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Systolic Anterior Motion of the Anterior Mitral Leaflet after Heart Transplantation

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TRANSESOPHAGEAL echocardiography (TEE) is a new tool for the perioperative treatment of cardiac surgery patients. We report a clinical case to illustrate the usefulness of TEE for evaluating morphologic and functional features of a cardiac graft. Mitral regurgitation (MR) secondary to left ventricular outflow tract obstruction with systolic anterior motion (SAM) of the anterior mitral leaflet was diagnosed preoperatively with TEE. Medical treatment of the recipient was changed consequently to prevent hemodynamic deterioration.

Case Report

A 50-yr-old man underwent an orthotopic cardiac transplantation because of a dilated cardiomyopathy. The donor heart was retrieved from a 53-yr-old man who died from a cerebral hemorrhage. The donor had been treated for hypertension. A transthoracic echocardiogram of the donor heart showed a normal left ventricle with normal systolic function and no valvular abnormalities. The donor heart was arrested with cold St Thomas' Hospital cardioplegic solution, excised, and then stored in 4°C cold saline solution.

The recipient was anesthetized with fentanyl (total amount, 80 μg/kg), pancuronium bromide, and midazolam and was monitored with a radial artery, pulmonary artery catheter and standard anesthesia monitors. A TEE was performed with a biplane transesophageal probe (5 MHz, Aloka 870; Aloka CO, Ltd., Tokyo, Japan). The donor heart was grafted according to the standard procedure with atrial anastomoses,[1] but keeping only a small atrial cuff around the four pulmonary vein orifices. Total ischemia time for the graft was 3 h. Weaning from cardiopulmonary bypass was performed using isoproterenol (0.05 $\mu g \cdot kg^{-1} \cdot min^{-1}$. Subsequent hemodynamic measurements showed sinus tachycardia (130 beats/min), systolic blood pressure of 105 mmHg, pulmonary capillary wedge pressure of 14 mmHg, cardiac index of 3.1 $l \cdot min^{-1} \cdot m^{2-1}$, and stroke volume of 42 ml. TEE examination showed a hyperkinetic left ventricle with global left ventricle hypertrophy (wall thicknesses of 14, 15, and 15 mm for anterior, septal, and posterior walls, respectively). Color flow mapping demonstrated grade II MR, assessed by the regurgitant jet diameter at the origin (4 mm) and the regurgitant jet area (5 cm^2). Bidimensional analysis showed a SAM of the anterior mitral leaflet (fig. 1). Mitral valve anatomy was otherwise normal. Given that medical management (hypovolemia, tachycardia, vasodilation, and enhanced inotropism caused by isoproterenol infusion) might cause or aggravate SAM and MR, isoproterenol infusion was replaced by 10-$\mu g \cdot kg^{-1} \cdot min^{-1}$ dopamine infusion (with expected alpha effects), and 500 ml modified fluid gelatine was quickly infused. Fifteen minutes later, heart rate decreased from 130 to 85 beats/min, cardiac index was 2.8 $l \cdot min^{-1} \cdot m^{2-1}$ with a stroke volume of 59 ml, and pulmonary capillary wedge pressure did not change. MR and SAM both completely resolved (fig. 2). The patient remained hemodynamically stable.

The postoperative course was uneventful. Postoperative transthoracic echocardiogram on day 10 showed a left ventricular hypertrophy with normal systolic function and trivial MR with normal mitral valve function.

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Discussion

We report a case of MR secondary to SAM of the anterior mitral leaflet after heart transplantation. This phenomenon has been previously described after other surgical procedures, including mitral valve repair and aortic valve replacement.[2,3] This phenomenon of left ventricular outflow tract obstruction with SAM and MR was initially described in hypertrophic cardiomyopathies with specific anatomic features[4,5]: a thickened interventricular septum and an anterior and medial displacement of the anterior mitral papillary muscle

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Key words: Echocardiography; mitral regurgitation.

leading to mitral-septal contact with possible SAM and MR and, finally, mitral leaflet elongation.[6] Two mechanisms have been proposed for such mitral-septal contact[7]: a flow drag on the protruding mitral leaflet or Venturi effect through a narrowed left ventricle outflow tract. Both mechanisms can be worsened by hemodynamic conditions such as tachycardia, vasodilation, hypovolemia, and increased inotropism. After heart transplantation, common use of the β-adrenergic agonist isoproterenol with resultant vasodilation, tachycardia, and increased myocardial contractility, along with iatrogenic hypovolemia to prevent graft dilation, might cause or aggravate SAM of the anterior mitral leaflet.[8] Although we did not document it in this case, the timing of recipient atrial contraction may also have a substantial effect on the atrial contribution to left ventricular filling as well as pulmonary venous return.[9] Intrinsic hypertrophy of the cardiac graft, although not observed on a technically difficult transthoracic echocardiogram, was likely a major predisposing factor for SAM of the anterior mitral leaflet and associated MR. Alterations in medical management elicited by the TEE findings of SAM of the anterior mitral leaflet with associated MR enabled us to correct the MR and prevent possible deterioration in the recipient's hemodynamic status.

Mitral regurgitation has been previously reported on postoperative transthoracic echocardiogram and TEE examinations performed on heart recipients.[10-12] However, the cause was different than in this case. Stevenson *et al.*[12] noted MR in 14 of 16 post-transplant patients and attributed this high rate to surgical techniques that resulted in an enlarged left atrium with mitral annular dilatation and resultant anuloventricular disproportion. This disproportion was explained as follows: left ventricle is restrictive because of increased contractility due to decreased vagal/tone or higher heart transplant rates; the mitral anulus is dilated in absolute value during the systole and, because of an abnormal geometry of the left atrium, configured as a "snowman" with the actual anastomoses. This anatomic configuration was not observed in our current case because we kept a minimal cuff of left atrium around the pulmonary vein orifices.[13] Other investigators suggested that multivalvular regurgitation observed after heart transplantation might be a result of mild edema of the cardiac structures.[10] Indeed, they found a significant left ventricular mass reduction within the first postoperative weeks, with a progressive resolution of valvular regurgitation. In a more recent work, the incidence of MR after cardiac transplantation was 48%; MR was mild and correlated with neither hemodynamic indices nor atrial distortion, by contrast with tricuspid regurgitation.[11] Taken together, all of these works show that MR is frequent after heart transplantation; nevertheless, the mechanism often remains controversial.

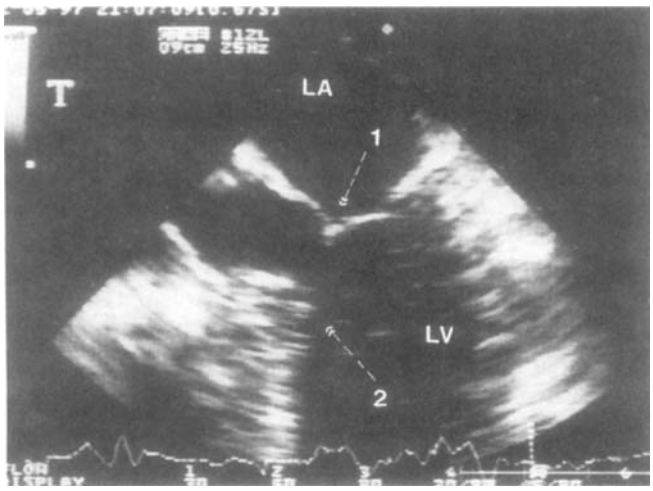

Fig. 1. Transverse view showing the systolic anterior motion of the anterior mitral leaflet (*arrow 1*) and the interventricular septum hypertrophy (*arrow 2*). LA = left atrium; LV = left ventricle.

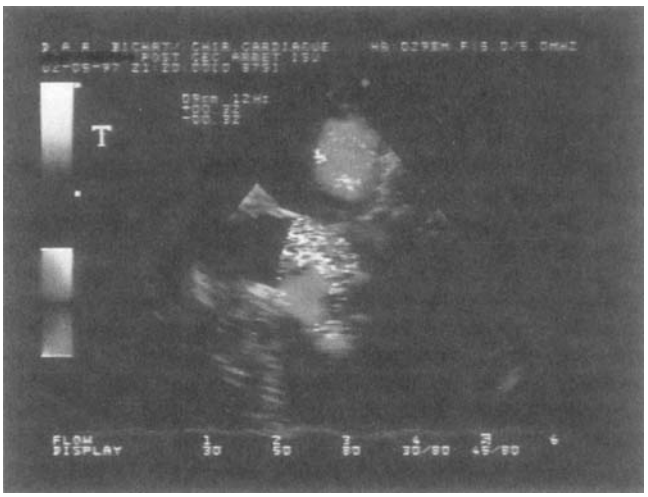

Fig. 2. Transverse view after the medication changes showing no mitral regurgitation.

References

1. Shumway NE, Lower RR, Stofer RC: Transplantation of the heart. Adv Surg 1966; 2:265-84

2. Mihaileanu S, Marino JP, Chauvaud S, Perier P, Forman J, Vissoat J, Julien J, Dreyfus G, Abastado P, Carpentier A: Left ventricular outflow obstruction after mitral valve repair (Carpentier's technique). Circulation 1988; 78(suppl I):I-78-84

3. Cutrone F, Coyle JP, Novoa R, Stewart R, Currie PJ: Severe dynamic left ventricular outflow tract obstruction following aortic valve replacement diagnosed by intraoperative echocardiography. ANESTHESIOLOGY 1990; 72:563-6

4. Schwammenthal E, Nakatani S, He S, Hopmeyer J, Sagie A, Weyman AE, Lever HM, Yoganathan AP, Thomas JD, Levine RA: Mechanism of mitral regurgitation in hypertrophic cardiomyopathy. Circulation 1998; 98:856-65

5. Wigle ED, Rakowski H, Kimball BP, Williams WG: Hypertrophic cardiomyopathy: Clinical spectrum and treatment. Circulation 1995; 92:1680-92

6. He S, Hopmeyer J, Lefebvre XP, Schwammenthal E, Levine RA: Importance of leaflet elongation in causing systolic anterior motion of the mitral valve. J Heart Valve Dis 1997; 6:149-59

7. Sherrid MV, Dellia E, Mogtader A, Dwyer EM: An echocardiographic study of the fluid mechanics of obstruction in hypertrophic cardiomyopathy. J Am Coll Cardiol 1993; 22:816-25

8. Udelson JE, Cannon RO, Bacharach SL, Rumble TF, Bonow RO: β-adrenergic stimulation with isoproterenol enhances left ventricular diastolic performance in hypertrophic cardiomyopathy despite potentiation of myocardial ishemia. Circulation 1989; 7:371-82

9. Valantine HA, Appleton CP, Hatle LK, Hunt SA, Stinson EB, Popp RL: Influence of recipient atrial contraction on left ventricular filling dynamics of the transplanted heart assessed by Doppler echocardiography. Am J Cardiol 1987; 59:1159-63

10. Cladellas M, Abadal ML, Pons-Llado G, Ballester M, Carreras F, Obrador D, Garcia-Moll M, Padro JM, Aris A, Caralps JM: Early transient multivalvular regurgitation detected by pulsed Doppler in cardiac transplantation. Am J Cardiol 1986; 58:1122-4

11. De Simone R, Lange R, Sack FU, Mehmanesh H, Hagl S: Atrioventricular valve insufficiency and atrial geometry after orthotopic heart transplantation. Ann Thorac Surg 1995; 60:1686-93

12. Stevenson LW, Dadourian BJ, Kobashigawa J Child JS, Clark SH, Laks H: Mitral regurgitation after cardiac transplantation. Am J Cardiol 1987; 60:119-22

13. Deleuze PH, Benvenuti C, Mazzucotelli JP, Perdrix C, Le Besnerais P, Mourtada A, Hillion ML, Patrat JF, Jouannot P, Loisance DY: Orthotopic cardiac transplantation with direct caval anastomosis: Is it the optimal procedure? J Thorac Cardiovasc Surg 1995; 109:731-7

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Case of Cowden's Disease that Caused Airway Obstruction during Induction of Anesthesia

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COWDEN'S disease, known also as multiple hamartoma syndrome, is a rare, predominantly inherited condition characterized by various ecto-, meso-, and endodermal benign and malignant tumors that may affect the skin; oropharyngeal, laryngeal, and gastrointestinal mucosa; thyroid; breast; and other organs.[1,2] We report a case of airway obstruction during induction of general anesthesia caused by the presence of extended multiple papillomas on the lingual tonsils, epiglottis, and the surrounding structure, for which a diagnosis of Cowden's disease was made postoperatively.

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Key words: Hamartoma; lingual tonsil; papilloma.

Case Report

A 55-yr-old woman was scheduled for elective mastectomy for cancer of the right breast. At the age of 12 yr, the patient had undergone bilateral tonsillectomy. Three years later, she underwent surgery for chronic sinusitis. At age 27 yr, she underwent partial thyroidectomy for benign adenomatous changes in the right lobe of the thyroid gland. The patient denied having had any complications from previous anesthesia. Preoperative plain radiograph and computed tomography images showed a giant adenoma in the left lobe of the thyroid gland that did not show any signs of compressing the trachea. The patient had neither history of decreased activity nor any obvious breathing difficulty.

Anesthesia was induced with intravenous propofol (1.2 mg/kg) and vecuronium (0.1 mg/kg). While performing tracheal intubation, visualization of the hypopharynx by direct laryngoscopy showed a mass of tumors occupying the airway. The epiglottis and the surrounding structures could not be identified because of the presence of numerous

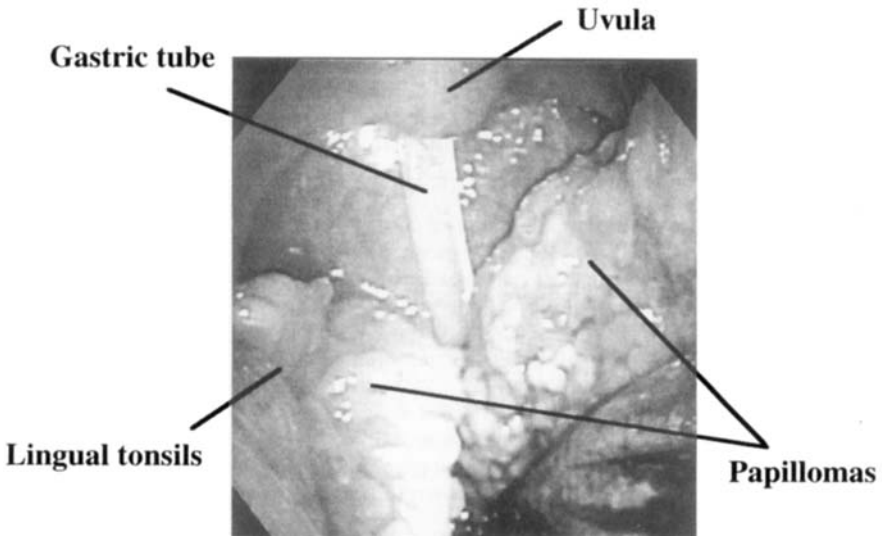

Fig. 1. Extensive confluent papillomatous lesions on the root of the tongue and hypopharynx as observed on fiberoptic laryngoscopy.

papilloma-like lesions. Two blind oral intubations were attempted without success. Bleeding from the papillomas was easily induced during this procedure. Thereafter, complete airway obstruction developed. A fiberoptic intubation attempt was also unsuccessful because of the nature of the papillomas and bloody secretions that obscured the visual field. The patient's oxyhemoglobin saturation had decreased to < 40%, and urgent tracheostomy was performed. A 6.0-mm ID endotracheal tube was inserted *via* the tracheostomy incision, resulting in an increase in oxyhemoglobin saturation to 100% with an inspired oxygen fraction of 1.0. The patient was delivered to the intensive care unit and treated under mild hypothermia (34°C) for 3 days to prevent brain damage from hypoxia. This resulted in full recovery of consciousness without any neurologic complications. Fiberoptic laryngoscopy performed in the intensive care unit showed extensive confluent papillomatous lesions on the root of the tongue and hypopharynx (fig. 1). However, the laryngoscope was not able to be advanced near the larynx because the hypopharynx was occupied by the extended papillomas. Laryngoscopy performed 5 days later showed that the papillomas on the lingual tonsils extended posteriorly beyond the tip of the epiglottis and filled the valleculae (fig. 2). Barium fluoroscopic examination also showed multiple papillomas on the hypopharynx and the esophagus (fig. 3). Eight days later, right mastectomy, left thyroidectomy, and resection of the papillomatous lesions on the lingual tonsils were performed. The pathology report showed that the lesions were hamartoma. Postoperative endoscopic examination showed a large number of polyps in the esophagus,

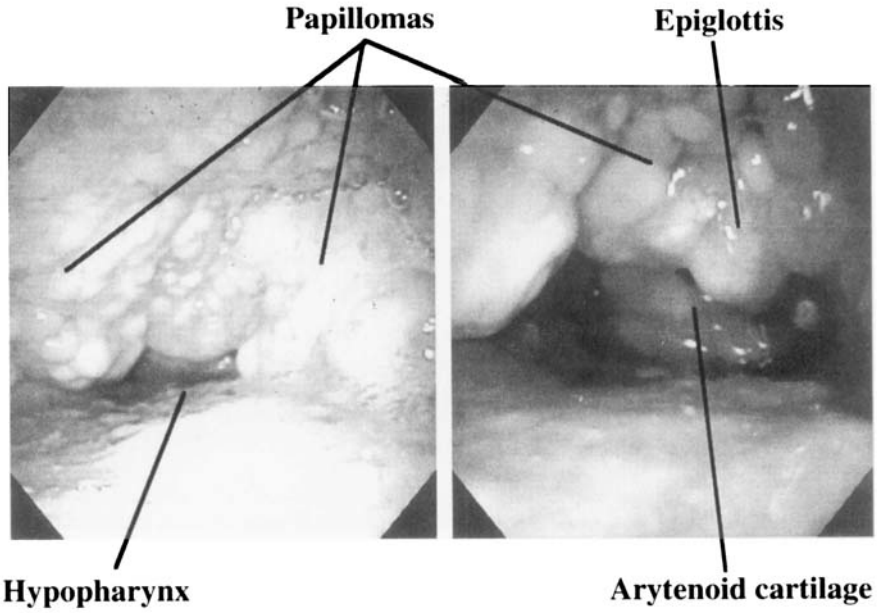

Fig. 2. The papillomas on the lingual tonsils extended posteriorly beyond the tip of the epiglottis and filled the valleculae (*left* and *right*).

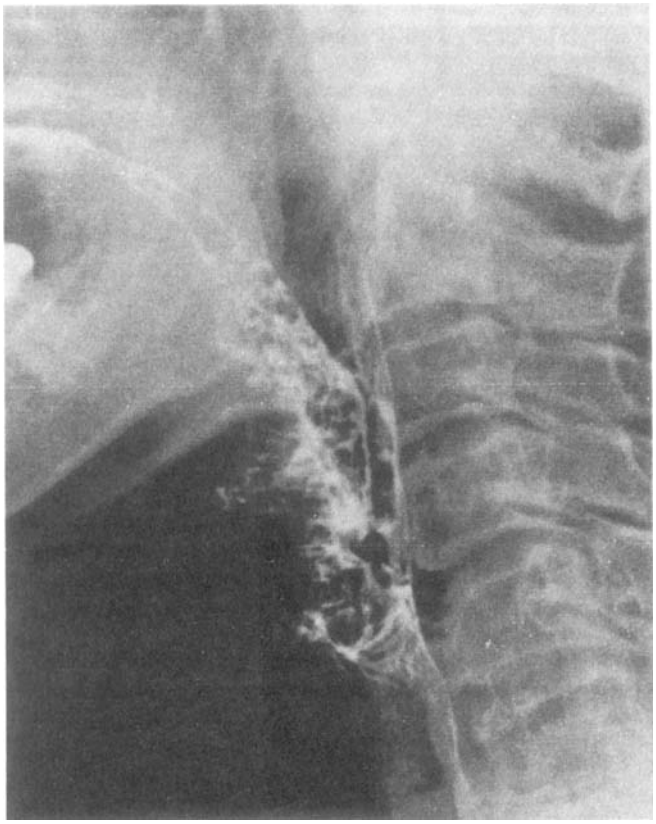

Fig. 3. Multiple papillomas on the hypopharynx and the esophagus in barium fluoroscopy.

hyperplastic polyposis and tubular adenomas in the stomach, and a hamartoma in the sigmoid colon. In addition, the patient's daughter and son also presented with some papillomas on the root of the tongue. On the basis of these results, the patient was diagnosed as having Cowden's disease.

Discussion

This is the first reported case of critical airway obstruction that developed during induction of anesthesia, was caused by oropharyngeal and laryngeal papillomas, and was later proved to be a rare condition of Cowden's disease.

Cowden's disease is a rare inherited genodermatosis, the major features of which are multiple hamartomatous papules and nodules on the skin and oropharyngeal, laryngeal, and gastrointestinal mucosa, together with thyroid and breast anomalies and polyposis of the gastrointestinal tract.[1,2] Since the first report of this disease more than 35 yr ago,[1] < 200 cases have been documented. In > 80% of reported cases, papular or verrucous oral lesions on the lips, tongue, gingiva, edentulous alveolus, buccal mucosae, palate, or tonsillar fossae have been found.[3-5] However, there has been no report of critical airway obstruction caused by pharyngeal and laryngeal lesions occurring during induction of anesthesia as a result of Cowden's disease.

In the present case, Cowden's disease was not diagnosed preoperatively, although the patient had a thyroid tumor and breast cancer. Ovassapian[6] described three cases of difficult intubations caused by a mass at the base of the tongue. He concluded that such cases cannot be identified by routine external physical examination of the airway and that anesthesiologists will continue to be surprised by unexpected difficult airways and difficult intubations. In fact, the lingual tonsils, being located on the posterior one third of the tongue in the hypopharynx, would not usually be visible by simple visual inspection of the oropharynx without the use of a mirror or endoscope. However, in the present case, we did a less-than-thorough airway examination preoperatively. If we did as thorough an evaluation of the preoperative intra-oral examination and on the computed tomography scans as was possible, it would be possible to predict the patient's oropharyngeal condition.

A difficult airway is particularly challenging for anesthesiologists. In the present case, we performed a fiberscopic trial in an attempt to achieve intubation within a limited period. However, our decision was inappropriate because we had known about the lack of space in the hypopharynx in which to advance the fiberscope and the bloody secretions that obscured the visual field. Laryngeal mask airway, Combitube (Kendall, Argyle, NY), or transtracheal jet ventilation might be of value to try to establish ventilation, but we do not know if the laryngeal mask airway would work in this particular case. We decided to perform tracheostomy when the patient's oxyhemoglobin saturation had become critical. However, we should have proceeded more rapidly to a tracheostomy once we encountered trouble.

In summary, multiple papillomas on the lingual tonsils, epiglottis, and the surrounding structure that occupied the hypopharynx caused difficulty in airway management and prevented tracheal intubation during induction of anesthesia. This is the first reported case of difficulty with airway management in a patient with the Cowden's disease.

References

1. Lloyd KM, Dennis M: Cowden's syndrome: A possible new symposium complex with multiple system involvement. Ann Intern Med 1963; 58:136-42

2. Weary PE, Gorlin RJ, Gentry WC Jr, Comer JE, Greer KE: Multiple hamartoma syndrome (Cowden's disease). Arch Derm 1972; 106:682-90

3. Devlin MF, Barrie R, Ward-Booth RP: Cowden's disease: A rare but important manifestation of oral papillomatosis. Br J Oral Maxillofac Surg 1992; 30:335-6

4. Smid L, Zargi M: Cowden's disease: Its importance for otolaryngologists. J Laryngol Otol 1993; 107:1063-5

5. Porter S, Cawson R, Scully C, Eveson J: Multiple hamartoma syndrome presenting with oral lesions. Oral Surg Oral Med Oral Pathol 1996; 82:295-301

6. Ovassapian A: Fiberoptic Airway Endoscopy in Anesthesia and Critical Care, 1st Edition. New York, Raven, 1990, pp 149-62

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Failure of a Patient-controlled Analgesia Pump in a Hyperbaric Environment

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TECHNOLOGIC progress has led to the development of simple and reliable epidural patient-controlled analgesia (PCA) pumps, resulting in their widespread use with the consequent appearance of new challenges. We present a case of PCA pump failure inside a hyperbaric chamber.

Case Report

A 52-yr-old obese woman with renal failure, who had been receiving hemodialysis for 8 yr and had refused a kidney transplant, developed necrotic lesions on the wall of the lower abdomen as a result of vascular calcification secondary to hyperparathyroidism. Anaerobic infection of the affected area necessitated antibiotics, daily debridement, and hyperbaric oxygen therapy (HBO) in a multiplace chamber (TEDSA, Cartagena, Spain). Pain relief was provided by epidural PCA with a CADD-PCA 5200 PXC pump (Pharmacia Deltec Inc., St Paul, MN). However, during the first HBO treatment period, the warning alarm on the PCA pump sounded. Consequently, the anesthesiologist who initiated the epidural PCA treatment also accompanied the patient during the second HBO session, during which the alarm again sounded, and we were able to confirm the malfunction of the pump.

To verify the functioning of this particular PCA pump in a hyperbaric environment, we conducted a series of 20 pressurizations with the same pump in the same HBO conditions, *i.e.*, within a multiplace chamber reaching 2.5 absolute atmospheres (ATA), a pressure equivalent to a depth of 49.5 ft of sea water. The pump behaved the same each time. When 2 ATA was reached, the alarm rang, the liquid crystal display screen started blinking, and the pump stopped. On reducing the pressure to 1.9 ATA, the pump resumed working without needing to be reprogrammed. At 2 ATA, it was impossible to reprogram the pump.

To determine if this problem was related just to the pump in question, we submitted it plus another two later models of the pump (CADD-PCA 5800) from the same manufacturer to HBO. They were each programmed identically to release 0.5 cm^3/min of physiologic saline solution. The results show that although there were no marked differences in flow (the readout on the pumps does not show decimals, only cubic centimeters released) there was a difference in the pressure at which the pumps stopped working and then resumed again (table 1).

Discussion

Hyperbaric oxygen therapy provides oxygen at pressures > 1 ATA.[1] Its therapeutic and iatrogenic effects are a result of the high concentration of oxygen and the increase in barometric pressure. HBO is used with surgery and antibiotic therapy to treat necrotizing lesions.[1,2] However, reasons for alterations in the normal functioning of PCA systems within a hyperbaric environment include the following[3]: (1) explosion (the pump motor contains brushes that produce sparks); (2) battery leakage caused by the high pressure; (3) collapse of the protective face plate covering the controls caused by high pressure; (4) entry of air into the reservoir or tubes (this is especially dangerous in intravenous PCA).

In this case, and after the experiment, we believe that

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Key words: Hyperbaric oxygen; drug delivery pump.