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ANKYLOSING SPINAL HYPEROSTOSIS

強直性脊椎骨肥厚症のレントゲン像

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ATOMIC BOMB CASUALTY COMMISSION

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HIROSHIMA AND NAGASAKI, JAPAN

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ROENTGENOLOGICAL ASPECTS OF ANKYLOSING SPINAL HYPEROSTOSIS

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SUMMARY

All available roentgenograms of ABCC-JNIH Adult Health Study Hiroshima subjects diagnosed as having ankylosing spinal hyperostosis (ASH) or ankylosing spondylitis were reviewed and 1 case of ankylosing spondylitis and 175 cases of ASH were detected. The most frequent sites of ASH were in the mid and lower portions of the thoracic spine; these lesions were rarely found in the upper portion.

At first, a few vertebrae were affected, and then involvement expanded. In some cases, involvement spread to the entire thoracic spine. The anterior and lateral aspects of the vertebral bodies were mainly affected — the left side less severely, due to aortic pulsations. The hyperostotic structure vertically spanning the anterior aspect varied in thickness up to about 10 mm. However, the rate at which the hyperostosis proliferated did not appear to be specific for any particular vertebra.

Males were about 12 times more frequently affected with this condition than females, especially those in the older age groups. Though diabetes mellitus and hypertension have been reported in other studies to be associated with ASH, we could find no such association between these diseases.

Despite the criteria for differential diagnosis, it is sometimes difficult to distinguish ankylosing spondylitis and ASH roentgenologically. The importance of accurately diagnosing these two entities lies in the need for early treatment of ankylosing spondylitis. The radiologic features helpful in diagnosis are described, and a review of the pertinent literature is included.

要 約

強直性脊椎骨肥厚症あるいは強直性脊椎炎と診断されているABCC一予研成人健康調査の広島の対象者の胸部とその他の適用できるレントゲン写真を再検討し、強直性脊椎炎1例と強直性脊椎骨肥厚症175例を認めた。後者の初発部位は、胸椎中部および下部であることが最も多く、胸椎上部であることはまれであった。

初めに少数の椎体が侵され、それから病変は次第に拡大し、いくつかの症例では、病変は全胸椎に広がっていた。主として椎体の前面および側面が侵されていたが、左側面では、大動脈の拍動のためにその程度は軽い。椎体前面に縦に広がっている過骨性構造物は、厚さ10mmくらいにまでなることがあるが、ある特定の椎体に特異的に骨過形成が進行するとは認められなかった。

男性の罹患率は女性の約12倍で、特に高齢者層に多かった。糖尿病や高血圧と強直性脊椎骨肥厚症との関係を示す報告があるが、われわれは、このような関係を見出すことはできなかった。

鑑別診断基準はあるけれども、強直性脊椎炎と強直性脊椎骨肥厚症をレントゲンの鑑別することはしばしば困難である。この二つの疾病を鑑別することの重要性は、強直性脊椎炎は早期治療を必要とすることにある。診断の助けとなるレントゲン像について述べ、文献的に考察した。

INTRODUCTION

Ankylosing spinal hyperostosis (ASH) consists of bone proliferation on the anterior and lateral aspects of the vertebral bodies which eventually becomes relatively thick. Usually, the thoracic spine is chiefly involved. Clinically, there are no symptoms or physical findings. The etiology of ASH is still unknown.

This condition has been described in the literature for nearly 100 years. Oppenheimer in 1942¹ examined the ossification in the anterior longitudinal ligament, called it "Spondylitis Ossificans Ligamentosa," and considered it a physiological phenomenon — not a disease entity. In 1950 Forestier and Rotes-Querol² first proposed that it be regarded as a distinct disease entity and named it senile ankylosing hyperostosis of the spine. Since then, many investigators have proposed numerous names for this abnormality including: "Forestier's Disease," by Lackner,³ "Vertebral Osteophytosis," by Bick,⁴ "Physiologic Vertebral Ligamentous Calcification," by Smith et al,⁵ and "Spondylosis Hyperostotica," by Ott.⁶

The first report of ASH in Japan was published in 1961.⁷ We describe here the results of our review of all available pertinent roentgenograms of subjects with this abnormality who participated in the ABCC-JNIH Adult Health Study during defined periods of time.

MATERIAL AND METHODS

The Adult Health Study, the major clinical surveillance program of ABCC-JNIH, continues to provide biennial clinical examinations of A-bomb survivors and comparison subjects (originally numbering 20,000) for the purpose of detecting late radiation effects of the atomic bombs.⁸ All participants routinely receive complete physical examinations and laboratory studies. Postero-anterior (PA) stereoscopic and lateral chest roentgenography are included among these examinations. Other examinations, including radiography and fluoroscopy, are performed when clinically indicated.

All chest roentgenograms and all other available radiographs of the spine and pelvis of all Adult Health Study participants in Hiroshima who were diagnosed and coded from July 1962 through June 1974 as having ASH or ankylosing spondylitis were reviewed and 92 cases were detected. A similar

緒 言

強直性脊椎骨肥厚症は、椎体の前面および側面の骨増殖であって、最終的にはかなりの厚さになる。普通は主として胸椎が侵される。臨床的には症状はなく、理学的所見もない。強直性脊椎骨肥厚症の病因はいまだに不明である。

本症は、100年くらいも前から文献にその記述がみられる。1942年に Oppenheimer¹ は、前縦靱帯に発生した骨化巣を観察し、それを Spondylitis Ossificans Ligamentosa と呼んだが、これは疾病でなく、生理的現象であるとなした。1950年に Forestier および Rotes-Querol² は、初めて本症を明確な疾病と考えるべきであることを提案し、それを senile ankylosing hyperostosis of the spine と称した。それ以来、多くの研究者はこの異常に種々の病名を提案している。例えば、Forestier's Disease (Lackner),³ Vertebral Osteophytosis (Bick),⁴ Physiologic Vertebral Ligamentous Calcification (Smithら),⁵ および Spondylosis Hyperostotica (Ott)⁶ などがある。

日本における強直性脊椎骨肥厚症の最初の例は、1961年に報告された。⁷ 本書では、一定期間中に受診した ABCC 一予研成人健康調査対象者の中でこの異常を有すると診断されている者について、適用できるすべてのレントゲン写真を再検討したので、その結果を述べる。

材料および方法

成人健康調査は、ABCC 一予研によって実施されている主要臨床調査プログラムである。最初に選定した合計 20,000 人の原爆被爆者と比較対照者について、原爆放射線被曝の後影響の究明を目的として、引き続き 2 年ごとに臨床検診を行っている。⁸ すべての受診者は、完全な診察および種々の臨床検査を受ける。その検査の一つとして胸部の背腹方向立体撮影と側方向撮影が行われ、臨床的に必要と認められた時にその他のレントゲン撮影や透視検査も行われる。

1962年7月から1974年6月までの間に強直性脊椎骨肥厚症または強直性脊椎炎と診断されて符号化されている広島成人健康調査対象者全員について、その胸部レントゲン写真および脊椎と骨盤部の適用できるレントゲン写真を再検討し、強直性脊椎骨肥厚症92例を認めた。長崎でも

review was attempted for Nagasaki subjects, but it was unsuccessful because of a relative lack of diagnosing ASH there, where the entity was considered an essentially normal finding with age. In addition, from 1 September 1974 through 30 June 1975, an intensive search was made in Hiroshima for all ASH among the prospectively-examined subjects, producing 83 additional cases. It should be noted that our review of earlier films made before the beginning of coding in July 1962 revealed some cases where ASH had been present but not coded.

The form used for data collection during this review is shown in Appendix 1. All roentgenograms were reviewed without knowledge of sex, age, or clinical history of T65 A-bomb radiation dose.⁹

RESULTS

We detected 175 cases of ASH and 1 case of ankylosing spondylitis. Among the 175 subjects with ASH, there were 157 males (89.7%) and 18 females (10.3%) (Table 1). Age at detection ranged from 37 to 82 years, with a mean of 60.7 years. The abnormality was detected most frequently in subjects in their 60's with a total of 74 cases (42.3%). It was next most frequently detected in 58 cases (33.1%) in the 6th decade and then in the 8th decade in 24 cases (13.7%).

同様の再検討を試みたが、同市では本症は本質的に年齢に伴って生じる正常な所見であると考えられたために、長崎で強直性脊椎骨肥厚症と診断されている例は比較的少なく、成果はなかった。そのほか、1974年9月1日から1975年6月30日までの受診者について計画的に調査を行い、広島で強直性脊椎骨肥厚症全例の徹底的検出に努めた結果、新たに83例を得た。なお、1962年7月に符号化の業務が開始される以前の初期の頃のレントゲン写真の再検討も行った。その結果、強直性脊椎骨肥厚症があってもその診断が符号化されていない例を若干認めたことを付記したい。

今回の再検討で資料収集に用いた用紙を付録1に示した。すべてのレントゲン写真は、性別、年齢、T65原爆放射線被曝線量⁹等を伏せて再検討を行った。

結 果

強直性脊椎骨肥厚症 175 例と強直性脊椎炎 1 例を認めた。強直性脊椎骨肥厚症 175 例のうちの 157 例 (89.7%) は男性、18 例 (10.3%) は女性であった (表 1)。発見時の年齢は 37 歳から 82 歳の範囲にわたり、平均 60.7 歳であった。この異常が発見されたのは 60 歳代が最も多く、合計 74 例 (42.3%) あった。これに次いで 50 歳代の 58 例 (33.1%) が多く、70 歳代の 24 例 (13.7%) がこれに続く。

TABLE 1 FREQUENCY DISTRIBUTION OF ANKYLOSING SPINAL HYPEROSTOSIS
SUBJECTS BY AGE AT DETECTION AND SEX

表 1 強直性脊椎骨肥厚症例の度数分布：発見時年齢別・性別

Age at Detection	Male	%	Female	%	Total	%
30-39	2	1.3	0	0.0	2	1.1
40-49	14	8.9	3	16.7	17	9.7
50-59	50	31.8	8	44.4	58	33.1
60-69	70	44.6	4	22.2	74	42.3
70 +	21	13.4	3	16.7	24	13.7
Total	157	100.0	18	100.0	175	100.0

Age range 37-82 years

Our follow-up of these subjects ranged from 3 to 18 years. Onset of the condition was most frequent in the mid or lower thoracic spine. ASH rarely began in the upper portion of the thoracic spine. At first a few vertebrae were affected, but involvement eventually spread. In the early stages of ASH, ossified "bridges" of relatively low density developed

これらの例について 3 年ないし 18 年にわたって経過の観察ができた。本症は、胸椎中部または下部に初発することが最も多く、胸椎上部から発症することはまれであった。初めに少数の椎体が侵され、それから病巣は次第に拡大した。強直性脊椎骨肥厚症の初期では、椎間腔の前方に陰影度の比較的薄い骨性の「橋構造」ができる。これ

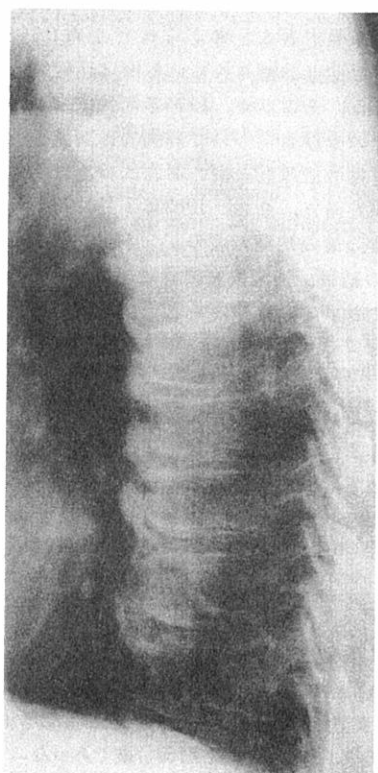


Figure 1 "Wave-like" marked hyperostosis in a 60-year-old male (MF [redacted]).

図1 60歳の男性(MF番号[redacted]). 強度の「波状」の骨過形成がみられる。

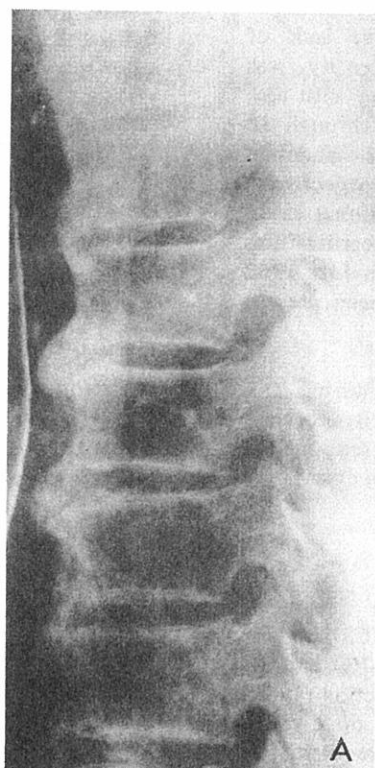
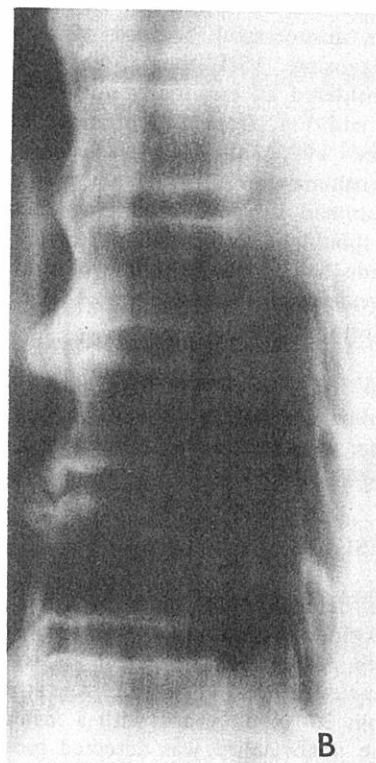


Figure 2 (a) lateral and (b) anteroposterior tomograms of 72-year-old male showing marked hyperostosis (MF [redacted]).

図2 72歳の男性(MF番号[redacted])の(a)側方向および(b)腹背方向断層撮影像。強度の骨過形成がみられる。



immediately anterior to the intervertebral spaces. In most cases, they developed between the junctions of ligaments and vertebral bodies. As ASH progressed, the ossification gradually increased in density and eventually covered most of the vertebral body, increasing in thickness, sometimes to nearly 10 mm. In the final stage, many vertebral bodies were covered by these densities which usually had a wave-like appearance, and were less frequently linear. The "crests" of these waves were located at the levels of the intervertebral spaces (Figure 1).

The hyperostosis was usually more dense than the vertebral bodies themselves. The anterior margins of the vertebral bodies were usually distinguishable from the hyperostosis by a vertical radiolucent zone between them (Figure 2). The hyperostosis increased in both density and thickness and involved an increasing number of vertebral bodies. Many stages of development were often seen simultaneously in a single spine. In some cases, all of the

は、靱帯と椎体との結合部の間にできることが多い。強直性脊椎骨肥厚症の進行に伴って化骨巣の陰影度は次第に増強し、その椎体のほとんど全面を覆うようになる。その厚さも増し、時には10mmくらいに達することがある。最終段階では、多数の椎体が覆われ、その陰影は直線状よりは波状の様相を呈することが多い。この波状陰影の頂点は椎間腔の高さに相当する(図1)。

骨過形成は椎体自体よりも陰影度が濃いことが多かった。椎体前面と骨過形成との間は、普通は垂直な放射線透過部によって区別される(図2)。骨過形成は、陰影度と厚さがともに次第に増し、他の多数の椎体に広がっていった。同一例に本症の種々の進行段階がしばしば同時にみ

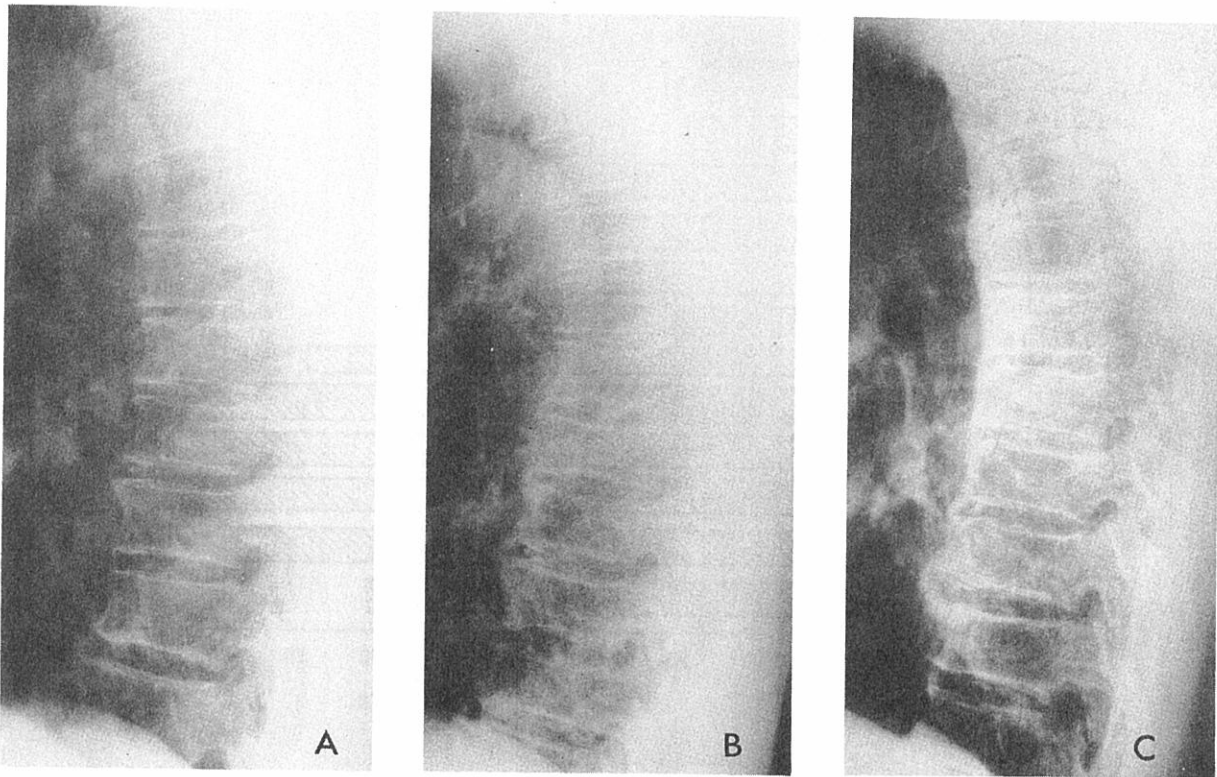


Figure 3 Lateral projection of the thoracic spine of a 51-year-old male, (a) before onset at 58 years, (b) at onset, and (c) at 62 years of age showing development of marked hyperostosis (MF 1234).

図3 男性1例(MF番号1234)の胸椎側方向撮影像。(a)発症以前の51歳の時の所見, (b)58歳で発症した時の所見, ならびに(c)強度の骨過形成を示す62歳の時の所見。

thoracic vertebrae were ultimately affected. This extension is illustrated in Figure 3.

Based on a review of the last available roentgenograms of each subject, the percentage of ASH cases with involvement of each thoracic vertebra is shown in Figure 4. The 9th thoracic vertebra was most frequently involved, followed by T10, T8, T11, and T7, in that order. The upper thoracic vertebrae were less frequently involved, especially in the region of T1 and T2.

The anterior and lateral aspects of the vertebrae were mainly affected. Among 12 cases who received thoracic spine examinations, the right side predominated in 11 (Figure 5). The hyperostotic structure vertically spanning the anterior aspects of the vertebrae varied in thickness up to 10 mm. The rate of proliferation of the hyperostosis was not observed to be specific for any particular vertebra. As seen in Figure 3, though the onset

was delayed, eventually the disease spread to all thoracic vertebrae in some cases. This progression is illustrated in Figure 3.

Based on a review of the last available roentgenograms of each subject, the percentage of ASH cases with involvement of each thoracic vertebra is shown in Figure 4. The 9th thoracic vertebra was most frequently involved, followed by T10, T8, T11, and T7, in that order. The upper thoracic vertebrae were less frequently involved, especially in the region of T1 and T2.

The anterior and lateral aspects of the vertebrae were mainly affected. Among 12 cases who received thoracic spine examinations, the right side predominated in 11 (Figure 5). The hyperostotic structure vertically spanning the anterior aspects of the vertebrae varied in thickness up to 10 mm. The rate of proliferation of the hyperostosis was not observed to be specific for any particular vertebra. As seen in Figure 3, though the onset

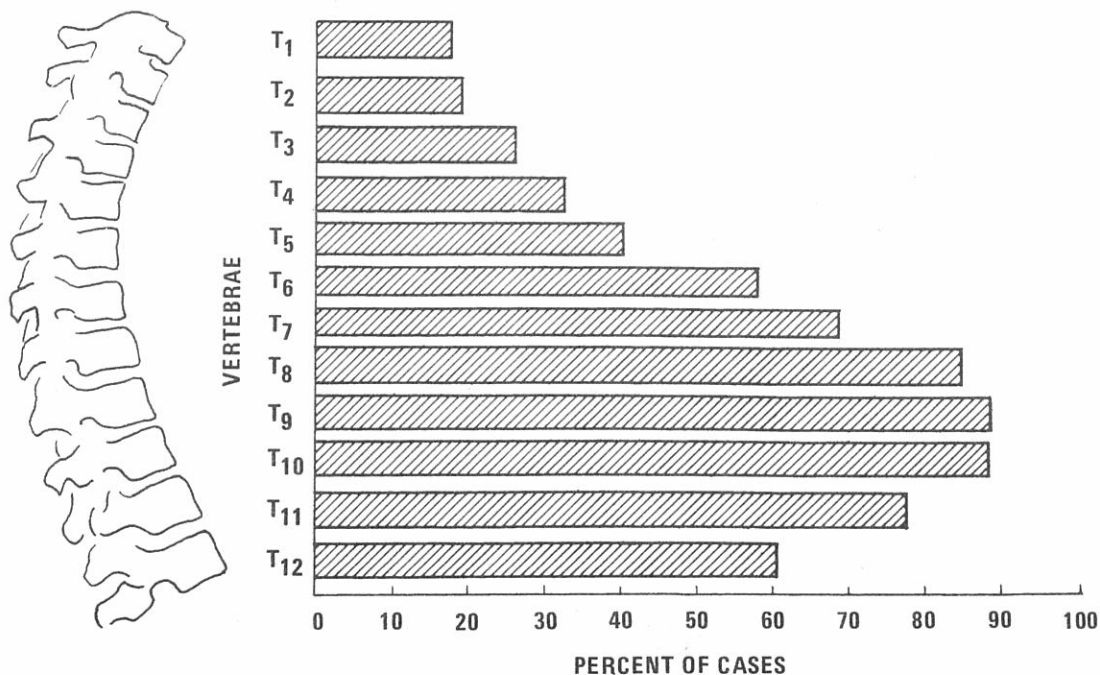


Figure 4 Percentage of 175 ankylosing spinal hyperostosis cases with involvement of each thoracic vertebra (at most recent examination).

図4 各胸椎が侵された175例の強直性脊椎骨肥厚症の百分率分布(最終検査時)

was in the lower thoracic vertebrae, at the last examination the upper vertebrae were involved with a thicker and more dense hyperostosis than the portion originally affected.

The lumbar and cervical spine are said to be involved less frequently than the thoracic portions of the spine. All 175 subjects in this study received PA and lateral chest roentgenography. Lumbar spines and pelves of 112 were examined with AP; 52 of them with lateral, and a few of them with oblique projections. We could not determine the true frequency of ASH in these two regions, because the lumbar spines and pelves of all ASH cases were not examined. Lumbar spines of 31 were affected by ASH in the 52 ASH cases who had lateral lumbar roentgenograms. No case of sacroiliac joint obliteration was found.

Members of the Adult Health Study were examined in the clinic and roentgenographically for varying numbers of times. Such variation could cause

とはいえ、最終検査時では、初発部位よりも胸椎上部の骨過形成が厚く、陰影度も濃いと認められた。

腰椎および頸椎は、胸椎よりも侵されることが少ないといわれている。今回の調査の対象となった175例全員は、胸部の背腹方向および側方向撮影が行われている。このうちの112例は腰椎および骨盤部の腹背方向撮影が行われており、52例では側方向撮影もあるが、斜方向撮影が行われている者は少ない。強直性脊椎骨肥厚症の全例について腰椎および骨盤部検査が行われていないので、両部位の正確な強直性脊椎骨肥厚症罹患率を決定できなかった。しかし、腰椎部の側方向撮影を受けていた強直性脊椎骨肥厚症52例のうちの31例は、腰椎も侵されていると認められた。ただし、仙腸関節の閉塞は1例もなかった。

成人健康調査対象者の受診回数やレントゲン検査回数に差がある。このために有病率の推定に偏りが起こりうる

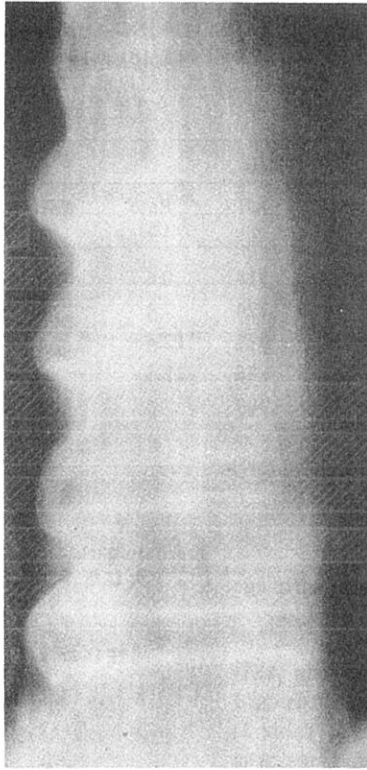


Figure 5 Anteroposterior tomogram of the lower thoracic spine showing a relative lack of ankylosing spinal hyperostosis on the left side in the region of the descending portion of the aorta (MF [REDACTED]).

図5 胸椎下部の腹背方向断層撮影像 (MF 番号 [REDACTED])。大動脈下降部の通過する左側面では、強直性脊椎骨肥厚症が比較的軽い。

prevalence estimates to be biased. Therefore estimates are based on examinations in the clinic during the period 1 September 1970 to 31 July 1971, because the combination of routine diagnosis and the intensive search should have detected nearly all of the ASH cases during this period. In addition, since there have been reports of an association of ASH with diabetes mellitus¹⁰⁻¹⁴ and a suggestion of an association with hypertension,¹⁰ data based on the medical records for this specific sample were analyzed for diagnoses of diabetes mellitus and of hypertension.

Table 2 shows the prevalence of ASH among Adult Health Study subjects examined during September 1970-July 1971. Among males, the prevalence was 7.5% and among females, 0.6%. The age-adjusted figures are 7.1% and 0.6%, respectively (about 12 to 1). Furthermore, except for the 40-49 age group, the age-specific ASH prevalence rates were significantly ($P < .01$) higher for males than for

と考えた。したがって、1970年9月1日から1971年7月31日までの間の成人健康調査受診者について、有病率の推定を行った。すなわち、この間に行われた通常検査と症例探知のための強力な努力の組み合わせにより、強直性脊椎骨肥厚症のほとんど全例が発見されたはずである。そのほか、強直性脊椎骨肥厚症と糖尿病との関係を示す報告¹⁰⁻¹⁴や高血圧との関係を示唆する報告¹⁰があるので、この期間の受診者の医学記録から求めた資料を用いて糖尿病および高血圧の診断の有無を調べた。

1970年9月から1971年7月までの成人健康調査受診者における強直性脊椎骨肥厚症有病率を表2に示した。有病率は男性7.5%、女性0.6%であった。年齢補正を行うとそれぞれ7.1%および0.6%となり、約12対1の比率である。その上、40-49歳年齢群以外は、強直性脊椎骨肥厚症の年齢別有病率も男性が女性より有意に高い ($P < .01$)。そのほか、男性の有病率は年齢とともに増

TABLE 2 PREVALENCE OF ANKYLOSING SPINAL HYPEROSTOSIS (ASH)
BY SEX AND AGE AT EXAMINATION AMONG ADULT HEALTH STUDY (AHS)
SUBJECTS EXAMINED SEPTEMBER 1970-JULY 1971

表2 成人健康調査受診者の強直性脊椎骨肥厚症有病率: 性別・診察時年齢別,
1970年9月-1971年7月

Age at Examination	Male			Female		
	ASH	AHS	Rate (%)	ASH	AHS	Rate (%)
40-49	2	314	0.6	2	676	0.3
50-59	16	224	7.1	4	460	0.9
60-69	34	275	12.4	5	438	1.1
70 +	22	176	12.5	0	298	0.0
Total	74	989	7.5	11	1872	0.6
Age-adjusted			7.1			0.6

females. In addition, the rate increased with age for males but not females.

Results of our search for correlation between ASH and other diseases, such as diabetes mellitus and hypertension, among the males are shown in Table 3; no attempt to investigate these associations was made for females, due to the paucity of female ASH cases.

The males in the Adult Health Study with ASH were significantly older than those without ASH, and in addition, conditions such as hypertension increase with age. Therefore it would be misleading to compare the crude ratios since the age differential between the two samples will tend to exaggerate the differences. In order to make valid comparisons, the combined male Adult Health Study sample was used as the standard population to compute age-adjusted ratios for the ASH sample and the non-ASH sample. As Table 3 shows, the age-adjusted prevalence of diabetes mellitus in the ASH and in the non-ASH samples was not significantly different. For hypertension, there was no association with ASH. The age-adjusted proportion with hypertension was the same ($P \approx 0.05$) in those with ASH as in those without ASH.

Correlations have been reported between ASH and hyperuricemia,¹³ and between ASH and obesity.¹⁵ Some investigators have vaguely referred to associations between ASH and elevated serum cholesterol,¹⁴ and between ASH and heavy manual labor. We could find no such correlations. We also studied frequencies of aortic arch calcification, elevated erythrocyte sedimentation rate, cataracts, syphilis,

大したが, 女性ではそのような増加はみられなかった.

男性における強直性脊椎骨肥厚症とその他の疾患, すなわち, 糖尿病ならびに高血圧との関係を検討した結果は, 表3に示した. 女性に強直性脊椎骨肥厚症が少ないので, このような関係の有無の検討は行わなかった.

成人健康調査対象者の中で強直性脊椎骨肥厚症を有する男性は, 本症を有しない者よりも年齢が有意に高いと認められた. しかも, 高血圧などの疾病状態は, 年齢とともに多くなる. したがって, 年齢の違いは両群間の差を増強する傾向を持つので, 粗有病率を比較すれば誤解を招くであろう. そこで, 有効な比較を行うため, 成人健康調査における男性合計を標準人口として用いて, 強直性脊椎骨肥厚症を有する群と有しない群における年齢補正有病率を計算した. 表3に示したように, 強直性脊椎骨肥厚症の有無別にみた糖尿病の年齢補正有病率に有意な差はなかった. 高血圧も強直性脊椎骨肥厚症との関係はなく, 強直性脊椎骨肥厚症を有する群も有しない群も高血圧の年齢補正有病率は同じであった ($P \approx 0.05$).

強直性脊椎骨肥厚症と過尿酸血症¹³あるいは肥満¹⁵との関係を示す報告がある. 血清コレステロールの増加¹⁴や重労働と強直性脊椎骨肥厚症との関係を莫然と示唆した研究者もある. しかし, われわれは, このような関係を見出すことはできなかった. 大動弓部石灰沈着, 血沈の亢進, 白内障, 梅毒あるいは肺活量の低下の頻度につい

TABLE 3 PERCENTAGE OF ANKYLOSING SPINAL HYPEROSTOSIS (ASH) AND NON-ASH CASES WITH DIABETES MELLITUS AND WITH HYPERTENSION AMONG MALE ADULT HEALTH STUDY SAMPLE, EXAMINED SEPTEMBER 1970-JULY 1971, BY AGE AT EXAMINATION

表3 成人健康調査男性受診者中の強直性脊椎骨肥厚症罹患患者および非罹患患者における糖尿病と高血圧の百分率：診察時年齢別，1970年9月—1971年7月

Age at Examination	With ASH			Without ASH		
	No.	With Diabetes	%	No.	With Diabetes	%
40-49	2	0	-	312	22	7.1
50-59	16	3	18.8	208	35	16.8
60-69	34	5	14.7	241	47	19.5
70 +	22	2	9.1	154	31	20.1
Total	74	10	13.5	915	135	14.8
Age-adjusted			10.0			14.9

	With Hypertension			With Hypertension		
	No.	With Hypertension	%	No.	With Hypertension	%
40-49	2	0	-	312	47	15.1
50-59	16	8	50.0	208	56	26.9
60-69	34	13	38.2	241	92	38.2
70 +	22	11	50.0	154	75	48.7
Total	74	32	43.2	915	270	29.5
Age-adjusted			30.9			30.2

and decreased vital capacity, but no associations with ASH were established. No symptoms correlating with ASH were observed. No increase in the prevalence of ASH was found with increasing A-bomb radiation (T-65) dose.⁹

DISCUSSION

At onset, ASH resembles spondylosis deformans with its deformities of the vertebral bodies and narrowing of the intervertebral spaces. However, the ossification in ASH is different from the osteophytes of spondylosis deformans.

In the latter, the osteophytes arise from the edges of the vertebral bodies themselves, proliferate smoothly, and are clearly continuous with the vertebral bodies. ASH can also develop on the anterior or lateral margins of the vertebral bodies, usually vertically, with the intervertebral spaces intact. When these two diseases coexist in the same spine, they can progress simultaneously.

The osteophytes of spondylosis deformans are also often seen in the cervical and lumbar spine where

でも検討したが，強直性脊椎骨肥厚症との関係は認められなかった。今回の調査では，本症と関係を示す症状は見られなかった。原爆放射線 (T65) 被曝線量⁹ の増加に伴って強直性脊椎骨肥厚症の有病率が高くなるということも認められなかった。

考 察

発症時における強直性脊椎骨肥厚症は，椎体の変形および椎間腔狭小化を示し，変形性脊椎症に類似している。しかし，強直性脊椎骨肥厚症における化骨形成は，変形性脊椎症で見られる骨突起形成とは異なっている。

後者では，骨突起は，椎体自体の辺縁から発生して平滑に増殖するものであり，椎体とは明らかに連続している。強直性脊椎骨肥厚症も椎体の前縁または外側縁に発生することがある。その場合，それは縦方向に発生することが多く，椎間腔は侵されない。同一脊柱に両疾患の併発があると，同時に進行することがある。

変形性脊椎症では，体重や運動のための過重負担が加わる頸椎および腰椎に骨突起形成が多い。特に腰椎におい

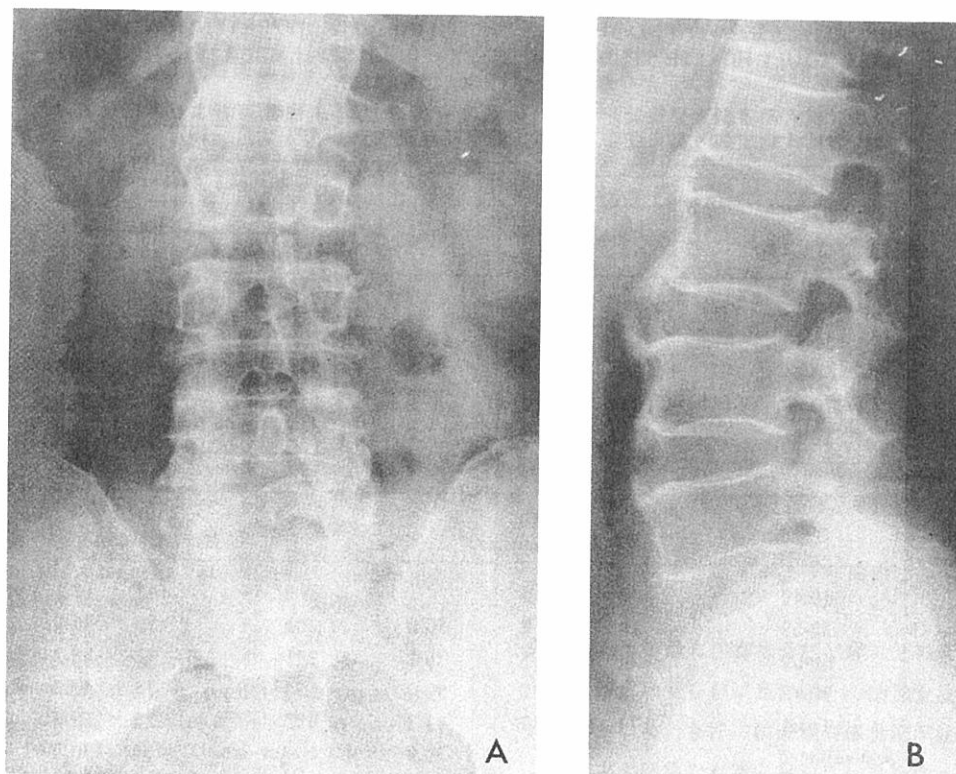


Figure 6 (a) Anteroposterior and (b) lateral projections of the lumbosacral spine of a 71-year-old male showing marked hyperostosis of the spine and unaffected sacroiliac joints (MF [redacted]).

図6 71歳の男性(MF番号 [redacted])の腰椎仙骨部の(a)腹背方向および(b)側方向撮影像。脊椎の強度の骨過形成がみられるが、仙腸関節は侵されていない。

weight and movement cause stress. It can become extensive, especially in the lumbar region. However in the thoracic spine it is rare for true osteophytes to become large, but if large ones occur here, they are usually accompanied by ASH. Another distinguishing feature is that spondylosis deformans is more frequent among males than females. Nathan found it twice as frequent in autopsied males.¹⁶ Some ASH cases may have been included in his series, though this was not mentioned. However we found ASH about 12 times as frequently among males as among females.

We detected one case of ankylosing spondylitis. It is important to differentiate this disease from ASH because of the necessity of early treatment of the former. The main points in the differential diagnosis are shown in Appendix 2.¹⁷ Clinically, subjects with ASH are elderly and have no symptoms, contrary to ankylosing spondylitis. Radiologically, the two entities are sometimes

て著しく発達することがある。しかし、胸椎では、真の骨突起形成が大きくなることはまれであり、大きな骨突起がみられる場合は、強直性脊椎骨肥厚症が併発していることが多い。変形性脊椎症のもう一つの特徴は、女性よりも男性に多発することである。Nathan は、剖検例における罹病率は男性が2倍も高いと認めている。¹⁶ その報告では、強直性脊椎骨肥厚症が含まれていたか否かの記述はないが、若干例含まれていたかもしれない。しかし、今回の我々の調査では、男性の強直性脊椎骨肥厚症罹病率は女性の約12倍であった。

今回の調査で強直性脊椎炎を1例認めた。この疾病は早期治療が必要であるので、強直性脊椎骨肥厚症との鑑別が重要である。鑑別診断の重点を付録2に示した。¹⁷ 臨床的には、強直性脊椎骨肥厚症を有する者は高齢であり、強直性脊椎炎患者の場合と異なって症状は全くない。この二つの疾患は、特に強直性脊椎骨肥厚症が全椎体に

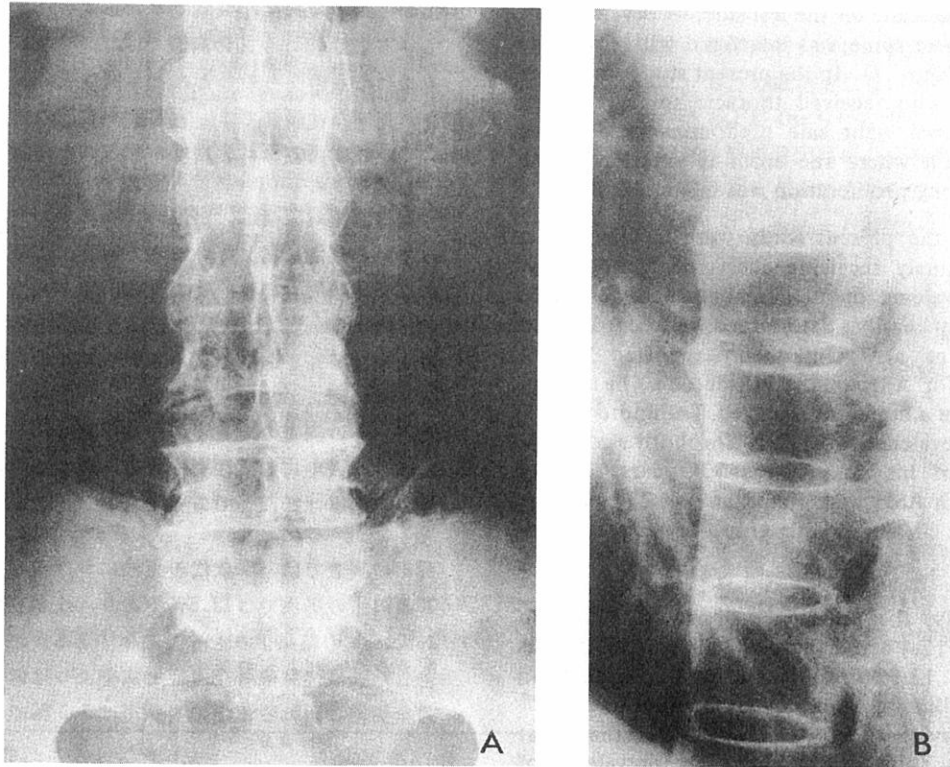


Figure 7 (a) Anteroposterior projection of the lumbosacral and (b) lateral projection of the thoracic spine of a 70-year-old male with ankylosing spondylitis. The apophyseal and sacroiliac joints are obliterated (MF [redacted]).

図7 強直性脊椎炎を有する70歳の男性 (MF 番号 [redacted]) の (a) 腰椎仙骨部腹背方向撮影像および (b) 胸椎側方向撮影像。突起関節と仙腸関節の閉塞がみられる。

difficult to distinguish, especially when ASH involves all spinal segments. Unlike ankylosing spondylitis, the apophyseal and sacroiliac joints are not involved in ASH. The thick and irregular proliferation of new bone continuous with the vertebrae, which distinguishes ASH, is illustrated in Figures 6 and 7.

Forestier and Lagier¹⁷ examined fresh ASH post-mortem spine specimens in the region of the anterior longitudinal ligament and found the ossification partially fixed to the anterior portion of the vertebral body. They reported that the ossification occurred initially in the connective tissue between the vertebral body and anterior longitudinal ligament, that it either invaded the latter or adhered to the former. These findings correspond well with the roentgenological signs.

A right side preponderance has been mentioned for ASH. The autopsied specimens of Culver and Pirson,¹⁸ Nathan,¹⁶ and Forestier and Lagier¹⁷

及んでいる場合は、レントゲンのに鑑別することがしばしば困難である。強直性脊椎骨肥厚症では、突起関節や仙腸関節が侵されないことが強直性脊椎炎と異なっている。強直性脊椎骨肥厚症の特徴である椎体に連続して発生する厚い、不整形な新骨増殖については図6および7に示した。

Forestier および Lagier¹⁷ は、前縦靱帯付近の強直性脊椎骨肥厚症罹患脊柱の新鮮な死後材料を検査し、化骨形成の一部は椎体前面に固定していたと報告している。その報告によれば、椎体と前縦靱帯との間の結合織に先ず化骨形成が生じ、それから後者へ侵入または前者に癒着するという。この結果はレントゲン写真上の所見とよく一致する。

強直性脊椎骨肥厚症は、右側が顕著といわれている。Culver および Pirson,¹⁸ Nathan¹⁶ ならびに Forestier および Lagier¹⁷ の剖検材料についての観察では、大動脈

demonstrated that, on the left side, its development in the thoracic spine was interfered with by aortic pulsation (Figure 7). In the present study, in nearly all subjects who received thoracic spine examinations there was right side predominance. In the lumbar region where the aorta is anterior to the spine, the bony proliferation was bilateral.

Subjects in the present study were members of a sample routinely receiving chest roentgenography, and the prevalence of such an abnormality can thus be relatively easily determined. According to Tsujimoto et al.¹⁴ Julkunen¹⁹ reported a 7% prevalence for ASH in the 7th decade. In a study of subjects in a home for the aged, Tsujimoto et al.¹⁴ found a prevalence of 17.3% for both sexes; the mean age of his sample was 73 years. His 38 subjects with ASH were found among 220 patients investigated. Males in this group had a prevalence of 25.7%.

In our study, the prevalence in males in the 7th decade was 12.4% and that for males over 70 years of age was 12.5%, results somewhat higher than those of Julkunen as quoted by Tsujimoto et al.¹⁴ but the prevalence of ASH was lower than that reported by Tsujimoto et al.¹⁴ The prevalence of ASH for all males over 40 years of age was 7.5%.

Associations between ASH and other diseases, especially with diabetes mellitus, have been reported. A comparison of the prevalence of diabetes among ASH and non-ASH cases and of the prevalence of ASH among diabetics and nondiabetics in several studies is shown in Appendix 3. Tsujimoto et al.¹⁴ reported a rate of 31.9% manifest or latent diabetes mellitus among 120 ASH subjects, and 24.5% ASH subjects among 94 diabetic patients over the age of 40 years. Julkunen et al.¹¹ reported a prevalence of 21% for ASH among 510 subjects with diabetes mellitus, though in the corresponding control group only 4% of 148 nondiabetics had ASH. Hájková et al.¹⁰ among 101 diabetes mellitus patients, found 40 with ASH. Schoen et al.¹³ found 125 (25.0%) ASH patients among 507 diabetes mellitus patients, though in the corresponding control group of 347 nondiabetics, only 9 (2.6%) ASH patients were found. Ott et al.¹² initially found 18 known diabetes mellitus patients among 100 known ASH subjects. With further diagnostic effort, he found 32 additional diabetics among these 100 ASH subjects. Among 82 diabetics, he found 40 (48.8%) had ASH. Moreover, at least in Ott's¹² study, many diabetics were first found by virtue of the ASH findings by roentgen films; the film interpretation and clinical diagnoses apparently were indepen-

dependent of the thoracic spine left side development was interfered with by aortic pulsation (Figure 7). In the present study, in nearly all subjects who received thoracic spine examinations there was right side predominance. In the lumbar region where the aorta is anterior to the spine, the bony proliferation was bilateral.

今回の調査対象者は、定期的に胸部レントゲン検査を受けている集団に属している者であり、この種の異常の有病率を比較的容易に決定できる。辻本ら¹⁴の報告によれば、Julkunen¹⁹は60歳代における強直性脊椎骨肥厚症の有病率は7%であると認めている。辻本ら¹⁴は、養老院収容者の調査で、男女を合計して17.3%の有病率を認めたが、その調査の対象者の平均年齢は73歳であった。被検者220人中に強直性脊椎骨肥厚症が38例発見され、男性の有病率は25.7%であった。

我々の調査では、60歳代の男性における有病率は12.4%、70歳以上の男性では12.5%であり、辻本ら¹⁴の引用したJulkunenの結果よりもやや高率であるが、辻本ら¹⁴の報告した強直性脊椎骨肥厚症有病率よりは低い。40歳以上の全男性における本症の有病率は7.5%であった。

強直性脊椎骨肥厚症とその他の疾患、特に糖尿病との関係があると報告されている。強直性脊椎骨肥厚症を有する者と有しない者における糖尿病有病率ならびに糖尿病を有する者と有しない者における強直性脊椎骨肥厚症有病率を比較したいくつかの調査の結果を付録3に示した。辻本ら¹⁴は、強直性脊椎骨肥厚症120例中に顕性あるいは潜在性糖尿病を31.9%の率で認め、一方、40歳以上の糖尿病94例のうち24.5%に強直性脊椎骨肥厚症があったと報告している。Julkunenら¹¹は、糖尿病510例中における強直性脊椎骨肥厚症の有病率は21%であったが、非糖尿病の対照群148例のわずか4%に強直性脊椎骨肥厚症が認められたという。Hájkováら¹⁰は、糖尿病101例中に強直性脊椎骨肥厚症を40例認め、一方、Schoenら¹³は、糖尿病507例中に強直性脊椎骨肥厚症を125例(25.0%)発見したのに対し、非糖尿病の347例中に強直性脊椎骨肥厚症がわずかに9例(2.6%)あったにすぎない。Ottら¹²は、強直性脊椎骨肥厚症を有する100例のうちに既知の糖尿病例を最初に18例認め、次いで、追加診断検査を行った結果、この100例中に糖尿病を新たに32例発見した。また、糖尿病を有する者を82人検査し、本症を40例(48.8%)発見した。その上、少なくともOtt¹²の調査では、レントゲン写真上の強直性脊椎骨肥厚症の所見に基づいて糖尿病が初めて認められた例が多数あった。すなわち、レントゲン写真の読影と臨床診断は互いに独

dently performed. In our study, before age adjustment, the prevalence of diabetes mellitus is 13.5% among the ASH subjects and 14.8% among those without ASH; the first figure is lower than that reported by Tsujimoto et al¹⁴ and Ott et al.¹² In addition, the prevalence of ASH was 6.9% among those with diabetes and 7.6% among those without diabetes; the first figure is considerably lower than that reported by others.

Schoen et al¹³ could not demonstrate a relationship to hypertension in their study. We investigated the possibility of an association between ASH and diabetes mellitus and between ASH and hypertension, but no associations could be established.

立して行われたものである。我々の調査では、年齢補正を加える以前の糖尿病有病率は、強直性脊椎骨肥厚症例では13.5%、一方、本症を有しない例で14.8%であった。前者は辻本ら¹⁴およびOttら¹²の報告した数字よりも低い。なお、強直性脊椎骨肥厚症の有病率を糖尿病の有無別にみると、それぞれ6.9%と7.6%であり、前者は他の研究者が報告している率よりも低い。

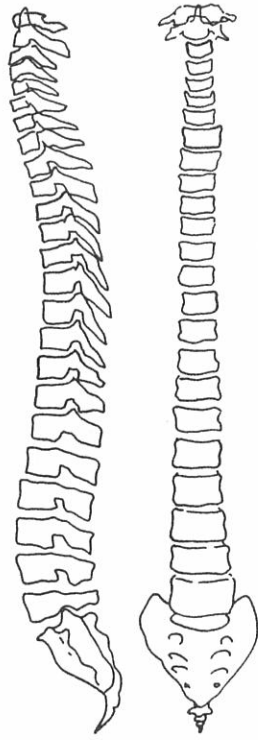
Schoen ら¹³の調査では、高血圧との関係は認められていない。我々も、強直性脊椎骨肥厚症と糖尿病や高血圧との関係の有無を調べたが、何らの関係も証明することはできなかった。

APPENDIX 1

FORM - ROENTGEN ASPECTS OF ANKYLOSING SPONDYLITIS & ANKYLOSING SPINAL HYPEROSTOSIS

付録1 書式—強直性脊椎炎および強直性脊椎骨肥厚症のレントゲン像

Department of Radiology ATOMIC BOMB CASUALTY COMMISSION ROENTGEN ASPECTS OF ANKYLOSING SPONDYLITIS & ANKYLOSING SPINAL HYPEROSTOSIS							
MF No. _____		Name _____		<input type="checkbox"/> Male <input type="checkbox"/> Female	Date of Birth _____		
				<input type="checkbox"/> Hiroshima <input type="checkbox"/> Nagasaki			
	Date of Exam.	Age	Osteoporosis	Hypertrophy	Joint Space Narrowing	Compression Fracture	Grade
T-spine							
L-spine							Sacroiliac Joint Involvement
C-spine							
Signs & Symptoms				Remarks: _____			
1 Back pain (+, -)				_____			
2 Fever (+, -)				_____			
3 Iritis (+, -)				_____			
4 Heart disease ECG (+, -)				_____			
5 Vital capacity (+, -)				_____			
6 RA test (+, -)				_____			
7 ESR (+, -)				_____			
8 Others				_____			
L-spine AP (+, -)				_____			
LAT (+, -)				_____			
Impression				<input type="checkbox"/> Possible <input type="checkbox"/> Probable <input type="checkbox"/> Definite			



APPENDIX 2 DIFFERENTIAL DIAGNOSIS

付録2 鑑別診断

	Ankylosing Spinal Hyperostosis	Ankylosing Spondylitis
CLINICAL		
Age	Elderly	Adolescents and young adults
Posture	Normal or slight kyphosis	Pronounced kyphosis in 60%
Vertebrae	Dorsal stiffness	Ascending lumbar ankylosis to total ankylosis
Pain	Mild dorsolumbar; radiating rare	Maximum during night Frequent thoracic, lumbar & sciatic radiating pain
Respiratory movements	Unaffected	Progressively diminished
Invalidism	Moderate	Often considerable
Erythrocyte sedimentation rate	Normal	Elevated in 75%
RADIOLOGICAL		
Vertebral bodies & discs	Continuous thick outgrowth extends entire height of vertebral bodies on lateral and oblique views Unaffected trabeculae	Early rarification at vertebral angles Squaring of vertebrae Syndesmophytes especially on anteroposterior view Osteoporosis
Apophyseal joints	Unaffected	Joint spaces partially or completely obliterated
Sacroiliac joints	Unaffected	Fusion bilaterally

After Forestier and Lagier¹⁷

APPENDIX 3 COMPARISON OF PREVALENCE OF DIABETES AMONG ANKYLOSING SPINAL HYPEROSTOSIS (ASH) AND NON-ASH CASES AND PREVALENCE OF ASH AMONG DIABETICS AND NON-DIABETICS IN SEVERAL STUDIES

付録3 強直性脊椎骨肥厚症を有する者と有しない者における糖尿病有病率ならびに糖尿病を有する者と有しない者における強直性脊椎骨肥厚症有病率の比較

Investigators	ASH cases	with Diabetes	Non-ASH cases	with Diabetes
Tsujimoto et al ¹⁴	120	32 %		
Ott et al ¹²	100	50 %		
Present study	74	14 %	915	15 %

Investigators	Diabetes cases	with ASH	Non-diabetes cases	with ASH
Tsujimoto et al ¹⁴	94	25 %		
Julkunen et al ¹¹	510	21 %	148	4 %
Hajkova et al ¹⁰	101	40 %		
Schoen et al ¹³	507	25 %	347	3 %
Ott et al ¹²	82	50 %		
Present study	145	7 %	844	8 %

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