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James W. Buchanan
University of Pennsylvania, jwb@vet.upenn.edu

James H. Anderson

Robert I. White

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Abstract
Balloon valvuloplasty (BV) is currently the treatment of choice for pulmonic stenosis in humans and dogs. Before permission was obtained to attempt the 1st BV in a child in 1982, the safety and efficacy of the procedure were tested in 1980 in an English Bulldog with spontaneous pulmonic stenosis. A fatal outcome would have caused indefinite postponement of BV in human patients, a procedure that currently benefits over 25,000 patients a year worldwide. This article describes the initial test procedure and its fortunate outcome in spite of unrecognized coronary anomalies in the bulldog. A small balloon was used in the test procedure, and fatal disruption of the anomalous left coronary artery (CA) did not occur as it has in several bulldogs since that time.

Keywords
Coronary anomaly, English Bulldog, Pulmonic stenosis, Single coronary artery

Disciplines
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Brief Communication

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The 1st Balloon Valvuloplasty: An Historical Note

James W. Buchanan, James H. Anderson, and Robert I. White

Balloon valvuloplasty (BV) is currently the treatment of choice for pulmonic stenosis in humans and dogs. Before permission was obtained to attempt the 1st BV in a child in 1982, the safety and efficacy of the procedure were tested in 1980 in an English Bulldog with spontaneous pulmonic stenosis. A fatal outcome would have caused indefinite postponement of BV in human patients, a procedure that currently benefits over 25,000 patients a year worldwide. This article describes the initial test procedure and its fortunate outcome in spite of unrecognized coronary anomalies in the bulldog. A small balloon was used in the test procedure, and fatal disruption of the anomalous left coronary artery (CA) did not occur as it has in several bulldogs since that time.

Key words: Coronary anomaly; English Bulldog; Pulmonic stenosis; Single coronary artery.

Balloon valvuloplasty (BV) has been a common procedure since the 1st report in a child with pulmonic stenosis in 1982.[1] It is currently the treatment of choice in patients with valvular pulmonic stenosis and frequently is used in patients with mitral, tricuspid, or aortic stenosis.[2] It is estimated that over 25,000 valvuloplasties now are performed annually worldwide.

The procedure was developed initially in dogs with surgically induced pulmonary artery stenosis at Johns Hopkins University, but approval to try it in human patients was not obtained from the institutional review board until the safety of the procedure was established in an animal with spontaneous pulmonic stenosis and marked right ventricular (RV) hypertrophy. Accordingly, a young English Bulldog with congenital pulmonic stenosis was transferred from the veterinary hospital of the University of Pennsylvania to Johns Hopkins Hospital, where the dog was treated successfully by BV on October 7, 1980.[3] Subsequent coronary artery (CA) findings in this dog,[4] and other bulldogs with pulmonic stenosis in which BV proved fatal,[5] showed how fortunate it was that this 1st ''trial balloon'' procedure did not cause indefinite postponement of the use of BV in human patients.

Case Report

A 14-kg, 5-month-old male English Bulldog was referred to the Comparative Cardiovascular Studies Unit at the University of Pennsylvania because of a heart murmur and characteristic signs of pulmonic stenosis. Cardiac catheterization under general anesthesia confirmed the diagnosis of valvular pulmonic stenosis with RV systolic pressure of 70 mmHg and a 55-mm gradient. Angiocardiography showed a centrally located, 3-mm-diameter pulmonary valve jet and a pulmonary sinus diameter of 18 mm. An experimental iliac angioplasty catheter with a polyethylene balloon expandable to a diameter of 10 mm was positioned across the pulmonary valve and inflated. A narrowing of the balloon appeared at the level of the pulmonary valve when the balloon was filled with radiopaque contrast material. When the balloon was pressurized, the narrow segment expanded to a diameter equal to the diameter of the rest of the balloon. Femoral artery systolic pressure rose from 65 to 67 mmHg after the 1st dilation but increased to 87 mmHg after a 2nd dilation. RV systolic pressure was 40 mmHg immediately after valvuloplasty but returned to the prevalvuloplasty value of 70 mmHg 30 minutes later. The dog recovered uneventfully from the procedure and remained clinically normal for 2 years, then developed exercise intolerance and ascites. The ascites became refractory to medical therapy, so the dog was admitted for surgery at 3 years of age. The dog still had evidence of pulmonic stenosis in addition to atrial fibrillation and marked ascites. Angiocardiography showed a 6-mm-diameter pulmonary valve jet and a pulmonary sinus diameter of 18 mm. At surgery, an oval segment of autologous pericardium was partially sutured over the RV outflow tract and main pulmonary artery. During caval occlusion, a transmural incision was made under the pericardial patchgraft extending through the root of the main pulmonary artery and into the RV outflow tract. The final sutures of the graft were then tied, circulation was reestablished, and blood pressure returned to normal.

After the thorax was closed, systemic blood pressure began to fall. Despite various supportive measures, the dog developed ventricular fibrillation approximately 1 hour after the ventriculotomy under the patch graft. Transient external defibrillation was accomplished, but ventricular fibrillation recurred repeatedly, so the chest was reopened to permit direct cardiac massage. The right ventricle was fibrillating, but the left ventricle was cyanotic and asystolic. Direct cardiac massage, defibrillation, and the usual drugs for cardiac arrest were given, but the heart was unresponsive, and further attempts at resuscitation were abandoned. Examination of the heart revealed a large, severed CA that coursed around the root of the pulmonary artery at the juncture of the RV and main pulmonary artery. Later, dissection showed that the severed CA was an aberrant circumpulmonary left main CA originating from a single right CA.[4] The aberrant vessel was positioned deeply in the epicardial fat immediately adjacent to the valve annulus, where it compressed the root of the pulmonary trunk. The 3 pulmonary valve leaflets were slightly thickened and squeezed together, but there was no commissural fusion.

Discussion

Balloon angioplasty to treat coronary and other vascular diseases was an established procedure in 1980, but use of a balloon to dilate a stenotic heart valve raised concern over patient safety because it meant sudden increases in RV pressure and abrupt stopping of circulation. Although sudden obstruction of the circulation had no ill effects in ex-

From the Section of Cardiology, School of Veterinary Medicine, University of Pennsylvania, Philadelphia, PA (Buchanan); the Department of Radiology, Johns Hopkins University, Baltimore, MD (Anderson); and the Department of Diagnostic Radiology, Yale University, New Haven, CT (White).

Reprint requests: James W. Buchanan, DVM, M Med Sci, School of Veterinary Medicine, University of Pennsylvania, 3850 Spruce Street, Philadelphia, PA 19104; e-mail: jwb@vet.upenn.edu.

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perimental dogs, it was desirable to test BV on an animal with spontaneous pulmonic stenosis and marked RV hypertrophy.[3] The selection of a bulldog as a test animal was coincidental but could have been fatal and delayed approval of human BV for years. The dog happened to be the next veterinary patient with pulmonic stenosis and a cooperative owner. Bulldogs were known to have a predisposition for pulmonic stenosis,[6] but we were unaware of the high frequency of coronary anomalies in this breed.[7] If this fact had been known, a bulldog would not have been chosen to test BV safety.

Retrospective examination of the preoperative angiocardiogram in the test dog revealed faint evidence of an extra bulge on the cranial aspect of the aortic root similar to the ''aortic root sign'' in some human patients with single right CA.[8] Because the only previously reported case of single right CA and pulmonic stenosis also was a bulldog,[9] coronary arteries were studied carefully in subsequent bulldogs with pulmonic stenosis. Single right CA and circumpulmonary left main CA were found in 7 out of 8 bulldogs at this institution.[10] Histologic studies in a stillborn bulldog indicated abnormal development of the left main CA as the cause of single right coronary and pulmonic stenosis.[11]

BV is the recommended treatment for all dogs with discrete pulmonic stenosis, except when single right CA is present and BV may be fatal. In one series of 35 dogs undergoing BV, only 2 dogs died during the procedure, and both were bulldogs with single right CA. Both were found to have disruption or avulsion of the aberrant circumpulmonary left main CA at postmortem examination.[5] The procedure probably was not fatal in the test bulldog because the balloon catheter available in 1980 was a slightly oversized iliac angioplasty balloon about 10 mm in diameter. Manufacturing techniques were just being developed to make larger balloons with rapid inflate-deflate cycles.[12] Current recommendations for BV are to use a balloon diameter 120% of the diameter of the pulmonary valve annulus. Because the test bulldog had a pulmonary sinus diameter of 18 mm, a 20-mm balloon would be selected for BV in this dog today, and use of a 20-mm balloon probably would have been fatal in the test bulldog.

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