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Successful Pregnancy and Cesarean Delivery 22 Years after Separation in an Ischiopagus Tetrapus Conjoined Twin

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ISCHIOPAGUS tetrapus conjoining (twins with a common pelvis, four legs, and shared urinary, genital, and intestinal organs) occurs in approximately 1 in 800,000 live births.[1] Successful separation is still unusual, and survivors have significant long-term health problems. Pregnancy, although not recommended, can occur in some female survivors. We describe successful perioperative anesthetic management of cesarean delivery in a patient who is a separated ischiopagus twin.

Case Report

A 23-yr old primigravid, separated ischiopagus tetrapus twin with hypoplastic mandible, uterine didelphys, and absent pelvic musculature presented in preterm labor at 35 weeks gestation. The patient had been joined to her sister at the pelvic rim. The twins had duplex urogenital sinus openings and a shared colon. The two girls were separated at 15 months of age at our institution.[2] The twin died shortly thereafter because of recurrent episodes of pneumonia. Our patient underwent a total of 23 operations during general anesthesia between birth and her early teens. There were no anesthetic complications or airway difficulties during these operations. Surgeries included ileostomy, colostomy, formation of a nonneurogenic urinary neobladder, vaginoplasty, multiple perineoplasties with closure of vesicovaginal fistulae, repair of abdominal wall defects, leg lengthening, pelvic and hip reconstruction, and lower back surgery. The patient led an active life; she won a teen beauty contest, finished high school, and entered the work force as a supermarket cashier.

Her prenatal course was complicated by hyperemesis gravidarum and multiple urinary tract infections secondary to her altered urinary anatomy. She received nitrofurantoin for bacterial suppression for the final 10 weeks of her pregnancy.

Maternal weight gain added to the gait disturbances caused by the patient's chronically enlarged intertrochanteric distance (1.5 times normal). This prevented her from pursuing her job near the end of pregnancy. Her reconstructed pelvic bones created an obstructed pelvic inlet that was best described as a broad flat oval. Vaginal delivery was not considered possible.

A magnetic resonance image of the pelvis without gadolinium contrast at 30 weeks gestation was ordered to describe the pelvic anatomy and to facilitate surgical planning. It revealed uterine didelphys. The pregnancy was located in the superior uterus, which abutted the abdominal wall. The placenta was posteriorly placed. The fetus had a vertex presentation. No fetal anomalies were noted. Inferior and anterior to the gravid uterus was a smaller, empty uterus; both uteri joined into a single vagina. The empty uterus was that of her twin, who after separation did not have a vagina or a uterus.

The patient was counseled to avoid pregnancy in the future. An elective cesarean delivery at 38 weeks in combination with tubal ligation was planned. However, the onset of preterm labor at 35 weeks hastened the delivery plan. Amniocentesis revealed unfavorable fetal lung indices, so dexamethasone was given to enhance fetal lung maturity. Cesarean delivery was performed 3 days later because the fetal heart rate pattern showed mild repetitive decelerations with regular uterine contractions.

There were no cardiovascular, respiratory, or neurologic symptoms. Physical examination revealed a 1.5-m-tall, 59-kg parturient. She had a hypoplastic mandible, a Mallampati score of 2, and a free range of movement in the temporomandibular joints and cervical spine. An ileostomy and multiple lower abdominal scars were noted. A vertical midline lumbosacral scar was present. There was no lumbar lordosis. Neurologic examination was normal. Full blood count, electrolytes, chest x-ray, electrocardiogram, and echocardiogram were within normal limits.

Little information was available about the patient's previous back surgery; therefore, she underwent lumbosacral magnetic resonance imaging without gadolinium contrast to evaluate the feasibility of neuraxial anesthesia (figs. 1 and 2). The lumbar spine had normal alignment and segmentation with preserved disc spaces. The spinal cord seemed to be normal, with the conus medullaris terminating at T12–L1. A near-midline skin tag overlaid the L2–L3 interspace. At the L2–L3 interspace, the distances from the skin to the epidural space and the spinal nerves were 3.4 and 3.8 cm, respectively. Caudal images demonstrated not only a malformed pelvis with splayed iliac bones but also a midline dorsal defect in the caudal sacrum through which fat invaginated. No neural tissue was present in the sacrum, and the overlying soft tissues appeared normal.

The patient wanted to be awake during the delivery and requested that her surgery be performed during regional anesthesia. Because physical and radiologic evaluation suggested that neuraxial anesthesia was possible, we decided to attempt combined spinal–epidural anesthesia. We planned to change to a general anesthetic if the epidural or intrathecal spaces could not be identified, if the block level was too high or too low, or if the fetal status had suddenly deteriorated.

The skin tag noted on the magnetic resonance image allowed us to identify the L2–L3 interspace. In this patient, the Tuffier line would also have been an accurate landmark because it projected over the L4 spinous process. The patient sat during block placement. The epidural space was reached in a single attempt with an 18-gauge Tuohy needle at a depth of 4.0 cm. A 27-gauge Whitacre spinal needle was advanced through the epidural needle into the spinal fluid. Twelve milligrams hyperbaric spinal bupivacaine, 0.75%, and 100 μg morphine were injected. A 20-gauge catheter was inserted 5 cm into the epidural space. We laid the patient supine with left lateral tilt to displace the uterus off the inferior vena cava, which was in the normal position to the right of the vertebral column. A C6 sensory level was obtained within 12 min. The patient maintained good upper extremity strength throughout the case.

Surgery proceeded with a paramedian incision through the atrophic

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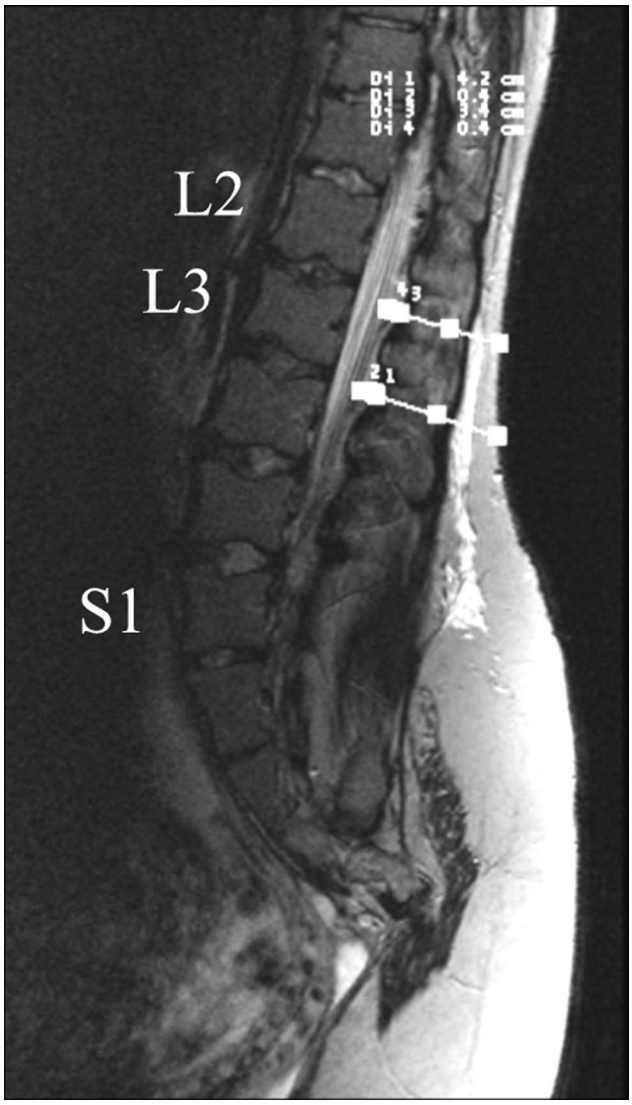

Fig. 1. T2-weighted sagittal magnetic resonance image of lumbar spine at 35 weeks' gestation. Annotations *3* and *4* at the L2–L3 level mark the distances from skin to epidural space (3.4 cm) and epidural to spinal nerves (0.4 cm), respectively.

abdominal wall. The patient's ileostomy functioned well throughout her pregnancy course and at the time of cesarean delivery was covered with an adhesive polyurethane membrane, which allowed direct visualization throughout the case. She had no rectus abdominus muscles and limited abdominal internal-oblique musculature. The abdominal musculature was thought to have been lost at the time of initial twin separation or was congenitally absent. Great care was taken at surgery to avoid these reconstructed areas, and additional efforts at abdominal closure were utilized to reinforce her attenuated fascial tissues with an overlapping technique to avoid future hernia formation.

A classic uterine incision was performed for visualization and to avoid the neobladder. A healthy male newborn (1- and 5-min Apgar scores of 9; weight, 2,200 g) was delivered. The solitary fallopian tube was ligated. Adhesions between the small bowel and the uterus were carefully lysed. The surgery lasted 66 min. Epidural dosing was not required, and the epidural catheter was removed at the end of surgery. Motor and sensory function returned to normal within 3 h.

Satisfactory postoperative analgesia was achieved with nonsteroidal analgesics in addition to the single dose of intrathecal morphine. The postpartum course was uneventful apart from a brief period of nausea.

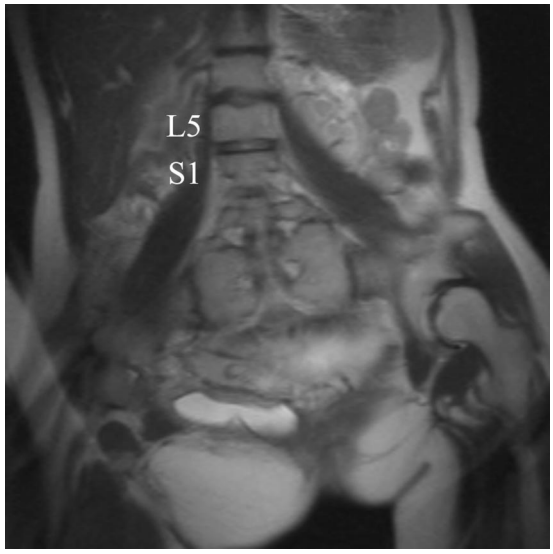

Fig. 2. T2-weighted coronal magnetic resonance image of lumbar spine. Note the open sacral hiatus and partial sacral agenesis.

The mother was discharged from the hospital in good condition on postpartum day 3. Feeding difficulties and mild respiratory issues secondary to prematurity complicated the infant's neonatal course. He was discharged home on day 9 from the neonatal intensive care unit, with a weight 2.18 kg.

Discussion

The incidence of conjoined twins is 1 in 50,000 to 1 in 100,000 deliveries in the United States.[3] Seventy percent of all conjoined twins are female. The ischiopagus type is rare, constituting only 6–11% of all conjoined twins. Only 63% of ischiopagus twins are separable.[4] Each case involves sharing of various normal or anomalous parts of the urinary, genital, and lower digestive systems. Separate, complete vertebral columns are found in ischiopagus twins; however, there is a high incidence of hyperextension, lordosis, scoliosis, and sacral fusion. In the tetrapus subtype of ischiopagus twinning, there are four legs and one large pelvic ring. Congenital vertebral anomalies (including thoracic hemivertebrae, missing lumbar vertebrae, and sacral agenesis) and visceral anomalies (including dextrocardia and lung hypoplasia) may accompany the pelvic and lower extremity problems.[1,5]

The only previously published record of a successful pregnancy in a separated conjoined twin is of a patient of the omphalopelvoischiopagus type.[5] Cesarean delivery was performed to avoid disruption of the fragile pelvic floor and reconstructed perianal area. An abdominal x-ray film showed lumbar lordosis, a short sacrum, and iliac crests with old osteotomies and absent pubic rami. Surgery with a midline vertical skin incision and vertical fundal uterine incision proceeded during epidural anesthesia.

Magnetic resonance imaging delineated our patient's nearly normal lumbar vertebral anatomy and gave us accurate information about the distance from skin to the epidural space. We choose to avoid using gadolinium contrast. Magnetopharmaceuticals such as gadolinium are considered safe for use during pregnancy, but these drugs are not innocuous.[6] Clinical trials in pregnancy are limited by small numbers and lack of reliable data on neonatal and maternal outcome.[7–9] The uncertainty of these risks outweighed in our minds the benefit of possible additional diagnostic information we could have obtained with gadolinium. The defect in closure of the sacral segments is most likely a result of the surgical separation of the bony fusion to the patient's twin rather than representing a true, congenital spina bifida. Although congenital lumbosacral spinal anomalies are generally considered to be a contraindication to neuraxial anesthesia, epidural catheters have been safely placed in some spina bifida patients after radiologic imaging to outline spinal anatomy.[10]

As conjoined twin separation techniques improve, more patients will survive to develop adult medical problems. This is the first report of combined spinal–epidural anesthesia for cesarean delivery in a pregnant separated ischiopagus twin. Magnetic resonance imaging defined the pelvic and vertebral anatomy and assisted the planning of the surgical and anesthetic procedures. A successful outcome was achieved despite the patient's multiple uterine, pelvic, and orthopedic anomalies.

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Peripartum Substitution of Inhaled for Intravenous Prostacyclin in a Patient with Primary Pulmonary Hypertension

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WE report a parturient with severe primary pulmonary hypertension who was receiving chronic intravenous prostacyclin (epoprostenol, PGI_2, Flolan®; GlaxoSmithKline, Research Triangle Park, NC) and changed to inhaled prostacyclin prepartum. Our strategy was to take advantage of the *selective* pulmonary artery (PA) vasodilation afforded by inhaled prostacyclin to minimize systemic side effects such as arterial hypotension and antiplatelet effects seen with intravenous administration. We theorized that this approach would allow for uninterrupted PA vasodilation while simultaneously minimizing the risk of antiplatelet effects and would thereby permit safer epidural catheter placement necessary for effective analgesia for planned forceps-assisted vaginal delivery.

Case Report

A 36-yr-old woman, gravida 4, para 3, weighing 77 kg was diagnosed with severe primary pulmonary hypertension during her sixth week of pregnancy and was receiving intravenous prostacyclin at 11.78 ng · kg^{-1} · min^{-1}. She presented at 36 weeks gestation for elective induction of labor with intravenous oxytocin, to be followed by a forceps-assisted vaginal delivery.

We obtained special permission from our institutional review board to use inhaled prostacyclin on our existing Food and Drug Administration Investigational New Drug Permit. Written, informed consent was obtained from the patient. Invasive monitoring was established with an arterial line and a PA catheter. An initial attempt at obtaining a wedge pressure was unsuccessful, and no further attempts were made. Initial vital signs showed a PA pressure of 80/37 mmHg, with a systemic

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blood pressure of 125/74 mmHg. We then started inhaled prostacyclin *via* facemask, while continuing intravenous prostacyclin. We used a concentration-based regimen for prostacyclin inhalation: 20,000 ng/ml nebulized at 8 ml/h (which corresponded to 35 ng · kg^{-1} · min^{-1}). A continuous nebulization system (MiniHEART® nebulizer; Westmed, Tuscon, AZ) was attached to a facemask with a Venturi attachment. Oxygen was administered to the Venturi attachment (to achieve an inspired oxygen concentration of 50%) and to the MiniHEART® nebulizer (prefilled with 15 ml prostacyclin) at 2 l/min. A constant infusion of prostacyclin at 8 ml/h was maintained to the nebulizer to replace nebulized losses. PA pressures decreased further, to 68/25 mmHg, and the intravenous prostacyclin was carefully weaned off over 30 min.

One hour later, a lumbar epidural catheter was placed, and a T12 sensory level was established with incremental doses of 0.25% bupivacaine. The patient remained stable, with a blood pressure of 139/82 mmHg, a PA pressure of 71/30 mmHg, and a cardiac index of 4.0 l · min^{-1} · m^{-2}. Two hours after epidural placement, we restarted intravenous prostacyclin at half the previous dose and rapidly titrated the dose upward as tolerated. This was done because there were no published reports of the efficacy of inhaled prostacyclin in this specific setting. Inhaled prostacyclin was stopped when a dose of 10 ng · kg^{-1} · min^{-1} intravenous prostacyclin was reached.

Over the next 3–5 h, the patient's PA pressures increased to levels above baseline (88/21 mmHg), while her systemic blood pressure (103/52 mmHg) and cardiac index (3.4 l · min^{-1} · m^{-2}) decreased. We decided that the reported benefits of selective PA vasodilation with inhaled prostacyclin would be desirable.[1–3] Inhaled prostacyclin was restarted at the previous concentration-based dose (20,000 ng/ml at 8 ml/h), and intravenous prostacyclin was rapidly weaned off. There was an immediate favorable response. The PA pressure decreased to 68/27 mmHg, and arterial blood pressure subsequently improved to 129/80 mmHg.

Six hours later, labor was induced with oxytocin. PA pressures increased somewhat during uterine contractions (peaks of 70–80/24–30 mmHg) but declined to baseline during uterine diastole (60–64/12–24 mmHg). A healthy infant was delivered by forceps 3 h after the start of the labor induction. The postpartum uterine tone was normal, and there was no excessive bleeding. The epidural catheter was removed shortly after the delivery. Inhaled prostacyclin was continued for 8 more hours to allow for smooth reintroduction of intravenous prostacyclin. The patient's postpartum course was uneventful, and she was discharged home on postpartum day 4.

Discussion

Primary pulmonary hypertension during pregnancy is a disease with significant morbidity, and a mortality rate as high as 50% has been reported.[4] Death may occur during or immediately after cesarean or vaginal delivery.[5,6] Most deaths, however, occur within the first postpartum week and are due to acute right ventricular failure and cardiovascular collapse. Successful management of right ventricular failure may be guided by invasive hemodynamic monitoring to optimize intravascular volume, reduce right ventricular afterload, and support right ventricular contractility. In this patient, a high forceps delivery was planned to avoid excessive pushing during delivery, which we anticipated would have deleterious effects on PA pressures and right ventricular function. We did not plan an elective cesarean delivery because maternal mortality is higher with cesarean than vaginal delivery.[4]

Inhaled and intravenous prostacyclin have been extensively studied, and their efficacy and side effects have been well described. We elected to switch this patient to inhaled prostacyclin for several important reasons: (1) it is as effective as inhaled nitric oxide,[3,7,8] is less costly, and does not carry the potential risk of toxicity seen with inhaled nitric oxide; (2) it has minimal systemic absorption[9,10] and therefore would potentially limit the antiplatelet activity seen with intravenous prostacyclin; and (3) the patient had already demonstrated a favorable response to intravenous prostacyclin.

Inhaled prostacyclin has been studied over a dose range of 0–50 ng · kg^{-1} · min^{-1}. No systemic side effects such as systemic hypotension or platelet dysfunction was observed over this dose range even though prostacyclin metabolites, with known antiplatelet effects, increased significantly.[11] We used a concentration-based regimen for prostacyclin inhalation: 20,000 ng/ml nebulized at 8 ml/h (which corresponded to 35 ng · kg^{-1} · min^{-1}). Prostacyclin and its major metabolite, 6-keto-prostaglandin F1α, are potent inhibitors of platelet aggregation *in vitro*,[9,10] raising concern about the use of regional anesthesia in patients receiving systemic prostacyclin. No clinical evidence of platelet dysfunction or excessive bleeding has been reported with inhaled use. Significant *in vitro* inhibition of platelet function does not usually occur until prostacyclin has been inhaled for 4–6 h.[9] In human studies, inhaled prostacyclin has not been shown to have a significant clinical effect on postoperative bleeding in postoperative cardiac surgery patients.[9] Prostacyclin is rapidly hydrolyzed (T1/2 = 5 min) to 6-keto prostaglandin F1α at acid or neutral pH. Prolonged antiaggregatory effects (up to 48 h) are seen only at alkaline pH (pH $>$ 7.8). Platelets incubated with (or exposed to) prostacyclin recover spontaneously and respond to aggregating agents within 15–60 min, depending on the initial prostacyclin concentration.[12]

We carefully considered both the potential risk of epidural hematoma due to the antiplatelet effects of prostacyclin and the risk of rebound pulmonary hypertension seen with intravenous drug withdrawal. At the time, we did not have access to test for potential antiplatelet function effects during the transition of intravenous to inhaled prostacyclin. Despite a normal prothrombin time, a normal activated partial thromboplastin time, and a normal platelet count antepartum, we could not fully exclude a potential or residual antiplatelet effect. A prolonged antiplatelet effect, however, seemed unlikely at normal pH,[12] and we anticipated plasma concentrations to be lower with inhaled than with intravenous prostacyclin, which would further minimize the risk of an epidural hematoma due to an antiplatelet effect.[12] Although we do not know if our approach was entirely safe, under the circumstances, our plan seemed reasonable.

In conclusion, inhaled prostacyclin seems to be a safe

and effective alternative to intravenous prostacyclin in the peripartum period. Its hemodynamic profile is favorable, it is relatively inexpensive, and it is easy to administer to spontaneously breathing patients. It may offer advantages over intravenous prostacyclin in those patients requiring a regional anesthetic technique.

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Preadmission Anesthesia Consultation Using Telemedicine Technology: A Pilot Study

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TELEMEDICINE has been defined as the delivery of healthcare and sharing of medical knowledge over a distance using telecommunications systems.[1] It uses modern high-speed telecommunication systems that allow interactive video-mediated clinical consultations. Telemedicine enables the delivery of healthcare irrespective of geographic location or ability to travel to tertiary healthcare centers.[2] In Canada, a significant population lives in remote regions away from tertiary care centers. At Toronto Western Hospital, 15% of patients are referred from remote regions. Telemedicine can potentially reduce travel costs and improve accessibility to health care.

Although telemedicine has been used by other medical and surgical specialties, there have been no reports in the literature evaluating telemedicine technology for anesthesia consultations.[3–5] We report the technical aspects and implementation of telemedicine anesthesia consultation, the first 10 patients interviewed, and their satisfaction regarding telemedicine consultations.

Case Report

Telemedicine Technology

The University Health Network in Toronto recently developed a partnership with the Northern Ontario Remote Telecommunication Health (NORTH) Network to provide telemedicine clinical consultations to residents of central and northern Ontario in Canada. The NORTH Network started in 1998 and now connects more than 65 distant sites throughout Ontario and Manitoba.

Telemedicine Setup

Both sites are equipped with videoconference television monitors and cameras that allow live two-way communication.

The Remote (Patient) Site

A light source is connected to two analog cameras (AMD-2500s; AMD Telemedicine Inc., Lowell, MA). The first camera functions as the room camera, and the second serves as the airway camera for intraoral views. A digital electronic stethoscope (AMD-3550; AMD Telemedicine Inc.) permits the transmission of heart and lung sounds.

Connection

The NORTH network uses the existing communications network Smart Systems for Health, provided by Bell Canada (Montreal, Quebec, Canada), operating with a bandwidth of 384 kbps.

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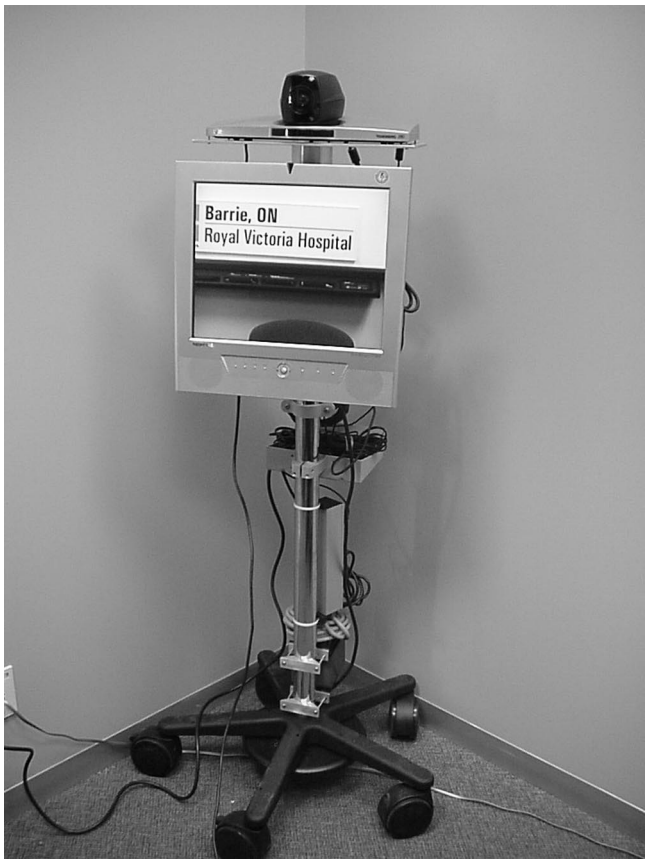

Fig. 1. Portable telemedicine unit with the viewing monitor and camera mounted on the unit.

The Consultant Site

The Tandberg 880 portable videoconference unit (Tandberg, New York, NY) (fig. 1) is mounted on a mobile stand and is located in the anesthesia preadmission clinic. The setup incorporates a monitor, a camera, a desktop computer, and a digital stethoscope. When connected to the remote site, the anesthesiologist can visualize, hear, and auscultate the patient using the digital stethoscope system (AMD-3550). The anesthesiologist inserts the digital stethoscope earpieces in exactly the same manner as a conventional stethoscope. The audible frequency range can be varied manually, depending on whether high-pitched or low-pitched sounds are being auscultated.

Identification of Candidates and Prearrangements

Institutional research ethics board approval was obtained for the study. Patient referrals were made by the surgeon's office. Potential candidates were identified by the preadmission booking clerk if their address was located outside of the Greater Toronto Area and if a telemedicine center was located near their home. The patient was then contacted by telephone and asked whether he or she wished to participate. An anesthesiologist with a special interest in telemedicine was then contacted regarding the suitability for telemedicine consultation. Patients who were mentally challenged, those who did not reside near a telemedicine facility, and those with complex medical issues that required additional preoperative investigations that were unavailable at the distant telemedicine site were excluded.

For agreeable candidates, medical information was then requested from the patient using the institutional preoperative patient questionnaire and from their family physician using the institutional preoperative history and physical examination form.

Telemedicine Consultation

An anesthesiologist was present at the consultant site while a nurse accompanied the patient at the remote site during anesthesia consultation. The anesthesiologist took a history from the patient as in a conventional consultation. Examination of the airway and respiratory and cardiovascular systems was performed. Mouth opening and the Mallampati score were assessed using the airway camera. The patient was then turned, and a side-view visual assessment of the airway profile, thyromental distance, and neck movement was made using the room camera. The digital stethoscope was used to auscultate the heart and lung sounds. The nurse at the remote center was instructed on the positioning of the stethoscope on the patient's chest and precordium. The rest of the consultation was conducted as per a conventional consultation.

Data Collection

Data were collected by an anesthesia research fellow. Degree of satisfaction by the patient and consulting and attending anesthesiologist were graded on five-point Likert response scales.[5] Postoperatively, the patient was visited by the anesthesia research fellow and asked whether he or she was satisfied with the telemedicine consultation. The consulting anesthesiologist was asked to rate his satisfaction with the telemedicine format after completion of the consultation. The attending anesthesiologist was asked on the day of the operation to rate his satisfaction with the telemedicine anesthesia consultation.

Results

Results from the first 10 consecutively completed telemedicine preadmission anesthetic consultations performed in 2003 were shown. Two patients were male, and eight were female. Four were had an American Society of Anesthesiologists (ASA) physical status classification of II, and six had an ASA physical status classification of III. The age of the patients was 58 $\pm$ 14 yr. The time to complete the telemedicine anesthetic consultation was 31 $\pm$ 7 min.

Nine of 10 patients stated that they were highly satisfied, and 1 of 10 were satisfied with telemedicine anesthesia consultation. Four anesthesiologists performed the telemedicine anesthesia consultation. Telemedicine consultation was satisfactory to both the consulting anesthesiologist and to the attending anesthesiologist. Eight of 10 consulting anesthesiologists were highly satisfied, and 2 of 10 were satisfied with the telemedicine consultation format. Ten of 10 attending anesthesiologists were highly satisfied with the preoperative anesthesia consultation. There were no reports of missing information from the attending anesthesiologists.

During preoperative screening, one patient was deemed inappropriate for telemedicine anesthesia consultation because he had obesity and inadequately investigated sleep apnea. The patient was assessed in person, and additional investigations were performed. One pa-

tient had her operation postponed as a result of the anesthesiologist noting documented results of an abnormal cardiac perfusion scan during the telemedicine consultation. After a coronary angiogram was obtained, the patient proceeded to surgery.

Discussion

The results of this pilot study indicate that preadmission anesthesia consultations using telemedicine technology can be successfully performed. Patients and consulting and attending anesthesiologists are very satisfied with telemedicine consultation.

Nine of 10 patients in this study were highly satisfied with anesthesia consultation by telemedicine. There are no reports in the literature evaluating telemedicine technology for preoperative consultations by anesthesiologists. In patients who underwent conventional preadmission anesthesia consultations, 92% thought that there was improved perioperative care, and 84% thought that they were less anxious as a result of the consultation.[6] Eighty-eight percent of patients surveyed by Murchison[7] indicated that the anesthesia consultation was beneficial to them. Ninety-three percent thought that the preadmission anesthetic consultation before cardiothoracic surgery was valuable.[8] The incidence of patient satisfaction in this study is in keeping with what is reported in the literature for conventional anesthesia consultation[6–8] and with telemedicine studies in other disciplines.[2,4,5,9] In 140 telemedicine pediatric consultations, 90% preferred the telemedicine format over the conventional consultation, and 71% were completely satisfied with the consultation.[9]

All attending anesthesiologists found that the quality of anesthesia consultation using telemedicine was satisfactory, and there was no missing information. A retrospective study of 43 patients booked for dentoalveolar surgery during general anesthesia found that 95% of the patients undergoing preadmission telemedicine consultations were judged to have been adequately assessed by oral and maxillofacial surgical residents for general anesthesia and nasotracheal intubation.[10] A junior surgical trainee, not an anesthesiologist, evaluated the airway using an airway camera.

The majority of the consulting anesthesiologists were satisfied with the telemedicine format for consultation. Because the physical examination by the anesthesiologist consists primarily of airway examination and heart and lung auscultation, the telemedicine format lends itself well to anesthesia consultations. The airway camera enables the anesthesiologist to assess the airway in a manner similar to that of a conventional consultation, but with better visualization of the posterior pharynx because of illumination. The digital stethoscope allows adequate examination of the cardiovascular and respiratory systems.

There are several limitations to the telemedicine consultation process. First, there are privacy concerns for the patient because they are being asked to provide personal details and exposure of the chest for auscultation. Second, telemedicine consultation does not permit any physical contact between the physician and the patient. Third, the patient and the anesthesiologist cannot speak at the same time.

This pilot study indicates that telemedicine preadmission anesthesia consultation can be successfully completed for patients residing in remote areas. Patients and anesthesiologists are satisfied with telemedicine consultations. However, a formal cost-effectiveness study must be performed before adopting this technology over conventional consultations on a wider scale. A randomized controlled trial with one group randomized to telemedicine consultation and the other to conventional consultation is needed. The aspects of cost, patient satisfaction, missing data, and necessary in-person visitation can then be compared between the groups to fully evaluate the cost effectiveness of this promising novel approach to preadmission anesthesia consultation.

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Neonatal Effects of Long-term Maternal Phenoxybenzamine Therapy

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PHEOCHROMOCYTOMA is an uncommon cause of hypertension in pregnancy, and little is known about the effects of pheochromocytoma or its therapy on the neonate. In this report, two neonates were delivered by successive cesarean deliveries of a woman with an unresectable intracardiac pheochromocytoma on long-term phenoxybenzamine therapy. Both neonates exhibited respiratory distress and hypotension in the first 72 h of life, requiring ventilation and inotropic support.

Case Report

First Pregnancy

A 24-yr-old gravida 3 para 0 woman presented at 38 weeks' gestation for an elective lower-segment cesarean delivery. An unresectable intracardiac pheochromocytoma had been diagnosed 11 months previously after she presented with paroxysmal hypertension, palpitations, headaches, and diaphoresis.

The patient was maintained on 20 mg phenoxybenzamine twice daily and had an otherwise uneventful pregnancy. Cesarean delivery was performed during epidural blockade (20 ml bupivacaine, 0.5%, plus 100 μg fentanyl), with invasive arterial blood pressure monitoring in place. Blood pressure was around 150/100 mmHg throughout. Her postoperative course was uneventful.

The 4,245-g male infant had an Apgar score of 9 at 1 min. By 5 min, he developed mottling of the extremities and respiratory distress and an Apgar score of 8. He was admitted to the neonatal intensive care unit, where his mean arterial pressure (MAP) was 31 mmHg. He was treated with continuous positive airway pressure and a dopamine infusion *via* an umbilical venous catheter. At 5 h and on 10 cm H_2O continuous positive airway pressure, a chest radiograph showed interstitial edema consistent with transient tachypnea of the newborn. Arterial blood gases were as follows: pH, 7.28; partial pressure of carbon dioxide (P_{CO_2}), 48 mmHg; and partial pressure of oxygen (P_{O_2}), 56 mmHg; on a fraction of inspired oxygen (F_{IO_2}) of 0.30. The infant was intubated, given 100 mg/kg surfactant, and placed on synchronized intermittent positive-pressure ventilation. His MAP remained 35–38 mmHg despite administration of dopamine at 24 $\mu g \cdot kg^{-1} \cdot min^{-1}$. A dobutamine infusion was commenced at 20 $\mu g \cdot kg^{-1} \cdot min^{-1}$, but his MAP at 10 h of life was unchanged. Hydrocortisone, 5 mg/kg and then 2.5 mg/kg every 6 hours, was added. MAP increased to greater than 50 mmHg from 25 h, and dobutamine and dopamine were weaned and ceased at 27 and 41 h, respectively. MAP remained stable thereafter at 49–52 mmHg. The infant was extubated at 48 h of life.

The mother and the infant were discharged home well on day 6. Breast-feeding was advised against on this occasion.

Second Pregnancy

Thirteen months later, the patient was admitted at 35 weeks' gestation with recurrent postural hypotension and gestational diabetes. Phenoxybenzamine was decreased to 10 mg twice daily before cesarean delivery at 38 weeks' gestation.

Epidural blockade was achieved with a total of 23 ml bupivacaine, 0.5%, and 100 μg fentanyl with a 16-gauge intravenous cannula and radial arterial pressure monitoring. Intravenous phentolamine boluses and oral phenoxybenzamine were required perioperatively for hypertension. The patient was stable the following day on 20 mg phenoxybenzamine twice daily.

At cesarean delivery, a 4,665-g female infant was delivered with Apgar scores of 9 and 9 at 1 and 5 min. Respiratory distress was noted at 10 min. She was admitted to the neonatal intensive care unit with a MAP of 40 mmHg and was placed on continued positive airway pressure of 11 cm H_2O. She was intubated at 19 h after an increase in inspired oxygen requirement to 60% and was given 100 mg/kg surfactant. Chest radiographs showed increased reticular markings. At 40 h, MAP decreased to 32–34 mmHg, and dopamine at a rate of up to 5 $\mu g \cdot kg^{-1} \cdot min^{-1}$ was given for a period of 9 h. She was extubated at 72 h of life.

The mother and the infant were discharged home on day 9. No neonatal hypotension was observed with breast-feeding.

Discussion

Pheochromocytoma is a rare catecholamine-secreting tumor, accounting for 1 in 400–800 cases of hypertension.[1] More than 200 cases of coincidental pheochromocytoma in pregnancy have been reported,[2] occurring in 1 out of 54,000 pregnancies.[2] Before 1969, maternal and fetal mortality were 48% and 55%, respectively.[3,4] With antepartum diagnosis and the use of α-blocking agents, fetal mortality has decreased to as low as 14%, and maternal mortality has decreased to zero.[5] Medical management of pheochromocytoma in pregnancy has led to delivery of a healthy infant in most cases.[6–13]

The placenta is the only non-neuronal tissue to express the norepinephrine transporter, responsible for reuptake of norepinephrine from the synaptic cleft and plasma compartments.[14,15] Little or no catecholamines cross the placental barrier, but they are taken up by the transporter and metabolized by placental catechol-*O*-methyl transferase and monoamine oxidase.[14–16]

Dahia *et al.*[17] report the case of a woman who was diagnosed with pheochromocytoma at 24 weeks' gestation and treated with prazosin. During cesarean delivery

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at 34 weeks, umbilical norepinephrine concentrations were 7% of maternal, suggesting that only a small percentage of maternal norepinephrine crosses the placenta. The neonate had no cardiovascular or respiratory problems. The noradrenaline concentration in umbilical cord blood has been measured at approximately 10% of maternal concentrations in other cases of pheochromocytoma.[18] Therefore, the direct hazardous effects of maternal catecholamines on the fetus are of little importance.

The fetus has a high basal rate of catecholamine production and low circulating catecholamine concentrations.[15] Intrauterine catecholamine clearance by the fetus is higher than under any other physiologic conditions, at 100–200 ml · kg^{-1} · min^{-1},[14,15] compared with a clearance rate of 30–50 ml · kg^{-1} · min^{-1} in adults.[15] The placenta accounts for nearly 50% of total fetal norepinephrine clearance.[15]

Circulating catecholamine concentrations increase exponentially at birth to support adaptation to extrauterine life.[14,15,19] Umbilical cord blood catecholamine concentrations in normal term neonates have been measured at almost 23 nmol/ml, similar to levels seen in pheochromocytoma. Any blockade of catecholamine receptors may impair the neonate's ability to cope with hypoxia and other stressors.[19]

Catecholamines at birth stimulate inotropy and chronotropy, redistribute blood flow to vital organs, stimulate clearance of alveolar and interstitial lung fluid and surfactant production, promote glycogenolysis and lipolysis, activate nonshivering thermogenesis, and stimulate nervous system functions such as arousal, muscular tone, and development of chemoreceptor activity.[20] Failure of catecholamine activation has been associated with neonatal hypoglycemia and idiopathic apnea of prematurity.[20]

The secretion of surfactant and secretion of lung fluid is mediated by β_2-adrenergic receptors. Rats given a β_2 antagonist in the immediate neonatal period cannot survive hypoxia. However, specific β_1 antagonists do not cause an increase in hypoxia-related mortality, suggesting that β receptors are not critical to cardiac function in the hypoxic neonate. In the neonatal rat, phenoxybenzamine causes a loss of tolerance to hypoxia, leading to cardiac failure, and cardiac α receptors are present in greater concentration than are β receptors. This suggests that intact α receptors may be important in the maintenance of cardiac function in the first week of life.[19]

α-Blocking agents have been used successfully in pheochromocytoma in pregnancy since the 1950s.[3] Phenoxybenzamine, an irreversible α-adrenoceptor antagonist with a half-life of approximately 24 h, is the most common agent used.[1] There are many reports of good neonatal outcomes in the presence of maternal phenoxybenzamine.[21] Indeed, until 1989, there were no reports of adverse fetal effects.[5] In two reported cases, phenoxybenzamine has been shown to cross the placenta.

Kothari *et al.*[2] reported a case of a woman who was diagnosed with bilateral pheochromocytomas at 26 weeks' gestation and treated with 60 mg phenoxybenzamine four times daily, propranolol, and nifedipine. She underwent cesarean delivery at 29 weeks. Apgar scores were 5 and 8 at 1 and 5 min, respectively, and the neonate was intubated and ventilated. No mention was made of neonatal hypotension, and the neonate was discharged at 7 weeks. The fetal:maternal plasma phenoxybenzamine ratio was 1.13:1, indicating that phenoxybenzamine crosses the placenta and accumulates in the fetus.

Santeiro *et al.*[22] reported a case of a woman who was diagnosed with pheochromocytoma at 33 weeks' gestation and managed with phenoxybenzamine and labetalol for 26 days before cesarean delivery. Apgar scores were 2 and 8 at 1 and 5 min, respectively, and the neonate was intubated briefly for poor respiratory effort. He also had mild hypotension for the first 3 days of life. The fetal: maternal plasma accumulation ratio of phenoxybenzamine was 1.6:1. The authors suggest that neonates of mothers receiving phenoxybenzamine should be monitored for the first few days of life for hypotension and respiratory depression.

Both neonates in this case exhibited hypotension and respiratory distress in the first 3 days of life. The fetus is largely isolated from high maternal catecholamine concentrations by the placenta but is exposed to phenoxybenzamine. Intact catecholamine function is important in adaptation to the extrauterine environment.

We postulate that α blockade by maternal phenoxybenzamine caused hypotension in these neonates. In the first neonate, dopamine and dobutamine did not seem to improve blood pressure. Glucocorticoids regulate the expression of cardiovascular adrenergic receptors and are used to counteract the down-regulation of these receptors in critically ill neonates.[23] They may have improved the hypotension in the first neonate, who was exposed to a greater dose of phenoxybenzamine and had a more prolonged and more resistant period of hypotension.

Because the neonatal heart has a large proportion of α-receptors, there may have been an element of cardiac failure contributing to the respiratory distress seen. However, the relation between phenoxybenzamine and respiratory distress is not clear.

There have been many reported cases of healthy neonates delivered to mothers receiving phenoxybenzamine. Because pheochromocytoma is usually resected if diagnosed in the first 20 weeks of gestation, these neonates were exposed to phenoxybenzamine for shorter periods of time than in this case.

There is no data on the transmission of phenoxybenzamine *via* breast milk or its effects on the breast-fed

neonate.[24] However, the milk-to-maternal drug plasma concentration for most drugs is 0.5–1.0, indicating that less than 1% of a maternal dose is available to the infant.[25] No hypotension was seen in the breast-fed infant in this case. We suggest that initiation of breast-feeding in a monitored environment is appropriate.

In conclusion, neonates born to mothers receiving phenoxybenzamine should be monitored closely in an intensive care unit, with particular observation for hypotension and respiratory distress.

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