THE ORTHOPAEDIC ASPECTS OF ONE HUNDRED CASES OF SPINA BIFIDA

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FIG. 1

FIG. 2

Of the many papers on the subject of spina bifida few are concerned mainly with the orthopaedic problems it presents. Macnab (1954) has indicated that, particularly since the advent of antibiotics, the early mortality rate is not as high as formerly, so that although the treatment of associated hydrocephalus is still not satisfactory, more patients may survive to require orthopaedic care. It is my purpose to describe some of the problems encountered in one hundred patients with spina bifida who sought advice at the Royal National Orthopaedic Hospital. Many had already been under treatment at other hospitals for some years, and this made some degree of selection of patients inevitable. Forty-two of these patients were male, and fifty-eight were female; four had a family history of the condition. Only a few had mild static hydrocephalus. Quite apart from the well-known sequelae of the neurological lesions such as club foot and trophic ulceration (Fig. 1), and related conditions such as dysplasia of the hip joint (Fig. 2), a wide variety of associated anomalies was found (Table 1).

In the five patients with café-au-lait type of skin pigmentation no other evidence of neurofibromatosis was found. Kessel (1951) has discussed the association of lateral intrathoracic extrapleural meningocele with spinal deformity in neurofibro-

TABLE I

Associated Anomalies in One Hundred Patients

1. Lumbo-sacral anomalies such as vertebrae of transitional type and vertebrae with abnormally shaped bodies, spinous processes and neural arches	32
2. Scoliosis (18 congenital, 2 idiopathic)	20
3. Congenital fusion of vertebrae, involving from 2-12 segments	18
4. Congenital lumbo-sacral spondylolisthesis	5
5. Hypoplasia of sacrum	5
6. Abnormal ribs (fusion, absence or deformity)	7
7. Syndactyly of fingers	2
8. Sacro-coccygeal dermoid	1
9. Café-au-lait type of pigmentation of trunk and limbs	5
10. Osteochondritis juvenilis (hip 3, spine 3)	6
11. Congenital absence of pectoralis major 2, thyroglossal cysts 3, congenital absence of vagina 1, imperforate anus 2, and macromastia 1	9
Total	110

matosis, but no case resembling those described by him was encountered.

The Spinal Lesion

For the purpose of describing the spinal lesion the patients have been divided into two groups:—

- (1) Twenty-three patients with myelocele or meningocele.

- (2) Seventy-seven patients with spina bifida occulta.

GROUP I: Myelocoele and Meningocele

All but two of the twenty-three patients had extensive involvement of the nervous system. Most of them had had operations on the myelocoele or meningocele within a few months of birth but there was no definite evidence of neurological improvement in any case. The neural arch defect usually involved five or more segments and in one patient no less than twelve arches were defective. Both laminae were often completely absent, and the site of the lesion was most frequently lumbo-sacral. When the defect was small it was nearly always centrally placed (Tables 2 to 5).

Analysis of the Defects in Twenty-three cases of Myelocoele and Meningocele

TABLE 2

Number of vertebrae involved	Number of patients
1	2
2	3
4	2
5	4
6	4
7	2
8	3
9	2
12	1

TABLE 3

Size of neural arch defect	Number of vertebrae involved
1. Less than 1 cm. in width on the radio-graph	33
2. Subtotal	14
3. Total	81

TABLE 4

Site of lesion	Number of patients
Cervical	1
Thoraco-lumbo-sacral	2
Lumbar	4
Lumbo-sacral	15
Sacral	1

TABLE 5

Site of small neural arch defects	Number of vertebrae involved
1. Central	30
2. Right laminar	1
3. Left laminar	2

GROUP II: Spina Bifida Occulta

In this group half of the patients showed involvement of only one or two vertebrae while the remainder showed defects as extensive as those found in myelocoele and meningocele. When only one segment was involved it was the first sacral in seventy-five per cent. of cases. The laminar defects were smaller in a higher proportion of the patients than in Group I, but in the extensive lesions the laminae were usually completely absent, just as in the myelocoele group. The commonest site of the

lesion was again lumbo-sacral and the smaller defects were usually placed centrally (see Tables 6 to 9).

Analysis of the Defects in Seventy-seven cases of Spina Bifida Occulta

TABLE 6

Number of vertebrae involved	Number of patients
*1	28
2	11
3	7
4	4
5	9
6	7
7	5
8	3
9	2
11	1

*This was the first sacral segment in 21 of the patients.

TABLE 7

Size of neural arch defect	Number of vertebrae involved
1. Less than 1 cm. in width on the radio-graph	141
2. Subtotal	8
3. Total	113

TABLE 8

Site of lesion	Number of patients
Cervical	2
Cervico-thoracic and sacral	1
Cervical and lumbo-sacral	1
Thoracic	2
Thoraco-lumbar	1
Thoraco-lumbo-sacral	3
Lumbar	7
Lumbo-sacral	37
Sacral	23

TABLE 9

Site of small neural arch defects	Number of vertebrae involved
1. Central	118
2. Right laminar	12
3. Left laminar	11

Cutaneous and subcutaneous changes were present in just over fifty per cent. of the patients with spina bifida occulta (Table 10); in the remainder the skin and subcutaneous tissues of the back were normal, although in five there was bony gap large enough to be easily palpable.

TABLE 10

Cutaneous and subcutaneous evidence of spina bifida occulta in 39 patients	Number of patients
Hairy patch	22
Haemangioma and telangiectasis	16
Palpable bony defect	16
Scarring	9
Lipoma	8
Lumbar linear scleroderma	3
Dimples	3
Pigmentation of skin	1

Of the seventy-seven patients in Group II fifty had neurological lesions. The presence either of

cutaneous and subcutaneous changes or of extensive laminar defects indicated that involvement of the nervous system was likely, although they were occasionally present when the central nervous system was normal. Of those cases without superficial evidence of spina bifida fifty per cent. had neurological lesions (Tables 11 and 12).

TABLE 11
Number of patients

Cutaneous and subcutaneous changes with neurological lesion	31
Cutaneous and subcutaneous changes with normal C.N.S.	8
No superficial changes with neurological lesion	19
No superficial changes with normal C.N.S.	19

TABLE 12

<i>Number of vertebrae involved</i>	<i>Number of patients with neurological lesion</i>	<i>Number of patients with normal C.N.S</i>
1	14	14
2	4	7
3	7	0
4	4	0
5	8	1
6	6	1
7	3	2
8	2	1
9	2	0
11	0	1

Tables 6 and 12 indicate that about fifty per cent. of the patients with spina bifida occulta of the first sacral segment had neurological lesions. This high figure is of course the result of the selection of patients to which I have already referred. By contrast Campbell Golding (1950) has found spina bifida occulta of the first sacral segment in nineteen per cent. of adult patients subjected to routine radiological examination.

Neurological Lesions

Neurological examination of all the hundred patients showed the following features:—

1. Nervous involvement was usually more severe and more extensive in the myelocoele and meningocele group but equally severe disturbances were found occasionally in spina bifida occulta.
2. The lower lumbar and sacral segments of the cord were most frequently involved.
3. Sensory and motor paralysis usually occurred together but either could be present alone.
4. Both the sensory and the motor paralysis were usually bilateral. In the unilateral cases the numbers of right and left lower limbs affected were equal.
5. Motor paralysis was usually of the lower motor neurone type but in thirty per cent. of the patients signs of pyramidal tract involvement were present. In most patients the paralysis

was incomplete; certain muscles tended to escape and various patterns of paralysis thus emerged.

Paralysis Affecting the Hip

Twenty-one patients had paralysis of muscles controlling the hip. In five it was unilateral, slight and accompanied by a similar degree of paresis in the rest of the limb. In sixteen the paralysis was severe, bilateral and accompanied by extensive paralysis more distally.

Three main groups were observed:—

1. Paralysis affecting all muscles uniformly but varying in severity from case to case.
2. Paralysis in which certain muscles were strong and others weak.
3. Paralysis associated with dysplastic changes in the hip (Table 13).

TABLE 13

<i>Condition of the hip in 21 patients</i>	<i>Number of hips</i>
Normal	5
Uniform hip paralysis	14
Paralysis with muscle imbalance	12
Paralysis with dysplastic changes	10
	41

(One patient had a disarticulation through the hip at the age of fourteen years for a flail limb)

The first group showed three main clinical pictures:—

(a) Slight general weakness, all muscles being graded about 4 on the M.R.C. scale. These patients either had no complaints or felt that the affected hip was somewhat weaker than the other.

(b) A moderate degree of paralysis causing a Trendelenberg type of limp from abductor weakness.

(c) Severe paralysis with the joint completely or almost completely flail. In none of five such hips was dislocation present.

In the second group paralysis was again variable in severity and the striking feature was the existence of muscle imbalance. In two patients the flexors were normal, the extensors weak and the other muscles fairly evenly balanced. Flexion deformity of the hip ranging from 50 to 60 degrees was present. In the remaining ten hips in this group the flexors and especially the adductors were stronger than their antagonists—a cause of spontaneous dislocation of the hip described by Watson-Jones (1926) and Hart (1928). In five of these hips the joint was dislocated (Fig. 3). In four of the other five the short rotator muscles were not severely paralysed, their grading being between 3 and 4. This suggests that these muscles normally may play an important part in maintaining the head of the femur within the acetabulum.

In the third group dysplasia of the hip was present as well as paralysis, four presenting the

FIG. 3.—Bilateral paralytic dislocation of the hip.

typical appearance of congenital dislocation and six having a shallow socket with or without malformation of the acetabular rim.

The type of paralysis was again that of muscle imbalance of variable severity. Fig. 2 shows a subluxated hip with an inadequate socket in a patient with almost complete paralysis of the lower limbs. Fig. 4 shows the hips of a girl of six years who had a large myelocoele and severe paralysis of the hip muscles. The flexors and adductors were just a little stronger than their

FIG. 4.—The radiograph at the age of six years of a child with an extensive lumbo-sacral spina bifida and severe paralysis of the lower limbs. The acetabula are well formed, but there is some degree of coxa valga.

antagonists and the acetabula were well formed. Five years later (Fig. 5) the left hip was beginning to subluxate and the acetabulum had become malformed. In five of the dysplastic hips dislocation had occurred and subluxation was present in two.

Blundell Jones (1954) has recently discussed the role of coxa valga in various types of paralytic dislocation of the hip. He found that an increase of the angle between the neck of the femur and the shaft was invariable and that it was most severe when paralysis was present in the first two years of life. Piersol (1930) has stated that the normal angle ranges from 110 to 144 degrees in adults, the average being 125 degrees. At birth the angle of the neck may be 160 degrees, diminishing to its permanent angle by puberty. Blundell Jones (1954) found that the major part of the reduction in the angle took place during the first five years of life.

I have been unable to confirm his finding that coxa valga is "always present." In the thirty-six hips of this series in which varying degrees of paralysis were present, fifteen showed definite coxa valga, the others being within the normal range. Of the twelve dislocated and subluxated hips six showed coxa valga, which was only severe in four instances. Paralysis had, of course, been present in all these patients since birth.

Fig. 6 shows the radiograph of the hip

FIG. 5.—The radiograph of the same child at the age of eleven years now shows a defective roof to the left acetabulum and early subluxation of the hip joint.

FIG. 6.—A radiograph of a child with extensive lumbo-sacral spina bifida and severe paralysis. There was considerable imbalance of the muscles controlling the left hip joint with complete paralysis of the short rotators. The left acetabulum is shallow and the coxa valga is severe. The hip has already dislocated.

FIG. 7.—The right lower limb of a young girl to show a marked external rotation deformity of the foot in relation to the femur (see text). Several years previously a gross equino-varus deformity had been corrected by radical tarsectomy. Next a fixed flexion deformity of the knee joint was corrected by complete tenotomy of the hamstrings. The scar just above the patella marks the site of a painless pressure sore from the post-operative plaster. The left leg was completely flail, and when a fracture of the femur occurred it was promptly amputated through the site of fracture.

boy with a large meningo-myelocele. This patient demonstrated all three of the factors which I believe tend to produce dislocation of the hip in these patients:—

- (a) Severe paralysis; the flexors and adductors were stronger than their antagonists, whilst the short rotators were completely paralysed.
- (b) A shallow acetabulum.
- (c) Severe coxa valga.

The Knee and the Leg

Nineteen patients presented deformities of the knee joint; flexion deformity was by far the most frequent, and was bilateral in half of these patients. In the others the opposite knee was either recurved, flail or normal (Table 14).

TABLE 14				
Condition of the knee joints in 19 patients				
Flexion deformity	..	..	..	27
Back knee	..	..	..	4
Flail	..	..	..	4
Normal	..	..	..	3

The amount of flexion deformity varied from 10 to 60 degrees, 30 degrees being frequent. In the majority the hamstrings and calf were strong

and the quadriceps weak; occasionally the hamstrings were spastic. In a few the main cause of deformity was capsular contracture following years of nursing with the joint flexed. Only one of the four instances of back knee was severe. As might be expected, minor degrees of external rotation deformity of the lower limb were frequent in patients with flexion deformities of the hip and knee. Fig. 7 shows a severe deformity

in a girl with a very strong biceps femoris and a tensor fasciae femoris contracture.

The Foot

Of all deformities, those of the feet were the commonest (Table 15).

TABLE 15	
Bilateral symmetrical deformity ..	30
Bilateral asymmetrical deformity ..	6
Unilateral deformity	27
Total	63

Equino-varus was much the commonest deformity and was frequently accompanied by secondary cavus. A few patients had a cavo-varus deformity only, some equinus being masked by the inversion of the hind foot. A smaller number presented with pes cavus alone; calcaneo-cavus, simple valgus and total paralysis were less common (Table 16).

TABLE 16	
Condition of 126 feet in 63 patients	Number of feet
Equino-varus	48
Varus only	9
Cavus	23
Calcaneo-cavus	6
Valgus	3
Flail foot	10
Normal	27

Pes Equino-varus. The patients with equino-varus showed a wide range of the degree of paralysis. As a rule all muscle groups were involved to some extent, though the calf sometimes escaped completely. The posterior leg muscles were much stronger than those in the anterior compartment and the invertors always stronger than the peronei, which were usually severely paralysed. In most of the patients tibialis posticus was stronger than tibialis anticus. The deforming factors therefore are the strong calf, which itself tends to increase varus of the foot in the presence of equinus, the weak peronei and the relatively stronger tibial muscles. The most severe deformities occurred when the calf and tibialis posticus were powerful. Adduction of the forefoot was less severe than in congenital clubfoot and the heel seldom so poorly developed.

Many of the patients had been under treatment for some time when they were reviewed. Correction of the deformity by conservative measures such as strapping, splints and repeated plaster casts had frequently been regarded as successful, but cessation of treatment was often followed by rapid relapse according to the degree of muscle imbalance present. Transplantation of the tendon of the tibialis anticus to the outer side of the foot was performed in ten instances; in one foot there was over-correction, while all the others still

showed equino-varus. In twenty instances Steindler operation had been performed and no further treatment was contemplated unless deformity recurred. In a third of the patients with equino-varus stabilisation of the foot by some type of triple arthrodesis was eventually performed. Half of these patients finally had feet of good shape and function and were able to wear ordinary shop shoes; in the other half some deformity remained, especially varus, due either to inadequate correction or to the continued action of deforming muscles.

A number of patients with mild equino-varus deformities had no treatment apart from inside irons with outside T-straps, varus wedges to the soles and heels and surgical boots. Nevertheless some deformity remained in most of the patients who had attended hospital. The infrequency of the attempts to balance the foot by suitable tendon transplantation after closed or open correction of the deformity suggested that the part played by muscle imbalance had not been sufficiently recognised.

Pes Cavus. Pes cavus was much less frequent than equino-varus. The incidence of bilateral and unilateral deformity was equal. Half of these patients complained of pain under the metatarsal heads, of weakness of the foot and of inability to wear shop shoes. Most of those with pain had fixed hyperextension of the metatarso-phalangeal joints, rigid clawing of the toes and callosities under the metatarsal heads. The severity of the cavus was very variable and even in those with no symptoms and normal sensibility some very severe deformities were observed. Electrical reactions showed evidence of denervation in only fifty per cent.

Other deformities of the foot were uncommon. Only six patients had the calcaneo-cavus deformity so frequent in poliomyelitis, and three had a valgus deformity caused by weak peronei in otherwise flail feet. Complete flaccid paralysis below the knee was present in ten instances.

Impairment of Sensibility and its Sequelae

Fifty patients showed loss of sensibility. In the myelocoele group sensory loss was severe and widespread; the lower lumbar and sacral dermatomes showed a similar frequency of involvement and the higher lumbar segments were also sometimes affected. In patients with spina bifida occulta the fifth lumbar and first sacral dermatomes were affected far more frequently than any other.

The impairment usually involved every type of both exteroceptive and proprioceptive sensation. As a rule, pain, touch and thermosensibility were equally affected. Vibration sensibility was fre-

FIGS. 8 and 9.—The clinical and radiographic appearances of a patient with neuropathic arthritis of the left ankle and tarsal joints.

quently diminished and even lost if the exteroceptive loss were severe and widespread. Diminution of deep pressure and pain sensibility was frequent, but complete loss was rare. Loss of postural sensibility was uncommon, nearly always confined to the toes and feet, and accompanied by extensive motor paralysis and severe loss of other forms of sensibility. When the sensory disturbance was severe, trophic changes in the soft tissues, bones and joints were frequent. The skin was thin, often shiny, and either dry or hyperhidrotic. The nails were either brittle, ridged or short.

Ulceration. Trophic ulceration recurring over periods of years had been present in fifty per cent. of the patients with sensory loss. The common sites were the postero-lateral aspect of the heel,

the outer side of the foot, the dorsum of the toes and the buttocks.

Perforating ulcers under the fourth or fifth metatarsal heads were present in six patients with equino-varus or cavo-varus deformities and severe sensory disturbances. They had all been present for years, healing slowly during periods of rest in bed and recurring on renewed activity. Complete correction of the deformity and the provision of suitable sponge rubber insoles was the only successful line of treatment, excision of the metatarsal heads and necks without correction of deformity being followed by early recurrence. Fig. 1 shows a patient with severe bilateral equino-varus deformity, a large trophic ulcer on the right foot and a perforating ulcer under the left fourth metatarsal head. The prognosis in

FIG. 10.—The radiograph of a boy aged four years to show excess callus formation after a spontaneous fracture of the upper end of the femur (see text)

perforating ulcers was bad; in fifty per cent. amputation became necessary because of increasing deformity, or of failure of attempts to correct it, or because of chronic sepsis.

Neuropathic Bones and Joints. Neuropathic arthritis of the ankle and tarsal joints developed in only one patient (Figs. 8 and 9). Spontaneous and almost painless fractures occurred in four patients. In two union took place normally; in the other two the fractures were notable for the development of excess callus. Fig. 10 shows the radiograph of a boy of four who had a severely paralysed limb and a dysplastic hip. After the failure of conservative methods, open reduction of the hip and later a rotation osteotomy of the femur had been performed. After removal of the plaster he developed a supracondylar fracture while lying in bed and later a subtrochanteric fracture with a great mass of callus. Further fractures followed and the limb was eventually

amputated. Histological examination showed normal callus and the "chondroid tissue" described by Baker (1946) in the hyperplastic callus of osteogenesis imperfecta was not observed.

Excessive callus in fractures associated with spina bifida has been reported by Gillies and Hartung (1938) who described two instances of fracture of the upper tibia with sclerosis, fragmentation and excessive callus formation; there was widening of the epiphyseal line and lateral displacement of the head of the fibula. Their cases bear a close resemblance to one case in this series. A young girl with spina bifida, severely paralysed lower limbs and gross sensory impairment came to hospital complaining of swelling of the upper tibial region of the left leg. A radiograph showed the changes described by Gillies and Hartung but no fracture (Fig. 11). The Wassermann and Kahn reactions were negative. One year later the tibia had become almost normal again (Fig. 12). These changes are probably the result of trauma in a limb with decreased sensibility.

Alterations in the Neurological Symptoms and Signs

The neurological deficit in spina bifida has been said to increase with growth, and Ingraham (1933) has stated that this occurs frequently between the ages of six and ten years. This is thought to be due—apart from the rare development of benign and malignant neoplasms or of increased growth of lipomata already present—to tethering of the cord and cauda equina at the site of myelodysplasia with traction on the cord above resulting from difference in the rate of growth between the neural and skeletal axes, that is to say, the same mechanism as for the Arnold-Chiari malformation. The onset or increase of neurological change is accepted by many as an indication for exploration of the cord with a view to freeing the nervous tissue and thus preventing further deterioration. An increase in deformity is not necessarily due to further involvement of the nervous system and a perforating ulcer may be due only to increasing activity or weight bearing. I found definite evidence of an increase in the neurological deficit such as the onset of urinary symptoms or the development of paralysis in a muscle previously known to be normal, in only five patients whose ages ranged from four to seventeen years.

Summary

1. One hundred patients with spina bifida have been examined from the orthopaedic point of view and the associated anomalies enumerated.
2. The spinal lesions in these patients are considered in detail.

FIG. 11.—The radiographs of a young girl with spina bifida and gross sensory loss in both legs. A painless swelling of the upper tibia developed. Note the sclerosis of bone with appearances of fragmentation and new bone formation. The epiphyseal line is widened and the head of the fibula is displaced laterally.

FIG. 12.—A radiograph of the same patient one year later showing spontaneous resolution of the changes. After a further year the films showed almost normal bone structure.

3. A description of the motor paralysis in the lower limbs and its relation to various deformities is given.
4. The impairment of sensibility, its incidence and sequelae are described.
5. Progression of neurological changes is discussed.

Postscript

I wish to record my thanks to Mr. H. J. Seddon for arousing my interest in this subject and for his helpful criticism, to Mr. K. I. Nissen and Mr. J. I. P. James for their continuous encouragement and advice, and to all those members of the Consultant Staff of the Royal National Orthopaedic Hospital who kindly allowed me to examine their patients. I wish also to record my appreciation of the ever ready aid of the Medical Records Department of the Royal National Orthopaedic Hospital,

the Department of Medical Photography and the Librarian of the Institute of Orthopaedics.

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(Postgraduate Medical Journal, July, 1954)

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