PREGNANCY IN ANKYLOSING SPONDYLITIS

A REPORT OF TWO CASES

BY

F. DUDLEY HART, A. C. H. BELL, and G. S. W. ORGANE

From the Westminster Hospital, London

Ankylosing spondylitis is relatively uncommon in females, the sex ratio usually lying in the region of 10 : 1, male : female. The question of pregnancy occurring in spondylitics, however, still arises as, compared with rheumatoid arthritis, the age at onset is considerably less, spondylitis usually starting in the third decade. In view of the early fixation of sacro-iliac joints and the general pelvic rigidity in advanced cases, the management of such a patient demands close co-ordination between physician, obstetrician, and anaesthetist. References to pregnancy in spondylitis are not common; Steinberg (1948) gives case histories of three patients, only one of whom was observed throughout pregnancy by the author. In view of the not uncommon dramatic reversal of the clinical picture in rheumatoid arthritis when pregnancy occurs, the two following case histories are of interest, for such improvement failed to occur in either subject.

Case Histories

Case 1. Married Woman, aged 31. Pain in the right hip on walking commenced in 1939. This was intermittent and disappeared completely in 1946. In 1943 pains across the back were attributed to fibrositis. In 1944 pains commenced in the shoulder girdle, accompanied by a feeling of stiffness. In 1947 an acute bout of pain and stiffness in the neck lasted for two weeks. During this year the pains in the back increased, being worse in the early morning when she felt very stiff. In 1948 the back pain was aggravated by coughing and sneezing, and the front of her chest was also painful. Neck pains returned. *X* rays of the neck were taken and were negative. Previous history and family history were irrelevant.

When first seen by one of us (F.D.H.) in July, 1948, the main complaint was of low backache and stiffness, worse in the morning and improving through the day. Her ribs ached on occasion. On examination physical signs were not marked. The spine appeared to move almost normally. In view of her history skiagrams were taken. These showed ankylosing spondylitis with advanced changes in both sacro-iliac joints, the left being completely ankylosed, the right partially. The intervertebral joints in the lumbar region did not appear to be involved. There was no ligamentous ossification. Hips and ischial tuberosities were normal, and a skiagram of the heart and lungs was also normal.

She was admitted to Westminster Hospital in September, 1948. The only positive findings on examination were restriction of chest expansion below the breasts to $\frac{3}{4}$ inch, slight limitation of movements of the neck and lumbar spine, and some tenderness over the left side of the sternum.

Laboratory Findings :

Sedimentation rate 8 mm. in one hour (Wintrobe) ; three weeks later 24 mm. in one hour.
Wassermann reaction, Kahn test, and gonococcal complement-fixation test negative.
Vital capacity 2,793 ml. (84 per cent. of normal).
Blood count, serum uric acid and urea, thymol turbidity and flocculation tests, and serum colloidal gold reaction all normal.

Deep *x*-ray therapy (Westinghouse) was given to cervico-dorsal and dorsi-lumbar regions by Dr. F. M. Allchin. No sacral port was used to avoid damage to the ovaries.

Dorsi-cervical one direct 15 × 10 cm. port. 1,200r skin.
Lumbo-dorsal one direct 23 × 10 cm. port. 1,200r skin.
Sternum one direct 15 × 10 cm. port. 900r skin.
Factors 200 K.V. 50 cm. F.S.D. 1·6 cu. H.V.L.

Breathing and postural exercises were given in addition.

Before discharge on November 3, 1948, chest expansion had increased to 2¼ inches, and back pain had diminished and mobility increased. Sternal tenderness disappeared completely. Sacral pain (an area not treated) had, however, increased.

On December 1, 1948, she was seen in the Rheumatism clinic. Her neck pains were now slight and gave her no trouble. Her chest felt free and there were no pains on coughing or sneezing. Aches and stiffness persisted in the untreated lumbo-sacral area. Chest expansion had risen to 3 inches. She was now tender over the left iliac crest. On January 26, 1949, sedimentation rate was normal.

Pregnancy.—The patient had been attending a fertility clinic as she and her husband were desirous of producing a family, but over a period of 5 years conception had not occurred. On attendance in September, 1949, she was found to be 4 months pregnant. Aching had recurred down the thoracic spine and tenderness was present over sacro-iliac joints, iliac crests (left more than right), and ischial tuberosities. *X* rays showed changes in both iliac crests, left more than right. The backache abated within three weeks, but bony tenderness in the above sites, worst in the left iliac crest, continued unabated throughout pregnancy. According to the patient, the back " seized up " on one occasion for a week when she was six and a half months pregnant.

Ante-natal progress was normal, the blood pressure never rose above 120/80 mm. Hg, and the urine contained no abnormal constituents.

Blood Test Results :

Wassermann reaction and Kahn test negative.
Blood group O (IV) Rh-positive.
Hb 81 per cent.

On February 2 1950, she was approximately 38 weeks pregnant, the presentation being a vertex left occipito-anterior with the foetal head deeply engaged in the pelvis.

Normal delivery was anticipated and labour commenced on February 15. The first stage lasted only 8 hours 10 minutes, spontaneous delivery of a live male child (6 lb. 15 oz.) taking place after 65 minutes in the second stage. The total duration of labour was 9 hours 55 minutes. The puerperium was quite uneventful and the child was breast fed.

Since pregnancy, bony tenderness continues. During pregnancy there was no alleviation of her symptoms and bony tenderness became more marked. Chest expansion two months after childbirth was reduced to 1¼ inches, and spinal flexion, registered photographically, was reduced to half what it had been before conception. *X* rays showed no appreciable changes in sacro-iliac joints and pelvis, but calcification had occurred in the soft tissues in the lateral aspect between L. 3 and 4.

Comment on Case 1.—Conception, attempted over a period of five years, occurred during a period of increased well-being and freedom from symptoms

following deep *x*-ray therapy to cervico-dorsal and lumbo-dorsal ports, the lower lumbar and sacral areas being untreated. Pregnancy had no alleviating effect; fresh symptoms and physical and radiological signs occurred during pregnancy. Childbirth was uncomplicated and uneventful.

Case 2. Married woman, aged 29. Backache was first noted in 1942 while serving in the A.T.S. at the age of 21. This was constant, but became worse after immobility ;

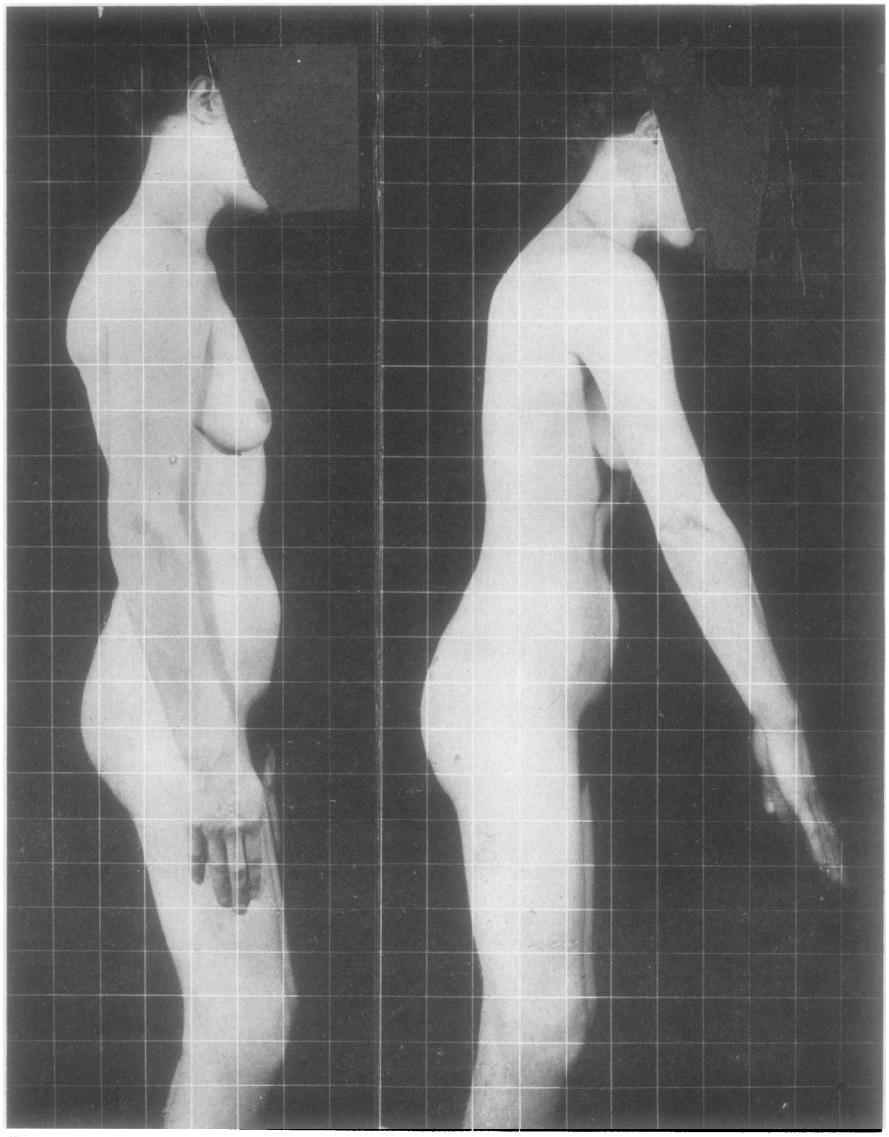

FIG. 1.—Patient 2 standing erect, July, 1948.

FIG. 2.—Patient 2 with full spinal flexion, attempting to touch her toes, July, 1948.

her worst time was on rising in the morning. Skiagrams taken showed changes in sacro-iliac joints and calcification of spinal ligaments. In the following four months pain extended up the spine and stiffness increased ; breathlessness was first noticed on exertion. Early in 1943 a plaster jacket was applied and this was changed subsequently for a plaster bed. She was discharged from the A.T.S. and remained in her plaster bed until April, 1944. She emerged with a completely stiff " poker back ", and her hips also fused. In October, 1945, her neck became stiff and deep *x*-ray therapy was given. Early in 1946 she married. About Christmas, 1947, the temporo-mandibular joints became affected. In June, 1948, both knees became painful and swollen. The previous personal and family histories were entirely negative.

She was admitted to hospital under our care in July, 1948. On examination she was a cheerful, optimistic subject. Chest expansion below the breasts was only $\frac{1}{2}$ inch. Breathing was almost entirely diaphragmatic. The spine was throughout entirely fused with flattening of the lumbar region; no movements of any sort were possible (see Figs 1 and 2). Jaws opened 1 inch. Hips were entirely rigid, fixed in slight flexion. Knee movements were restricted by 50 per cent.—complete flexion and extension being impossible. Ankle, elbow, wrist, and hand movements were all normal. Shoulders retained 75 per cent. of full range. Other systems were normal. She moved about briskly on crutches and was just able to shuffle short distances without them.

Radiographs.—Skiagrams of her entire skeleton showed complete fusion of hips, sacro-iliac joints, all intervertebral articulations, and all costo-transverse and costo-vertebral joints. Calcification of longitudinal ligaments was extensive (Fig. 3). She was a human ramrod, with movement only of her extremities.

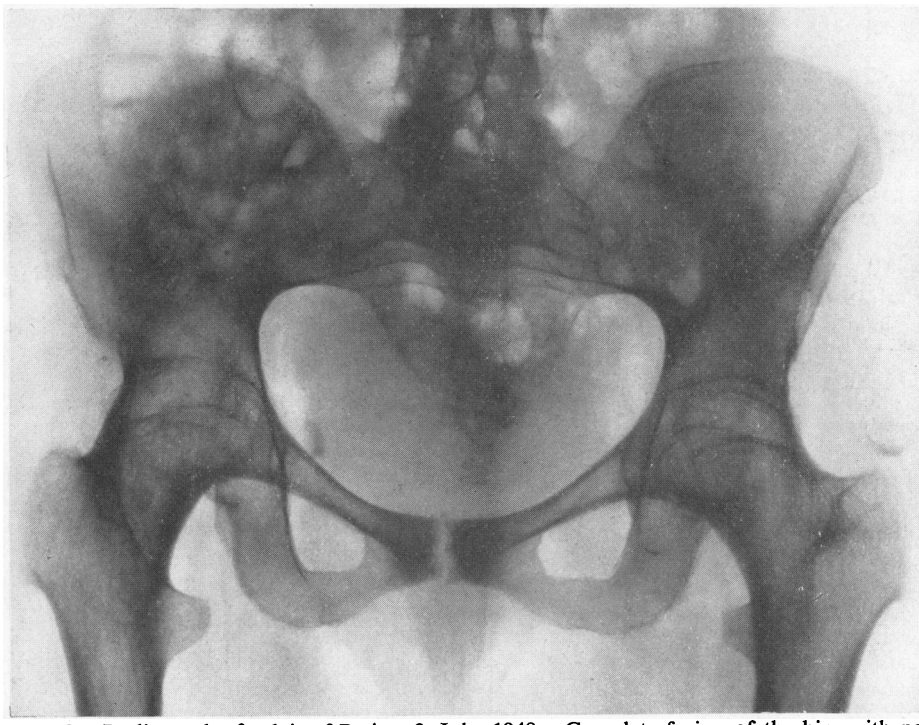

FIG. 3.—Radiograph of pelvis of Patient 2, July, 1948. Complete fusion of the hips with no movement at all under anaesthesia. Complete fusion of sacro-iliac joints. Changes present in the symphysis pubis. Longitudinal ligamentous calcification.

Laboratory Findings :

Blood count, Hb 69 per cent. (Haldane);
R.B.C. 4,350,000 per c.mm.;
Colour Index 0·8;
W.B.C. 9,700 per c.mm. with normal differential.
Sedimentation rate 30 mm. in one hour (corrected Wintrobe).
Wassermann reaction, Kahn test, and gonococcal complement-fixation test negative.
Thymol turbidity 1 unit.
Thymol flocculation negative.
Serum colloidal gold reaction 2+.
Vital capacity 2,030 ml. (61 per cent. of normal).

She was discharged and kept under out-patient observation. A woman of high morale, she did not allow her disability to restrict her activities and attended the cinema and theatre taking with her her own home-made portable chair, for she was unable to flex her thighs to sit on the ordinary sort.

Pregnancy.—She was very anxious to have a family, and as early as October, 1947, had consulted one of us (A.C.H.B.) who did not consider it likely she would become pregnant, but thought a Caesarean section would have to be done if conception occurred. At this time ankles could only be separated 3 inches. The uterus was small and retroverted. Investigation of infertility was not considered desirable, even if possible.

On November 9, 1949, she attended the Rheumatism clinic and was found to be ten weeks pregnant.

Laboratory Findings :

Sedimentation rate 31 mm. in one hour (corrected Wintrobe).
Vital capacity 2,100 ml. (69 per cent. of normal).
Hb 70 per cent. (Haldane).
Plasma proteins 7·35 g. per cent. (albumin 4·17 g., globulin 2·60 g., fibrinogen 0·55 g.).
Thymol turbidity test 1 unit.
Thymol flocculation test negative.
Serum colloidal gold reaction 1+.

Examination at the first visit to the Ante-natal clinic showed the hips to be fixed in adduction and slight flexion, no abduction being possible. At this time the patient was ten weeks pregnant. She stated that this followed normal intercourse, although in view of her deformity this had seemed impossible. Her blood pressure was 130/80 mm. Hg, the highest recording at any time during her pregnancy. The urine remained free from albumin and sugar throughout pregnancy.

Blood Test Results :

Wassermann reaction and Kahn test negative.
Blood group A (II) Rh-positive.
Hb 70 per cent.

On December 12, 1949, her condition was unchanged. The vital capacity had increased to 74 per cent.; there was no complaint of dyspnoea; her only complaint being of nausea, and occasional evening vomiting. On January 20, 1950, the vital capacity was 70 per cent. About this time she first noticed œdema of both legs up to the knee (this was slightly worse towards the end of the day, but also still present in the morning to quite a marked degree), and also a new symptom, aching in her right elbow. Two bouts of severe backache, possibly related to long periods of standing, were noted for a few days in the left sacro-iliac and mid-lumbar areas for a few days.

In April, 1950, she exhibited gross oedema of the legs, but no other evidence of toxaemia. The haemoglobin was now 53 per cent. She was admitted for rest and treatment for anaemia, two pints of blood (packed red cells) being given intravenously on April 29. On May 8, the haemoglobin had risen to 86 per cent., and the oedema was considerably less. By this time she was 36 weeks pregnant, the presentation being a

vertex left occipito-anterior. Her general condition was good. It was decided to deliver her at the 36th week by Caesarean section, because delivery *per vias naturales* appeared impossible and respiratory embarrassment was likely owing to fixity of the thoracic cage and the size of the pregnant uterus. The following day a lower segment Caesarean section was performed under general anaesthesia (see note below), and a live female child (5 lb. 11 oz.) was delivered.

The post-operative course was uneventful, normal involution taking place. The patient was semi-ambulant (a special chair being employed) from the second post-operative day in order to forestall any chest complications. The child made good progress and was breast fed. The patient was discharged on May 27, the 19th post-operative day. Subsequent progress was satisfactory. Oedema of the ankles gradually diminished. Intermittent aches in the right elbow and tenderness over the right olecranon wore off after two months. Joints were exactly as before conception and have remained so since. The daily aspirin ration remains constant at two.

Anaesthesia.—The patient was premedicated with atropine gr. 1/100 ; anaesthesia was induced with 0·2 g. thiopentone, and continued in a light plane with nitrous oxide—oxygen and a trace of ether. When she was already unconscious it was realized for the first time that her head was fixed on the neck in a fair degree of flexion, this led to considerable difficulty in maintaining an adequate airway; she was partly obstructed throughout, and there was some cyanosis at times. She recovered very rapidly from the anaesthetic, and there were no difficulties in the post-operative period. The child's movements were not very active, but it cried spontaneously.

In retrospect, it is clear that some form of local, regional, or nerve block would have been the most satisfactory solution. This would have taken the form of a bilateral posterior intercostal nerve block of the 9th, 10th, 11th, and 12th thoracic nerves, with some local infiltration at the lower end of the incision and behind the pubis.

Comment on Case 2.—One of the worst spondylitics attending the Rheumatism Clinic, this girl with rigid thorax and completely fused hips, pelvis, and entire spine, passed through her 36-week pregnancy with no significant reduction in her vital capacity readings (indeed, they appeared to improve slightly) and no complaint of dyspnoea except when lying with head flat or depressed. No improvement or deterioration was noted in her condition and sedimentation rates remained elevated throughout.

Discussion

In two of the cases quoted by Steinberg (1948) there was indication that pregnancy seemed to benefit the spinal condition, though in his Case 2 symptoms had first appeared during pregnancy. This improvement was not noted in our two cases, where remission, often so dramatic in rheumatoid arthritis in pregnancy, failed to appear. In one case there was progression of the disease clinically and radiologically, during pregnancy. Childbirth in this case was entirely normal as in Steinberg's Case 1. Our Case 2, one of the most severe attending the Rheumatism clinic, suffered none of the respiratory embarrassment that we anticipated, her vital capacity increasing slightly during pregnancy. There was no improvement in her condition in any other respect, but neither was there evidence of deterioration.

Summary

The management of pregnancy in two patients with ankylosing spondylitis is described and discussed. One mild case had an easy labour, but her disease appeared to progress during cyesis. The other, a severely ankylosed patient, was delivered of a normal child by Caesarean section in the 36th week. Her vital capacity rose slightly during pregnancy, and although her thorax was completely fused, no respiratory distress was experienced. In neither case was there any symptomatic improvement or sign of true remission during the months of pregnancy.

REFERENCE

Steinberg, C. L. (1948). *Annals of the Rheumatic Diseases*, 7, 209.

Grossesse dans la Spondylite Anquilosante
Rapport de Deux Cas

RÉSUMÉ

Les auteurs décrivent et discutent le maniement de la grossesse chez deux femmes atteintes de spondylite ankylosante. Chez une d'elles le travail se déroula facilement, mais pendant la grossesse la maladie semblait avancer. Chez l'autre femme l'ankylose était grave ; au bout de la trente-sixième semaine on fit l'opération césarienne et un enfant normal naquit. Pendant la grossesse, malgré la fusion complète de son thorax, on n'observa pas de détresse respiratoire et la capacité vitale s'était même élevée un peu. Dans les deux cas pendant toute la durée de grossesse il n'y eut pas d'amélioration objective ni subjective.

El Embarazo en la Espondilitis Anquilosante
Relato de Dos Casos

RESUMEN

Los autores describen y discuten el manejo del embarazo en dos mujeres con espondilitis anquilosante. En una de ellas la labor fué fácil pero durante el embarazo la enfermedad parecía progresar. En la otra mujer la anquilosis era grave ; al cabo de treinta y seis semanas un niño normal fué extraido por operación cesárea. Durante el embarazo, a pesar de la fusión completa del tórax, no se vió angustia respiratoria, y la capacidad vital estaba algo aumentada. En ambos casos durante el embarazo no hubo mejoría objetiva ni subjetiva.