The reported cases and our cases did not show classic clinical features of WMG. Moreover the shadows seen in chest x-ray films varied in appearance presenting diffuse infiltration, nodular lesion, hilar enlargement or pleural effusion, and the respiratory symptoms were also not diagnostic. Therefore, it was very difficult to determine clinically whether the disease was pulmonary WMG or some other lung disease in which IgM increased secondarily. Moreover, it is very difficult to distinguish reactive lymphoid lesions from neoplastic ones of the lung, especially in lesions composed predominantly of small lymphocytes like those in WMG. So we consider that immunologic tests for monoclonal IgM are the most important and best ways to establish a diagnosis of pulmonary WMG. In the present patients, we demonstrated cytoplasmic monoclonal IgM in tumor cells by the PAP method and surface monoclonal IgM in lymphoid cells obtained from bronchoalveolar lavage in case 1. In case 2, we did not obtain a sufficiently large specimen for diagnosis by transbronchial lung biopsy, but we obtained sufficient tissue by open lung biopsy. We demonstrated monoclonal cytoplasmic IgM by the PAP method and monoclonal surface IgM in a cell suspension obtained from the right lung. We established the diagnosis from these immunologic data. Thus, we wish to stress the importance of immunologic studies in making a diagnosis of pulmonary WMG, since histologic examination of biopsy specimens does not often show specific changes and there are few specific clinical features of the disease.

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Hypoplastic Coronary Arteries and High Takeoff Position of the Right Coronary Ostium*

A Fatal Combination of Congenital Coronary Artery Anomalies in an Amateur Athlete

David M. Menke, M.D.; Bruce F. Waller, M.D., F.C.C.P., and John E. Pless, M.D.

This report describes a previously unrecognized combination of congenital coronary artery abnormalities in the heart of a 30-year-old amateur athlete who died suddenly during a basketball game. Both right and left circumflex coronary arteries were half of their normal length (hypoplastic) decreasing posterior ventricular myocardial perfusion. In addition, the right coronary ostium rose 5 mm above the sinotubular junction (high takeoff position), which also contributed to decreased right coronary artery perfusion. This combination of congenital coronary arterial lesions should be added to the list of structural cardiac defects associated with exercise-related sudden death.

With increased participation in exercise have come several reports of sudden death in conditioned subjects dying during or shortly after vigorous exercise.4,4 Necropsy examination in these subjects has disclosed major structural cardiovascular abnormalities in the vast majority.4 In amateur or professional athletes 30 years of age or younger, congenital coronary arterial anomalies or hypertrophic cardiomyopathy account for most of the exercise-related sudden deaths.2,4 In the report which follows, we describe a previously unreported combination of congenital coronary arterial anomalies in a young amateur athlete who died suddenly while playing basketball.

CASE REPORT

A 30-year-old white man had been asymptomatic all of his life until he suddenly collapsed and died. During a basketball game, he suddenly fell to the floor with seizure-like activity. The paramedic rescue team found the subject in ventricular fibrillation, but attempts at resuscitation were unsuccessful. About three years before death, familial type 2 hyperlipoproteinemia was diagnosed on a routine physical examination with total serum cholesterol level of 454 mg/dl. During the same examination, resting electrocardiogram results was normal. The patient was a slender, well-

*From the Divisions of Cardiovascular and Forensic Pathology, Department of Pathology, and the Department of Medicine (Cardiology), Indiana University School of Medicine, the Department of Pathology, Methodist Hospital; and theKrannert Institute of Cardiology, Indianapolis.
Reprint requests: Dr. Waller, University Hospital, N-340, 926 Michigan Avenue, Indianapolis 46223

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conditioned amateur athlete who exercised several times per week playing vigorous games of basketball and racquetball.

At necropsy, the heart weighed 350 g. Examination of the major epicardial coronary arteries disclosed hypoplastic right and left circumflex arteries with total lengths measuring 4.5 and 2.5 cm respectively (normal lengths: right = 12, left circumflex = 5 cm) (Fig 1). The left main and anterior descending coronary arteries had normal lengths. Serial sectioning of the entire coronary tree into 5-mm long segments disclosed mild and focal proximal left anterior descending coronary narrowing by atherosclerotic plaques. The maximal luminal cross-sectional area narrowing was 26 to 50 percent. The remaining segments of anterior descending coronary artery were normal (Fig 1). The ascending aorta had extensive lipid deposits characteristic of type 2 hyperlipoproteinemia (Fig 2). The right coronary artery ostium was hypoplastic and arose 5 mm above the sinotubular junction (high takeoff position) (Fig 1, 2). Transverse sectioning of the cardiac ventricles from apex to base disclosed a small focus of transmural posterior myocardial scar involving left and right ventricular free walls (Fig 2). The remainder of the general necropsy examination revealed no other abnormality.

**Discussion**

Hypoplastic right and/or left coronary arteries have been observed previously in hearts of two athletes with exercise-related sudden death. The proposed mechanism of death in this condition is ischemia from hypoperfused myocardium. High takeoff position of a coronary ostium, defined as location of the ostium 5 mm or more above the aortic sinotubular junction, has been postulated as a cause of sudden death in a nonathlete. The proposed mechanism of death with this lesion also involves myocardial ischemia resulting from coronary hypoperfusion. Normally, the coronary ostia are located within the sinuses of Valsalva permitting maximal opportunity for coronary diastolic filling. Location of coronary ostia in the tubular portion of aorta, ie, above the sinuses of Valsalva, may decrease coronary perfusion. The adverse effects of the combination of these congenital coronary anomalies—high takeoff coronary ostium and hypoplastic coronary arteries—appear to have been additive in the above-described athlete. During exercise (or any cause of tachycardia), the length of mechanical and electrical diastole shortens. Reducing the diastolic period further enhances the adverse filling properties of high takeoff and hypoplastic coronary arteries. Morphologic evidence of chronic myocardial ischemia was present in our patient in the distribution of the right coronary artery (posterior left and right ventricular scarring).

The combination of high takeoff coronary ostium and coro-
nary hypoplasia should be added to the list of congenital coronary arterial anomalies associated with exercise-related sudden death.

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Surgical Management of Recurrent Spontaneous Pneumothorax during Pregnancy*
Sonny S. Dhalla, M.D.; and John M. Teskey, M.D., F.C.C.P.

We report three cases of recurrent spontaneous pneumothorax associated with pregnancy. All three cases had apical bullaeescence during their pregnancies.

Spontaneous pneumothorax in pregnancy is said to be a rare condition. There have been only 12 cases reported in the English literature in the last three decades.14 We present three case reports of spontaneous pneumothorax associated with pregnancy. All three patients were managed by operation during pregnancy which, to our knowledge, has not previously been reported.

CASE REPORTS

CASE 1

A 25-year-old Caucasian Gp3/p1, previously healthy with last normal menstrual period (LNMP) December 11, 1984, Breviindex positive on January 18, 1984, was admitted at four weeks’ gestation with a moderate-sized right spontaneous pneumothorax. This was treated by chest tube drainage with complete resolution. At five weeks’ gestation, the patient developed a moderate left spontaneous pneumothorax treated successfully by chest tube drainage. Left pneumothorax recurred at six weeks’ gestation and again was treated by chest tube drainage, but failed to resolve completely.

At eight weeks’ gestation, the patient electively underwent bilateral apical bullectomy and pleural abrasion, through midline incision, under general anesthesia. There was no postoperative complication.

At 32 weeks’ gestation, the patient developed mild cholestasis of pregnancy. At term, a healthy normal male infant (weight 3,175 g, with Apgar scores of 7 and 9 at one and five minutes respectively) was delivered with Simpson forces.

Case 2

A 25-year-old Caucasian Gp3/p1 previously healthy with LNMP September 26, 1981, Breviindex positive November 9, 1981, presented at eight weeks’ gestation with a small right pneumothorax which was treated with rest and observation. At 11 weeks’ gestation, a small recurrent right spontaneous pneumothorax was treated with needle aspiration. At 13 weeks’ gestation, she had further recurrence of the right spontaneous pneumothorax of moderate size, treated with chest tube drainage successfully. At 18 weeks’ gestation, a further recurrence of the right pneumothorax was treated by needle aspiration.

At 22 weeks’ gestation, the patient electively underwent right transaxillary apical wedge resection and apical pleurectomy under general anaesthesia. There were no postoperative complications.

At term, a healthy normal female infant (weight 3,010 g, with Apgar scores of 1 and 9 at one and five minutes respectively) was delivered with Kielland forces. A subsequent pregnancy two years later was uneventful.

Case 3

A 28-year-old Caucasian Gp3/p1 with previous history of left spontaneous pneumothorax at age 17 and right spontaneous pneumothorax at age 23, had a small spontaneous right pneumothorax in March, 1960, treated with observation. Her LNMP was May 28, 1980. At 14 weeks’ gestation, she was admitted with a small right pneumothorax and treated with observation.

At 24 weeks’ gestation, the patient electively underwent right transaxillary apical wedge resection and apical pleurectomy under general anesthesia. The fetus was monitored intraoperatively by ultrasound cardiocotometry. There was no postoperative complication.

She delivered, at term, a healthy normal male infant (weight 4,190 gm, with Apgar scores of 9 and 9 at one and five minutes respectively) with Simpson forces.

DISCUSSION

Spontaneous pneumothorax in pregnancy is said to be extremely rare, as witnessed by the paucity of reported cases in the English literature.4 However, we have seen three cases in the last four years at our hospital. Many authors14 have suggested that underreporting is responsible for the apparent infrequency of this condition, and our experience strengthens this viewpoint.

Of the 12 reported cases, only two occurred during the first trimester,13 and the rest in the last trimester, mostly at term. At our hospital, all three cases were seen in early pregnancy (4, 8, and 14 weeks). Two had no previous history of pneumothorax. All had recurrent episodes during their pregnancy prior to surgical treatment.

In a previously healthy young population, spontaneous pneumothorax is almost always related to the presence of small apical blebs or bullae without other significant pulmonary disease. The risk of recurrence following the initial episodes is almost 30 percent,14 and the risk thereafter is about 50 percent, with overall incidence of recurrence about