A LATE EFFECT ON THE CERVICAL SPINE IN ANKYLOSING SPONDYLITIS

BY

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Adults with advanced ankylosing spondylitis will occasionally exhibit a dramatic resorption of bone from the anterior surface of the cervical spine. This may be confused with the vertebral growth disturbances of juvenile rheumatoid arthritis and an erroneous inference may be drawn that in these cases ankylosing spondylitis must necessarily have begun in childhood (Wilkinson and Bywaters, 1958). It seems pertinent to call attention to this finding to emphasize that it may be acquired in adulthood and is not a "growth disturbance".

Case Reports

Case 1, a 43-year-old male, first noted stiffness and pain in the back with radiation down both thighs in 1939. These symptoms gradually progressed and he developed arthritis of both hips for which he was admitted to the University Hospital in 1944. Physical examination showed moderate rigidity and kyphosis of the dorsolumbar spine and limitation of motion in both hips. Chest expansion was reduced. Neck motion was

normal. There were no rheumatoid nodules. Radiological examination disclosed ankylosis of both sacroiliac joints and dorsolumbar spondylitis with "squaring" of the vertebral bodies, paraspinal ossifications, and fusion of the apophyseal joints. The hips were involved, but the cervical spine was normal (Fig. 1a). A diagnosis of ankylosing spondylitis was made and radiation therapy gave some relief of symptoms.

He was seen again in 1949 for recurrent episodes of faintness which were not relieved by food and were unrelated to posture. Physical examination now showed extreme kyphosis and spinal rigidity. Neck motion was partially limited. Neurological examination was negative. Cervical spine films (Fig. 1b) showed a dramatic resorption of bone from the anterior surfaces of C4, 5, and 6, so that the antero-posterior diameters of these vertebral bodies were reduced. The anterior margin of the cervical spine was continuous and smooth, as though it had been "planed down". The apophyseal joints seen in the lateral projection were fused, the intervertebral disks were reduced in height, and many were partially calcified, and the bones were demineralized. The cause of these symptoms was uncertain. The results of an oral

Fig. 1.—Case 1. (a) 1944 at age 27. Normal cervical spine. (b) 1949. (c) 1954. Extensive, progressive resorption of bone from the anterior surface of C4, 5, and 6. The vertical dimensions of these vertebral bodies are preserved. Note apophyseal ankylosis and atrophy of intervertebral disks.

glucose tolerance test suggested functional hypoglycaemia, but the symptoms continued despite a high carbohydrate diet. He was again seen in 1954 because of an exacerbation of the arthritis. Cervical spine films (Fig. 1c) at this time showed further resorption of bone from the anterior aspect of the cervical spine. Films were again obtained in 1960 and these showed no significant change, though flexion and extension views demonstrated fair motion in the atlanto-occipital and atlanto-axial joints.

Case 2, a 44-year-old male, first developed progressive pain and stiffness of the lumbar spine at the age of 20 years. Arthritis of the hips and shoulders began 5 years later. When the patient was first seen at the University Hospital in 1944, he had marked limitation of motion of the entire spine, including the cervical region. There was marked kyphosis of the dorsolumbar spine, and chest expansion was diminished. There was arthritis of the hips, shoulders, knees, and several joints of the right hand and left foot. Neurological examination was negative. Radiological examination showed typical, far-advanced changes of ankylosing spondylitis with extensive paraspinal ossifications, "squaring", and ankylosis of the sacro-iliac, apophyseal, and costo-vertebral joints. Cervical spine films were not obtained.

In 1954 x-ray films showed progression of these changes and severe kyphosis of the dorsolumbar spine. The cervical apophyseal joints were completely ankylosed and there was resorption of bone from the anterior surface of C6 and C7 and T1 (Fig. 2). Extensive ligamentous ossification was present. The intervertebral disks were atrophic and partially calcified and the vertebrae were demineralized.

Fig. 2.—Case 2, 1954 at age 38. Resorption of bone similar to that shown in Fig. 1 has taken place at C6 and 7 and T1. Advanced ankylosing spondylitis of the entire spine was present.

Comment

Superficial erosions of the anterior surfaces of the vertebrae, so-called "anterior spondylitis", are well known in ankylosing spondylitis and account for the characteristic "squaring" of the vertebral bodies (Romanus and Ydén, 1955). These erosions are frequent in the dorsolumbar region, but may also be observed in the cervical spine. The vertebral changes which are illustrated are remarkable in that the loss of bone substance was so extreme and occurred predominantly in the cervical region. We have noted this now in several patients with advanced ankylosing spondylitis. It seems to be a late manifestation associated with complete ankylosis and extreme kyphosis of the dorsolumbar spine, such that the cervicodorsal segments tend to be horizontal with the patient in the upright position. Continuous hyperextension of the neck becomes necessary to permit the patient to look forward rather than down. One may postulate that, as a result of this continuous stress, a physiological resorption of bone occurs at the anterior, convex surface of the cervical arch by Wolff's law of transformation (Weinmann and Sicher, 1955). It is likely that this effect is superimposed on superficial erosions in this region, and thereby accounts for this degree of bone loss. The particular segments which undergo this transformation vary and this probably depends on the preceding pattern of ankylosis.

Apophyseal ankylosis is a common sequel to the spondylitis of juvenile rheumatoid arthritis and characteristically, the adjacent vertebrae remain small in all diameters in this disease and the intervertebral disks atrophy and may calcify (Marten, Holt, and Cassidy, 1961). These changes are presumably due to growth disturbance and to atrophy secondary to non-use. This appearance may be distinguished from that noted above in which the antero-posterior diameters of the vertebral bodies are reduced. While ankylosing spondylitis may occasionally begin in childhood, it is probably quite uncommon, particularly before puberty.

Summary

Extensive resorption of bone from the anterior surface of the cervical spine is not uncommon in ankylosing spondylitis and is not to be confused with the growth disturbances seen in juvenile rheumatoid arthritis. Two examples of this deformity are illustrated. The radiological findings in one case clearly show that the onset occurred in adulthood, after the spine had reached full maturity. Furthermore, one may expect a generalized reduction in the size of the cervical vertebral bodies in

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juvenile rheumatoid arthritis, whereas in these adult cases the antero-posterior diameters are primarily affected.

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Effets éloignés sur la colonne cervicale dans la spondylarthrite ankylosante

RÉSUMÉ

Une résorption osseuse prononcée de la face antérieure de la colonne cervicale n'est pas rare dans la spondyl-

arthrite ankylosante, et on ne doit pas la confondre avec des désordres de croissance observés dans l'arthrite rhumatismale juvénile. On décrit deux exemples de cette déformation. Les images radiologiques dans un cas montrent clairement que la lésion avait débuté à l'âge adulte, après que la colonne ait atteint sa maturité. Chez ces adultes, c'est le diamètre antéro-postérieur qui est surtout atteint, alors que, s'il s'était agi d'une ancienne arthrite rhumatismale juvénile, on aurait observé une diminution totale de volume des corps vertébraux.

Efectos remotos sobre la columna cervical en la espondilartritis anquilosante

SUMARIO

Marcada reabsorción de hueso en la superficie anterior de la columna cervical no es infrecuente en la espondilartritis anquilosante, y no se debe confundir con disturbios de crecimiento vistos en la artritis reumatoide juvenil. Se describen dos ejemplos de esta deformidad. Los hallazgos radiológicos en un caso muestran claramente que las alteraciones empezaron en la edad adulta, después que la columna había llegado a su pleno desarrollo. En lugar de la anticipada reducción generalizada en el tamaño de los cuerpos vertebrales en la artritis reumatoide juvenil, en estos casos adultos los diámetros antero-posteriores se ven primeramente afectados.

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CORRIGENDUM

It is regretted that in the paper entitled "A Late Effect on the Cervical Spine in Ankylosing Spondylitis", by W. Martel, J. F. Holt, and W. D. Robinson, which appeared in the June, 1962, issue of the *Annals*, the three blocks for Fig. 1 were printed upside down. The correct version is shown below:

Fig. 1.—Case 1. (a) 1944 at age 27. Normal cervical spine. (b) 1949. (c) 1954. Extensive, progressive resorption of bone from the anterior surface of C4, 5, and 6. The vertical dimensions of these vertebral bodies are preserved. Note apophyseal ankylosis and atrophy of intervertebral disks.