In Utero Myelomeningocele Repair

Baby Steps and Giant Leaps for Fetal Surgery

AFTER decades of preparing the theoretical framework of fetal surgery,[1] the reality of it finally hit home in 2011 when Adzick *et al.* published their National Institutes of Health-supported study on outcomes after *in utero* surgery for myelomeningocele. Aptly named MOMS (Management of Myelomeningocele Study), the investigators terminated enrollment early after demonstrating that children who received *in utero* repair of myelomeningocele had a decreased incidence of ventriculoperitoneal shunt placement and less mental and motor disability at 30 months of age.[2] In this issue of Anesthesiology, Ferschl *et al.*[3] detail their early experience anesthetizing pregnant women undergoing *in utero* myelomeningocele repair. On the surface, it appears to be a review article. But we believe it belongs to a unique genre of scientific communication that provides an introduction to the anesthetic considerations for an emerging new technology. Unlike most review articles, there is little directly relevant anesthetic research data to review as fetal surgery takes its first steps—rigorous investigation in this area does not yet exist.

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Ferschl's description of anesthesia for fetal surgery mirrors similar pioneering articles in our field, such as Vandam's description of anesthetic considerations for renal transplantation[4] and early descriptions of anesthesia for liver transplantation,[5] laparoscopic cholecystectomy,[6] bariatric surgery,[7] and transcatheter aortic valve replacement,[8] to name a few. These publications have introduced us to emerging surgical technologies and heralded new eras marked by rapid evolution of surgical techniques, innovation, outcomes, and protocols. They have the power to stimulate and shape the development of research agendas around the emerging technology. For fetal myelomeningocele repair, examples of this might include the following: what is the most effective way to relax the myometrium during the procedure? Can certain anesthetic drugs or techniques prevent or reduce the risk of preterm labor? Is the fetus anesthetized and free of pain? Does it matter? What is the best comprehensive anesthetic approach that optimizes maternal and fetal well-being? Research into the anesthetic aspects of this highly complex procedure is still in its infancy.[9,10]

Ferschl *et al.* describe the anesthetic protocol from their center, which was one of the three that participated in MOMS. Although the protocol they describe is similar to that used at the other two original participating institutions, over time there will be evolution and divergence across centers that venture into this area. The challenges and strategies of successfully transferring this new technology from the trial centers to other medical centers are among the most pressing and difficult issues. Similar to solid organ transplantation and bariatric surgery, fetal surgery requires high levels of interprofessional teamwork, coordination, and longitudinal care to optimize outcomes and reduce and manage complications.

If *in utero* myelomeningocele repair is as beneficial as the MOMS trial suggests, then it would be desirable to expand the number of centers capable of performing it as rapidly as possible to ensure equitability of access. The intensive longitudinal care from mid-gestation surgery through delivery makes geographical location an access-limiting factor. As with other high-stakes, heavily resource-intensive, and costly programs, such as solid organ transplantation, how will these centers be chosen? Should there be criteria or limitations based on case volume, expertise, or geographic region? Hospitals stand to derive great benefit (financial, prestige, career

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advancement, *etc*.) from developing programs to provide this and other fetal therapies.[11] Tempering this zeal are the significant maternal and fetal risks of *in utero* surgery, such as uterine dehiscence and preterm delivery.[2]

In December 2011, one of us (Dr. Litman) participated as a representative of the American Society of Anesthesiologists in the Myelomeningocele Maternal–Fetal Management Task Force convened by the National Institute of Child Health and Human Development to establish (unpublished data) "minimum criteria for centers providing open fetal myelomeningocele repair." The development of these minimum criteria proved to be extremely challenging. What volume of cases should certify a pediatric surgeon, perinatologist, anesthesiologist, or neurosurgeon as competent? Excellent? Whatever the recommended criteria, whether it is based on case volume, or proven expertise, collaboration between centers, rather than in individual silos, will speed the advancement of this promising new therapy, while ensuring the very best and safest results. The Report on Maternal–Fetal Intervention and Fetal Care Centers from the American Academy of Pediatrics and the American College of Obstetricians and Gynecologists recently addressed this issue,[11] acknowledging the tenuous balance between providing geographical access to specialized procedures and maintaining excellence through high case volumes. This ideal balance across all regions is nearly impossible to achieve, and the challenge has been experienced by every other type of highly specialized emerging therapy. The American Academy of Pediatrics/American College of Obstetricians and Gynecologists report stressed that cooperation and collaboration between fetal care centers will promote reporting of short- and long-term maternal and fetal outcome data.

We applaud the recent proposed development of a fetal myelomeningocele repair registry by the North American Fetal Therapy Network (NAFTNet) [January 2013, verbal personal communication, Julie Moldenhauer, M.D., Assistant Professor, Departments of Clinical Obstetrics and Gynecology in Surgery, Perelman School of Medicine, University of Pennsylvania, Philadelphia, Pennsylvania]. Like the U.S. Scientific Registry of Transplant Recipients,‡ which was established in the 1980s, NAFTNet should facilitate and promote monitoring, track quality, identify centers of excellence and best practice, contribute to research and knowledge, cultivate collaboration, and demonstrate transparency to all stakeholders, including the public. Creation of the NAFTNet registry is consistent with our obligation to serve the public good with high standards, public responsibility, and accountability. Public access fosters our social contract and trusteeship, as well as our interest in maintaining professional integrity. Fetal surgery centers might carefully consider the lessons learned from the SRTR experience, including the controversies. The public availability of program-specific reports has been described as both "groundbreaking and controversial."[12] These public reports have many audiences including public and private payers, regulators, media, transplant centers, organ recipient candidates (and potential living donors), and are used for a host of purposes, both intended and unintended by the registry.[13] This high level of transparency and the expanding audience have had unintended consequences. Underperformance relative to peer programs, as reflected by poor program-specific outcome results, may have considerable negative consequences for programs. Such threats may cause centers to be more reluctant to accept higher risk patients, or to be more selective about accepting organs. Recently, a consensus conference of stakeholders was held to recommend modifications of reporting to address potential confounding factors related to baseline disease that may contribute to poor outcomes. Recommendations included careful revision of risk measurement and adjustment, changes to help protect and promote innovation, and provision of tools necessary to empower centers to facilitate greater quality assessment and improvement.[12] Lagasse recently emphasized the challenges and importance of accurate measurement of quality and outcomes risk-adjustment, as well as the complex political and financial implications of outcomes registries.[14]

Fetal myelomeningocele repair will not be the last complex, resource-intensive, large-team surgical intervention that will be introduced into the realm of anesthesiology. The speed with which emerging new surgical procedures take hold and spread from the pioneering programs to specialized centers, and then on to "value-added process" models[15] in the community seems to be accelerating. We should consider participating fully in NAFTNet, and its anticipated leadership role to ensure a systematic and collaborative approach to managing the development, implementation, assessment, refinement, improvement, safeguarding, promotion, and new program expansion and support of similar major emerging high-stakes perioperative advances in the future. The model's goal and our work should center upon value, namely, the outcomes and costs that matter to the primary stakeholders of our work, our patients.[16]

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Michael G. Richardson, M.D.,* Ronald S. Litman, D.O.†
*Department of Anesthesiology, Vanderbilt University School of Medicine, Nashville, Tennessee. †Perelman School of Medicine at the University of Pennsylvania, Department of Anesthesiology and Critical Care, The Children's Hospital of Philadelphia, Philadelphia, Pennsylvania. litmanr@email.chop.edu

‡ http://www.srtr.org/. Accessed April 1, 2013.

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