The resolution of hypertrophic pulmonary osteoarthropathy following treatment of subacute infective endocarditis

L. M. SHAPIRO  
M.B., Ch.B., M.R.C.P.  

J. MACKINNON  
M.D., F.R.C.P.

The Department of Cardiology, Dudley Road Hospital, Birmingham B18 7QH

Summary
A patient with a corrected coarctation of the aorta subsequently developed infective endocarditis on a congenital bicuspid aortic valve. Resolution of the radiographic bone changes of hypertrophic pulmonary osteoarthropathy was demonstrated following treatment of the infection.

Introduction
Finger clubbing is a common feature of subacute infective endocarditis (Hayward, 1960) but it is rarely associated with the painful arthropathy and periostitis of hypertrophic pulmonary osteoarthropathy (HPOA).

A case is reported which demonstrates the resolution of the periosteal new bone formation after treatment of the endocarditis.

Case report
A Sikh born in India in 1936 came to England in 1959. At a pre-employment medical examination in 1960 he was noted to have a BP of 140/95 mmHg in both arms, small volume femoral pulses and a precordial systolic thrill. A chest radiograph showed an abnormal aortic knuckle, rib notching and minimal cardiomegaly. A diagnosis of coarctation of the aorta and congenital aortic stenosis was made. An aortogram in 1962 confirmed the coarctation distal to the left subclavian artery and a systolic pressure gradient of 37 mmHg across the aortic valve was found. In 1964 the coarctation was resected and the BP fell to normal.

In October 1978 he suffered a right hemiparesis which resolved after 3 days, leaving residual dysphasia and was admitted to Dudley Road Hospital. He have a history of night sweats for 2 months and 4 rigors. His temperature was 38.5°C. Finger-clubbing and splinter haemorrhages were noted. The pulse was regular at 56/min, the BP was 100/85 mmHg in both arms and there was a basal systolic thrill and murmur. The spleen was just palpable and there was microscopic haematuria. His dental condition was good and he gave no history of joint pain or swelling. An ECG showed left ventricular hypertrophy with the rhythm alternating between 2:1 heart block (Mobitz II) and complete heart block. The cardiothoracic ratio was increased on the chest X-ray compared with a previous film. An echocardiogram (Fig. 1) demonstrated vegetations on a bicuspid aortic valve; left ventricular function was good. X-ray of the distal tibia showed periosteal new bone formation (Fig. 2a) as did the distal radii and femora. No infecting organism was isolated but a clinical diagnosis of subacute infective endocarditis was made and he was treated with i.v. benzyl penicillin and gentamicin, the doses of which were controlled by serum levels.

He remained pyrexial and developed aortic incompetence which progressed in severity throughout early December and culminated in left ventricular failure that was unresponsive to medical management.

At thoracotomy the diagnosis of endocarditis was confirmed and the bicuspid aortic valve was found to be detached at the anterior commissure where it had been undermined by an abscess extending to the region of the atrio-ventricular node. The diseased valve was replaced by a Björk-Shiley 21 mm prosthesis and a permanent epicardial demand pacemaker was implanted. Culture of the resected aortic valve grew no organisms. Following prolonged convalescence on antibiotics he made a full recovery and returned to work. At review in March 1979 finger clubbing was no longer present, and an X-ray of the distal tibia (Fig. 2b) showed total resolution of the subperiosteal bone changes.

Discussion
This case demonstrates the natural history of a patient with a corrected coarctation of the aorta and an associated bicuspid aortic valve. The bicuspid valve was the site of subacute infective endocarditis with resultant myocardial abscess formation causing heart block by involvement of the conducting tissue, valvular disruption and left ventricular failure. The infection was eradicated by aortic valve replacement and a prolonged course of antibiotics,
Case reports

Fig. 1. Aortic valve echocardiogram showing vegetations on a bicuspid valve.

Fig. 2. Radiograph of distal tibia (a) before, (b) after treatment.
even though it was not possible to identify the causative organism.

The HPOA described in this case was atypical as there was no painful arthropathy (Holling and Brodey, 1961). In most of the published reports HPOA is a complication of intrathoracic tumours or sepsis and the authors were unable to find a report of this condition associated with subacute infective endocarditis. Finger clubbing is known to resolve following excision of thoracic tumours or following withdrawal of a causative agent (Silk, Gibson and Murray, 1975). It also resolves with treatment of subacute infective endocarditis and, in the case presented, regression of the abnormal periosteal bone was also noted.

The aetiology of HPOA is not known but there are 3 theories. The resolution noted with vagal nerve section and thoracotomy is given as evidence that the vagus or intercostal nerves are an afferent pathway from the pulmonary focus. There is no described efferent pathway (Flavell, 1956). Circulating agents capable of inducing HPOA that are normally degraded in the pulmonary circulation are proposed to have a peripheral action because of the lack of inactivation in this condition or to pass through arterio-venous connections (Marie, 1890). The final theory which also has little experimental support is that it is due to increased peripheral blood flow (Holling and Brodey, 1961). It is very difficult to apply any of these theories to endocarditis, as there is no specific pulmonary lesion or arterio-venous communications and the cardiac output is likely to be normal or reduced.

References

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L. M. Shapiro and J. Mackinnon

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