ANKYLOSING SPONDYLITIS

THE LITERATURE UP TO THE CLOSE OF THE NINETEENTH CENTURY

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Ankylosing spondylitis, a disease causing pain and stiffness referable to the spine and large joints, has affected the human race for ages past. Traces are to be found in the skeletons of the earliest Egyptians and there is no reason to believe that it was then a new disease. Ruffer (1918) found evidence of true ankylosing spondylitis amongst the many cases of spondylitis deformans he came across dating back as far as 5000 B.C. Smith (1908) differentiated between the two diseases in his investigations of Egyptian and Nubian skeletons and found a fairly high incidence of the former in the graves he opened; interesting illustrations can be found in his atlases in the British Museum. Though it is an uncommon disease, it does cripple young men, and I therefore think it remarkable that references in the literature are so rare, at least up to the final quarter of the nineteenth century.

It is doubtful who was the first to describe the syndrome, but I am satisfied that the first adequate clinico-pathological account was given long before the generally accepted authors published their findings. Up to the present time, the credit has been distributed between such writers as Fagge (1877), Sturge (1879), von Bechterew (1892), Marie (1898), Leri (1899), and Strümpell (1897). In the 1880s many others added their cases, notably Clutton (1883), Bradford (1883) in the U.S.A., and Davies-Colley (1885) of Guy's.

As I shall show, to give credit to one or to divide it between a number of these eminent men would be unfair to physicians and surgeons of the earlier 1800s and before. Though there is little to be found in the literature, excellent papers have been written; a résumé of the historical data is given below.

The earlier writers are vague and the famous Caelius Aurelianus (5th cent. A.D.) in his *de Ischiadicis et Psadicis* includes a great number of conditions with similar symptoms. His *Psadicus*

(probably Pliny's *Psadicus Dolor*) corresponds in part to our disease; he refers to the attachments of the lower vertebral column and notes a stiff woodenness:

The patient is seized by pain in the nates, moves slowly, and can only bend or stand erect with difficulty (Mettler, 1947).

Sydenham in the latter half of the seventeenth century was one of the first to give a reasonable description of rheumatism; he divided it into three kinds, the second two following from the first:

- (1) Acute rheumatism or rheumatic fever.
- (2) Chronic rheumatism or modern rheumatoid arthritis.
- (3) Rheumatic lumbago.

Of the third type Sydenham writes (trans. J. Pechy, 1696):

It is properly called the Rheumatical Ach of the Loins, a violent Pain being fixed there, and stretching sometimes to the *Os Sacrum*. . . . Upon which account I have been heretofore mistaken, thinking it was produced from Gravel sticking in those Parts; whereas in truth it owed its rise to the peccant and inflamed Matter of the *Rheumatism*, which afflicts indeed only those Parts, the rest of the Body being untouched. This violent pain continues as the other Species [Chronic Rheumatism] if it be not cured after the same manner, grievously afflicting the poor Patient, so that he cannot lie in his Bed, but is forced to leave it, or to sit upright, rocking himself continually.

The description is a clinical one only, with no allusion to *post-mortem* dissection, but this is typical of Sydenham who was essentially a clinician. It was not until the following century that Morgagni developed the system of correlating clinical and *post-mortem* findings.

In 1691 Bernard Connor used as subject for his M.D. thesis at the University of Rheims a skeleton which he had found in a graveyard. This showed

an advanced stage of the disease (Fig. 1). Connor gave a full and detailed description, and (1695) reconstructed with fair accuracy the probable symptoms, making a few wild shots at the aetiology.

Fig. 1.—Skeleton described by Bernard Connor (1695)

The following are some excerpts from Connor:

As to the crooked and bending Shape of the Skeleton it is reasonable to suppose that it proceeded from the first Formation of the Foetus in the Womb, from the Eggs not having sufficient Room, or being accidentally pressed by some Abscess in the Womb or elsewhere, so that the Carina of the Backbone instead of running straight, was bent into a Circle and kept the same Figure when at full growth that these Bones had taken when soft and tender.

From the Construction of the Parts it necessarily follows that this Body must have been immovable . . . breathing achieved by diaphragmatic Movement

. . . Possibly the Foramen Ovale might continue open, and that by it and the Arterial Canalis the Blood might pass from the Cava to the Aorta, but a part of it passing through the lungs.

Whatever his shortcomings, Connor thus published the first pathological description of a case of ankylosing spondylitis and if his aetiological ideas are amusing to us, his reconstruction of the symptoms caused was logical.

Lyons (1831), writing to the *Lancet*, gives an interesting study of a man aged 36 who had had a 7½ years' history. Pain and stiffness had spread from his elbows and knees to his back, so that in 15 months he was unable to bend because of a rigid spine. Lyons noted that the local pain ceased as each part became fixed, a point now usually ignored or unrecognized. He noted no inflammatory changes. He referred to some totally ankylosed museum specimens, including one in Paris, one in the Hunterian Collection, and one in Trinity College, Dublin. That at Dublin (Fig. 2, opposite) had been the subject of an excellent description in a letter from the Bishop of Cork to the Earl of Egmont (1741). Lyons distinguished the condition of each of these skeletons from that of his own patient. These cases were in fact examples of myositis ossificans progressiva, and his patient was obviously afflicted by a different disease. He tried to arrange admission to a Dublin Hospital with an eye to an eventual *post-mortem* examination and pressed his case by noting that Hunter had given £100 for his skeleton which had had no history attached. Here was one, well documented, which with a little arrangement might be had for nothing! He placed a duty on the Dublin members of the profession to see to this and the man was demonstrated in Dublin shortly afterwards. However, the patient, a Manxman, decided, with excellent judgment, that if he wished to be buried according to custom he had better return to the Isle of Man. This he did and when he died, in order to outwit any "resurrectionists", his friends, in answer to his last wishes, cut up his body into small pieces with "saws and sledges". However, they did not reckon with the persistence of Lyons and his friends, for a year or so later the bones found their way back to the Museum of the Royal College of Surgeons of Ireland and the skeleton was reconstructed. The specimen no longer exists, but there is a description by Houston (1840) in the Museum Catalogue.

About the same time Adams and others in Dublin described ankylosed vertebrae, and Adams (1857), in his *Treatise on Rheumatic Gout*, described typical pain and stiffness and the postures these lead to.

He recognized the apparently healthy vertebral

Fig. 2.—Skeleton at Trinity College, Dublin, described by Robert, Bishop of Cork (1741).

body, and the intact disks with the accompanying bony changes in the ligaments. He included a drawing of an affected odontoid peg (Fig. 3) showing where the ligamentous attachments near the apex were partially calcified, giving the appearance of a “Phrygian cap”.* He noted that the proper function of the marrow was not interfered with.

When Adams addressed the medical section of the British Association at Bristol in 1836, he noted that in “chronic rheumatic arthritis” in women the small joints were affected, while in men it attacked the central joints. Although he thought that the disease

Fig. 3.—Drawing of odontoid peg by Adams (1857)

* Cf. Ruffer (1918).

was only a variant of rheumatoid arthritis, he knew of the differing sex incidence.

Todd (quoted by Adams, 1857) thought that the disease was due to an irritation of the ligaments rather than to inflammation, and reported the case of a girl of 25 who was completely ankylosed and unable to move, for whom he constructed a machine to hoist her into and out of bed.

Hare (1849), writing on curvature of the spine, described bony ankylosis of the lateral and longitudinal ligaments and of the apophyseal joints with fixation of the ribs to the vertebrae, but did not draw any clinical conclusions.

Brodie (1850), who described Reiter's syndrome a life-time before Reiter, put into print what must surely have troubled the inquisitive mind of Connor:

Every one who is conversant with pathological museums must have seen specimens in which the bodies of a greater or smaller number of vertebrae are firmly ankylosed, there being at the same time a deposit of bony matter here and there on the surface adhering to the bone beneath, and extending from one vertebra to the other. It is reasonable to suppose that such a change in the condition of the spine must have been the result of a long-continued chronic inflammation; but in no instance in which I have had the opportunity of observing these morbid appearances, have I been able to ascertain what were the symptoms by which the existence of the disease had been marked in the living person.

He then went on to describe a case which he had observed in 1841. His excellent clinical description might be that of a case seen to-day. The man complained of pain and stiffness in the dorsal and lumbar spine, buttocks, and thighs. The pain was frequently induced by sneezing, "which therefore he completely avoided". One knee joint was affected and the man also had iritis. The onset had been gradual and he had had remissions and exacerbations. Brodie treated him with mercury, iodides, and sarsaparilla, and the disease gradually burned itself out over a period of years, so that eventually the patient was left without symptoms except for "a rigid and inflexible spine".

Wilson (1856) described the case of a young labouring woman, admitted to the Baltimore Almshouse, who over a period of 10 years gradually stiffened from head to foot except for the lower jaw and the ribs. She only complained of pain periodically. Edwin Canton, assistant surgeon at Charing Cross Hospital, records rheumatic arthritis of the spine as a matter of course in some lecture notes of 1855. Adams (1857) had earlier quoted from some of his works.

Brodhurst (1858) showed the case of a young

lieutenant, aged 25, who after gonorrhoea became completely stiff with an ankylosed spine. He showed a second case the next year of a man aged 46 who had also had gonorrhoea; his whole spine was stiff and diaphragmatic breathing was noted. Commenting on these cases, Brodhurst ascribed the ankylosis to "muscular rheumatism", a theory at variance with the currently accepted gonorrhoeal aetiology which lasted till near the end of the nineteenth century.

Hilton (1860), writing on pain, described how he treated a man with pain in the neck with continued and complete rest in the horizontal position. The patient was cured but was left completely ankylosed(!) and later died of tuberculosis.

Von Thaden (1863) described a case which he called "spondylitis deformans" in a young adult man, but as far as one can gather, this was a true case of ankylosing spondylitis.

In 1870, pieces of two spines showing bony ankylosis were pictured in the *British Medical Journal*, one human (from the London Hospital), and one equine (from the Royal College of Veterinary Surgeons). The bony appearance of the longitudinal ligaments and the fact that they did not dip into the disk spaces were noted.

We now arrive at the last quarter of the nineteenth century. Fagge (1877) and Sturge (1879) have lately been mentioned as being the original writers and are compared favourably with hitherto accepted authors, such as von Bechterew (1892-99), Marie (1898), and Strümpell (1897) from whose work Fig. 4 (opposite) is taken. Marie gave a valuable exposé which justifies his name being affixed to the disease.

Leri, his pupil, completed his work by filling in the *post-mortem* details of the cases and wrote on his own account in the early twentieth century. It is interesting that von Bechterew's name is that perhaps most commonly associated with the condition; the reason for this remains a mystery unless it be because he was one of the brightest medical luminaries of his day. The cases he described were, at best, atypical examples of the disease as we know it to-day, exhibiting neurological features unfamiliar to us. It seems, from a translation of his work, that he thought that the fundamental lesion lay in the meninges rather than in the spine itself (von Bechterew, 1893, 1897, 1899). In his *post-mortem* notes, he describes the posterior nerve roots and the columns of Goll and Burdoch as being degenerated! I have never seen a case showing

Fig. 4.—Case described by Strümpell (1897).

added the skeletal details. Sydenham and Caelius Aurelianus must also share the honour.

The available literature of the time spotlights these names, but each was probably one among many contemporaries who thought along the same lines. Even in the eighteenth and early nineteenth centuries and no doubt before, the average medical man knew of the disease, though he usually regarded it as an extension of chronic rheumatoid arthritis, very much as the Americans do to-day. William Clark (1813), in some manuscript notes on rheumatism in the Charing Cross Hospital Medical School Library, mentions affection of the loins, abdomen, chest, and neck, and warns that when the pain has gone the stiffness will remain. Non-venereal iritis as an associated disease has been rediscovered in recent years, but the men of the early nineteenth century were already familiar with it; for example, Rosas (1837) of Vienna classified rheumatic inflammations of the eye into scleritis, keratitis, internal ophthalmia, and generalized acute and chronic ophthalmia, and described twelve cases.

These early writings on ankylosing spondylitis show a fairly clear understanding of the disease, from which we have since progressed little.

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neurological lesions apart from those due to associated disease such as disk herniation.

However, as I have shown earlier, all these authors were rather late on the scene. Nearly two centuries previously Connor had given a very full skeletal picture but had had no clinical opportunity, while in 1850 Brodie had shown the hand of the master. Lyons produced an early case which he followed, one might say, beyond the grave, giving descriptions of the clinical features to which Houston