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Ankylosing hyperostosis in American Blacks: a Longitudinal study

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SUMMARY We studied 63 consecutive patients with ankylosing hyperostosis to investigate any possible difference in clinical or roentgenographic features between whites and blacks. Our data suggest that (a) the disease may not be less common among blacks, (b) presence of extraspinal hyperostosis in this disease may be more frequent in black patients, and (c) the known male preponderance of the disease among whites may not occur among blacks.

Key words: Ankylosing Hyperostosis, Racial Incidence, Racial Characteristics, Ankylosing Spondylitis.

INTRODUCTION

Ankylosing hyperostosis of the spine (Forestier's disease) is a skeletal disorder characterized by excessive bone formation (hyperostosis) at skeletal sites where tendons and ligaments attach to bones; the most commonly involved sites are spine, ischial tuberosity, femoral trochanter, tibial tubercle and calcaneum (1,2,3). The extraspinal hyperostosis may occur without appreciable spinal changes; therefore, a new name — diffuse idiopathic skeletal hyperostosis (DISH) — has been proposed for this disorder (4).

In a series of patients, Resnick et al. (2,3) observed that the disease occurred mainly in whites. They were therefore unable to make any inference regarding possible racial influence on disease characteristics. We undertook the present prospective study to investigate any possible clinical or radiological differences between whites and blacks in the occurrence of this disease.

MATERIALS AND METHODS

We longitudinally studied 63 consecutive patients with ankylosing hyperostosis seen at Cleveland Metropolitan General Hospital. The diagnosis was based on published crite-

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ria (1-4). All patients had ossification of the anterior longitudinal ligament of the spine; those with ankylosing spondylitis, Reiter's syndrome, parathyroid disease, psoriasis, fluorosis, hypervitaminosis A, or Paget's disease were excluded from this study. The extraspinal localization of hyperostosis was also investigated by studying the roentgenographs of the pelvis, femurs, knees, shoulders, heels, elbows, forearms, hands and feet. The total patient population served by our hospital is almost equally divided between the two racial groups.

RESULTS

Table 1 lists the age, sex and race distribution of these patients. There were 30 blacks and 33 whites and their mean age was 66.75 years. A significant difference was noted in the sex distribution in the two racial groups; a male predominance was seen in whites but not in blacks ($X^2 = 5.65$, $p < 0.02$). By definition, all patients had the characteristic ossification of the anterior longitudinal spinal ligaments (fig. 1 and 2).

Extraspinal skeletal hyperostosis was studied in 33 patients (16 black, 17 white) and was noted in 87.5 % of blacks; and 35.3 % of whites (X^2 with Yates correction $= 6.72$; $p < 0.01$) (Table 2). The HLA-B27 test result was negative in 4 patients who were so tested.

DISCUSSION

Resnick et al (2,3) studied ankylosing hyperostosis primarily in whites and were thus unable to establish the racial incidence of the disease. In a hospital population which includes approximately an equal number of whites and blacks, we found that the incidence of the disease is similar in whites and blacks. Among our white patients, the disease was more common in males, as has been noted by Resnick (4). This male preponderance of the disease, however, was not observed among our black patients.

When the roentgenographic features of the disease were compared in the two racial groups, extraspinal skeletal hyperostosis was observed to be significantly more common among black patients as compared to whites. Such a difference has not previously been reported.

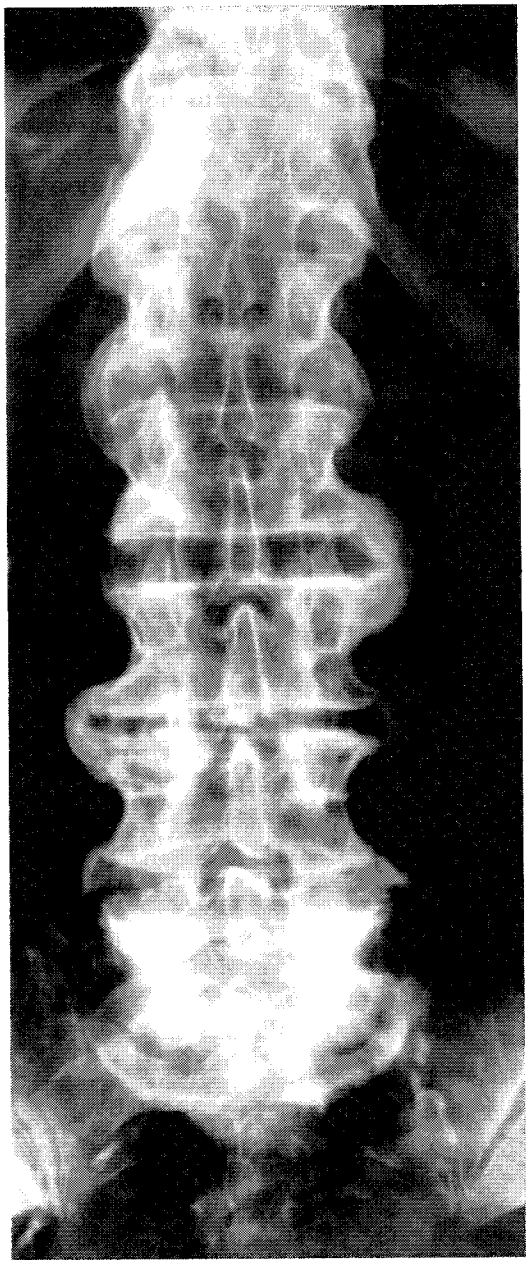
Shapiro et al. (6) reported an association between DISH and HLA-B27. This could not be confirmed by other investigators

Table I: Age, sex and race distribution of patients with ankylosing hyperostosis

Race	Number of patients	Males	Females	Age	
				Mean	Median
Black	30	14	16	67	71
White	33	25	8	66.5	66
TOTAL	63	39	24		

Table II: Extraspinal skeletal hyperostosis in the two racial groups

Race	Number of Patients studied	Number of patients with extraspinal skeletal hyperostosis	Significance
Black	16	14 (87.5 %)	
White	17	6 (35.3 %)	$p < 0.01$



FIGURES 1 and 2: Antero-posterior and lateral radiographs of the lumbar spine of a 71-year old black male with ankylosing hyperostosis, showing typical flowing type of ossification at the antero-lateral aspect of vertebral bodies, resulting in bumpy contour of the spine.

(5,7,8). All 4 of our patients that were so tested lacked B27. We had earlier studied the frequency of B27 in 10 other patients with DISH, equally divided between whites and blacks; none was found to possess B27 (unpublished observation).

The spinal ossification of ankylosing hyperostosis may be confused with ankylosing spondylitis, a disease that shows strong association with HLA-B27. However, there are certain features that help differentiate these two conditions. Ankylosing spondylitis usually tends to affect younger adults rather than the middle-aged and the elderly, and is associated with more slender vertical bony bridging (syndesmophytes) between vertebral bodies that can be differentiated from the anterior longitudinal ligament ossification of ankylosing hyperostosis. Furthermore, the apophyseal and sacroiliac joint abnormalities characteristic of ankylosing spondylitis are absent in ankylosing hyperostosis. However, in some patients such distinctions may be difficult. Goldin and Bleustone (9) have suggested that HLA-B27 typing can be of clinical help in such a situation, since ankylosing spondylitis shows a strong association with B27, while there is no such association with ankylosing hyperostosis.

In conclusion, our data show that ankylosig hyperostosis is not less common among blacks. The extraspinal hyperostosis may be more frequent in black patients, and the male preponderance of the disease seen among whites may not be present among blacks.

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