fig 5).

Most orally is the deep segment, followed aborally by the superficial and, most aborally, by the subcutaneous segment. In the anterior area of the anus, all 3 segments are joined. In addition, anteriorly, longitudinal muscle fibers subdivide the muscle radially (not seen on the 3-D reconstruction because only surface structures are represented). Furthermore, laterally and posteriorly, all 3 muscle segments can be identified clearly. The external sphincter appears not to make contact with any bony structures.

Both the funnel and the external sphincter are interconnected by additional intermediate muscle fibers. Originating from behind the pubic bone, these fibers run isolated from the funnel, separated from it by mesenchymal tissue. Behind the anus, the fibers join the external circular system. The cranial section of the intermediate fibers splits, with the inner and cranial fraction of the fibers running toward the coccyx. The caudal fraction of the fibers crosses behind the anal canal. This sling has been called the puborectalis sling. It cannot be distinguished from the uppermost (ie, the deep) part of the external sphincter.

When the ventral aspect of the funnel is electronicgraphically faded out, an additional narrow muscle band becomes visible. This originates at the inner aspect of the levator in close contact with the rectum. It inserts at the coccyx. It may be assigned to the rectococcygeus muscle (Fig 1).

DISCUSSION

The first promyeloblasts of the levator have been found in the sixth week of gestation, and those of the external sphincter in the seventh week of gestation.[2] Both main constituents of the anorectal continence organ, the levator funnel, and the external sphincter, have been shown as early as in the eighth week of gestation.[3] Our electronic reconstructions confirm these findings. Anatomic differences between sexes, however, could not yet be identified in our 14-week-old fetus. Nor was the

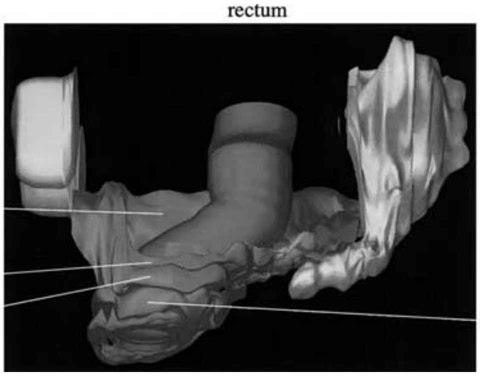

Fig 2. Levator muscle removed on the left side, exposing the uppermost margin of the levator on the right side.

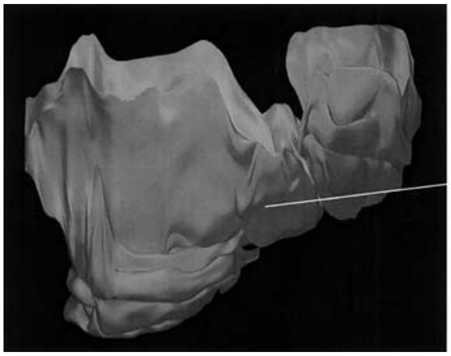

Fig 3. The ileococcygeus muscle as a separate structure.

anococcygeal ligament seen, which has been observed during surgery later in development.

The early development of the tripartite composition of the funnel also was known previously.[4] There is inconsistency in denominating the different muscle parts among anatomists and surgeons. The terms are misleading. Although called *pubococcygeus* and *ileococcygeus muscles*—suggesting origin and insertion, even function—in fact, only a small proportion of most caudally and posteriorly situated muscle fibers actually insert at the coccyx.

de Vries and Friedland[5] identified 2 layers of the pubococcygeus muscle, a craniolateral and a mediocaudal layer. Our findings confirm that the ventral funnel is 2 layered and that the cranial majority of the muscle mass does not make contact with the coccyx. Ventral to the anus, the caudal section further divides into 2 parts; only the cranial part contacts the coccyx and thus deserves the name *pubococcygeus*.

Our findings show that the puborectalis muscle is the major anterior component of the funnel, anchoring it to the symphysis. On its course posteriorly, it splits, together with the puborectalis. Posteriorly, it cannot be distinguished from the external sphincter. Other researchers also have seen one uniform structure.[2,3,6] The uniformity is so pronounced that the puborectalis muscle has even been considered an integral part of the external sphincter.[7] Even in the adult, this has been confirmed in magnetic resonance imaging.[8]

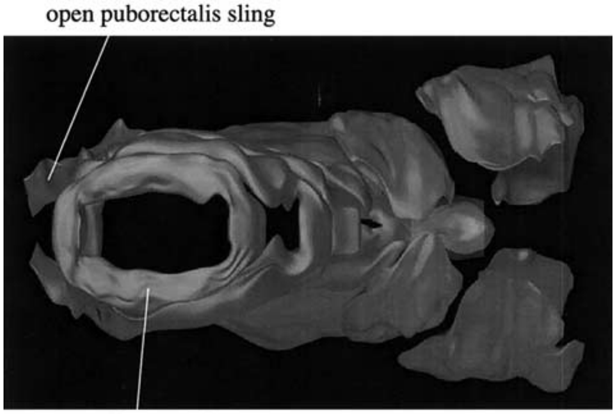

Fig 4. Puborectalis sling with open section anteriorly.

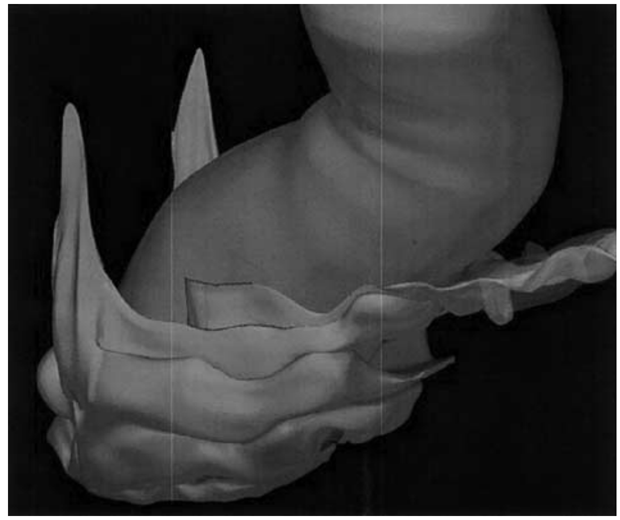

Fig 5. The 3 components of the externus muscle.

Most anatomists consider the external sphincter a 2-loop system[9]; surgeons are uncertain.[10,11] Levi has, in 18 human embryos, only seen 2 portions of the external sphincter.[2] The fetus investigated in our study showed 3 distinct circular structures.

Kluth et al[12,13] studied normal and abnormal fetal mice, and found, in normal embryos, that the region of the future anal opening can be identified soon after the very early establishment of the cloacal membrane. They concluded that the "fistula" in anorectal malformations resembles a normal anus at an ectopic position.

In anorectal anomalies of children, Paidas and Peña were unable to distinguish separate external sphincter components; they all appeared fused. In his usual surgical approach from the posterior, Peña[14] would encounter a longitudinal "muscle complex," which he considers to represent the junction of the levator muscle with the external sphincter. Electrical stimulation of the upper part (ie, the levator) would be answered by pulling the anus anteriorly. This is mostly the function of cranial majority of the pubococcygeus muscle mass, which does not make contact with the coccyx. Stimulation of the lower part (ie, the "muscle complex") results in a cranial movement and simultaneous closure of the anus. This appears to be the function of the caudal pubococcygeus muscle section. The current study is unable to contribute to the open question of the presence of an internal sphincter.[15]

In the organ of the fetus, we have found a clear tripartite funnel system and a tripartite circular system of muscles. The funnel is composed of the levator ani, iliococcygeus, and coccygeus muscles. The pubococcygeus and puborectalis muscles play an intermediary role. The ring system consists of the various parts of the sphincter ani externus muscle.

It appears from this study that the puborectalis is the key continence muscle, as on contraction its sling pulls forward where it is anchored to the pubic bone and simultaneously narrows circularly because it is also an integral part of the external sphincter. Possibly, its components are unable to contract individually and will react with mass contraction upon a physiologic stimulus.[7] The predominance of the puborectalis as an adaptation of both the levator ani and the pubococcygeus in this magnitude is only found in primates walking upright.[2]

The technique presented is now being applied to older fetal stages through to birth, as well as during postnatal stages. Comparative studies in animals and animations of isolated muscles are also planned to show comparative functional capacities. The animation technology allows us to animate every single muscle and to show its function in isolation and in combination with other muscles. Such studies may lead eventually to an improvement of surgical techniques.

REFERENCES

1. Haas A, Fischer MS: Three-dimensional reconstruction of histological sections using modern product-design software. Anat Rec 249: 510-516, 1997

2. Levi AC, Borghi F, Garavoglia M: Development of the anal canal muscles. Dis Colon Rectum 34:262-266, 1991

3. Fritsch H: Topography and subdivision of the pelvic connective tissue in human fetuses and in the adult. Surg Radiol Anat 16:259-265, 1994

4. Fritsch H, Fröhlich B: Development of the levator ani muscle in human fetuses. Early Hum Dev 37:15-25, 1994

5. de Vries PA, Friedland GW: The staged sequential development of the anus and rectum in human embryos and fetuses. J Pediatr Surg 9:755-759, 1974

6. Paidas CN, Peña A: Rectum and anus, in Oldham KT, Colombani PM, Foglia RP (eds): Surgery of Infants and Children, Scientific Principles and Practice. Lippincott-Raven, Philadelphia, PA 1997, pp 1323-1362

7. Shafik A: A new concept of the anatomy of the anal sphincter mechanism and the physiology of defecation. The external anal sphincter: Atriple loop system. Invest Urol 12:412-419, 1975

8. Hussain SM, Stoker J, Lameris JS: Anal sphincter complex: Endoanal MR imaging of normal anatomy. Radiology 197:671-677, 1995

9. Garavoglia M, Borghi F, Levi AC: Arrangement of the anal striated musculature. Dis Colon Rectum 36:10-15, 1993

10. Devroede G, Lamarche J: Functional importance of extrinsic parasympathic innervation to the distal colon and rectum in man. Gastroenetrology 66:273-276, 1974

11. Godlewski G, Prudhomme M: Embryology and anatomy of the anorectum. Basis of surgery. Surg Clin North Am 80:319-343, 2000

12. Kluth D, Hillen M, Lambrecht W: The principles of normal and abnormal hindgut development. J Pediatr Surg 30:1143-1147, 1995

13. Kluth D, Lambrecht W: Current concepts in the embryology of anorectal malformations. Semin Pediatr Surg 6:180-186, 1997

14. Peña A: Atlas of surgical management of anorectal malformations. Springer Verlag, New York, NY, 1990

15. Peña A, Hedlund H: Does the distal rectal muscle in anorectal malformations have the functional properties of a sphincter? J Pediatr Surg 25:985-989, 1990