LIPOMA OF THE CAUDA EQUINA*

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Although intradural lipomas of the cauda equina with extradural extension have been recognized for over a century, their importance is not widely known. The subcutaneous swelling is easily mistaken for a meningocele and since there is normal skin cover and no neurological involvement in the early stages, a conservative approach is usually advised. It is not generally appreciated that the subcutaneous lipoma is in fact a mushroom-like structure with a stalk which passes intradurally and is firmly adherent to the cauda equina, the filum terminale, and the conus medullaris.]

[In due course most of these lipomas cause neurological symptoms. The onset of neurological involvement may be early or late. It may be sudden and severe] and for this reason we now recommend early exploration, even in the absence of any neurological abnormality.

Review of the Literature

Johnson (1857a, b) gave the first accurate description of this condition. A subcutaneous swelling in the sacral region, first noted at 3 weeks, gradually increased in size, but the child was symptom-free until 9 months of age when he developed twitching and convulsive movements of the right leg. At operation a month later there was a subcutaneous fatty tumour the size of a small orange, with a stalk protruding through a small opening in the sacral canal. It was continuous above with the membranes of the spinal cord from which it was dissected free without opening the membranes. The child improved after operation, the abnormal movements of the leg disappeared, and he was able to stand and to take a few steps. Six weeks later he died of peritonitis. At necropsy a fatty mass was found within the dura extending upwards and pressing on the spinal cord and involving the roots of origin of the lowest spinal nerves, which seemed to be embedded in its substance. There was an associated malformation of the lower sacral vertebrae.

In spite of Johnson's very detailed description there have been surprisingly few reports in subsequent papers. Ingraham and Swan (1943) commented on the frequent association of an overgrowth of fatty tissue with spina bifida and meningeal defects, and noted that where a meningeal defect was present, the lipomatous tumour might extend intradurally for some distance and even cause intrinsic compression of the nerves or the cord. Ingraham and Lowrey (1943) reviewed 65 patients with spina bifida occulta in whom symptoms were present: 31 had abnormality of gait or other orthopaedic symptoms; 22 had a local abnormality of the lower spine, either a swelling or an overgrowth of hair or a scoliosis; 7 had incontinence of urine; and in the remaining 9 patients the presenting symptoms were unrelated and the spina bifida was an incidental finding. In 13 of these 65 there was an associated lipoma. They observed that the lipoma might be at any tissue level, from the cord itself to the subcutaneous tissue, and might extend directly from one layer to another, binding the various tissues together and preventing the normal movement of the cauda equina and the spinal cord with growth. In 5 patients they found a combined intradural and extradural lipoma.

Groff and Yaskin (1947) described a 12-year-old boy who had had a congenital lumbosacral meningo-myelocoele operated on at 3 years, without improvement in his urinary incontinence. A second operation at 12 years of age revealed a fatty tumour extending from the end of the spinal cord down the sacral canal, with the roots of the cauda equina firmly adherent to it. There was thought to be some improvement after operation, but the bladder still needed manual expression.

Bassett (1950) presented the first detailed study of lipoma of the cauda equina as an entity and stressed the importance of early diagnosis and operative treatment. He reviewed 9 patients, all female, including 2 sibs. One of his patients (Case 4), operated on at 2 months of age, remained symptom-free. Another (Case 8), also seen initially at 2 months of age without neurological deficit, had operation deferred; at 16 months there was gross

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weakness of the legs and inability to stand, with sensory and sphincter loss. Operation at that stage did not result in any marked improvement in motor power or bladder control. Of the remaining patients, 3 appeared to show some improvement in their neurological signs post-operatively, one was symptom-free at operation and remained so, one deteriorated markedly before operation and died in the post-operative period, one showed no change, and in the remaining one there was no follow-up.

A number of subsequent case reports also illustrate the progressive nature of the condition, and the poor results of late operation. Rand and Rand's (1960) book on intraspinal tumours in childhood contains two typical examples. The first, a boy, had progressive weakness of the legs from the age of 1 year and loss of sphincter control at the age of 4 years. Operation at the age of 7 years was not followed by any neurological improvement. The second, a girl, with motor and sensory changes as well as loss of sphincter control, was not benefited by surgery at the age of 8 years. Another case of interest was a 9-week-old infant, who had a sacral swelling since birth and two associated dermal sinuses. An intradural lipoma attached to the conus medullaris was continuous with the upper dermal sinus. The roots of the cauda equina were not involved. The lipoma was resected and no neurological deficit developed after operation.

Recently, Swanson and Barnett (1962) reviewed 9 cases of intradural lipomas in childhood. Four of these conform to lipoma of the cauda equina. In one child (Case 3) there were no associated signs at the age of 8 weeks, but by 4 years she had motor and bladder involvement. Another (Case 7) presented with a lumbosacral meningocele at the age of 5 months; because of the absence of neurological abnormality no operation was recommended. At the age of 10 years she had motor and urinary involvement. At operation in both these patients there was an extradural lipoma with an intradural extension involving the cauda equina.

As might be expected with a congenital lesion, lipomas of the cauda equina are essentially a paediatric problem. An unexpected contrast, however, is the interesting case of Di Biagio (1959). A 61-year-old woman had a swelling in the lumbar region since birth. It was not until the age of 53 that she developed weakness and sensory loss in the left leg, which became progressively worse. Subsequently the right leg was also affected and there was associated loss of bladder control. The lumbar swelling began to enlarge. The tendon jerks were pathologically brisk and there was a bilateral Babinski response. At operation the subcutaneous lipoma had an

intradural extension and was firmly bound up with the cauda equina. It was removed as far as possible. After operation there was a considerable improvement in her motor power, the sensory changes almost completely regressed, the abnormal tendon jerks became less brisk but the plantar response remained extensor.

Clinical Data

Twelve personal cases with typical features of lipoma of the cauda equina form the basis of this report. All have had detailed pre-operative neurological assessment and at least three months' post-operative follow-up. Three additional cases have been omitted because of inadequate post-operative follow-up and several others are still awaiting operation.

The first patient (Case 1), who drew our attention to this condition in September 1962, was a child previously thought to have a straightforward meningocele. However, there was a recent history of deterioration of bladder function and on examination she had an extensor plantar response and anaesthesia in the sacral distribution. Lipoma of the cauda equina was suspected and confirmed at operation.

The main clinical features of this and the subsequent 11 patients are summarized in Tables 1 and 2, and detailed clinical histories of three patients (Cases 1, 3, and 5) are given in the Appendix.*

Eight patients were male and four were female. There were no affected sibs nor was there any previous family history of a similar condition or of hydrocephalus or meningocele.

A swelling was present at birth in each case. An associated dermal sinus or a deep dimple was noted in four and a skin tag in an additional one. No neurological abnormality was noted at birth in any of the patients, and none developed hydrocephalus.

Onset of Symptoms. In infants it is often difficult to assess the exact age of onset of symptoms such as loss of sphincter control or sensory changes.

The earliest onset was in Case 3, who was examined in detail at birth and found to be normal. At the age of 3 weeks he had gross weakness of one leg. In contrast, Case 5 was symptom-free until the age of 3 years. Two patients (Cases 8 and 11) were still symptom-free at the time of operation.

Neurological Signs. The commonest neurological complications were motor weakness (9 patients) and

* Clinical histories of the remaining 9 cases are available from the author.

TABLE 1

Case No.	Sex	Age at Onset of Neurological Symptoms(yr.)	Age at Operation (yr.)	Duration of Follow-up (mth.)
1	F	2½	3	19
2	F	2	2 yr. 1 mth.	15
3	M	3 wk.	5 wk.	11
4 (Fig. 1)	F	2½	9 yr. 10 mth.	8
5	F	3½	3 yr. 7 mth.	7
6	F	1 yr. 9 mth.	11 yr. 9 mth.	5
7	M	1 yr. 6 mth.	5 yr. 8 mth.	4
8 (Fig. 2)	M	?	2 yr. 3 mth.	7
9	F	1	3 yr. 3 mth.	5
10	F	3 mth.	5 yr. 2 mth.	3
11	M	—	2 yr. 10 mth.	3
12	F	6 mth.	3 yr. 7 mth.	3

loss of bladder control (9 patients). The latter usually presented as dribbling incontinence, but 2 patients presented with retention with overflow and a number of others had excessive residual urine on catheterization. Difficulty with defaecation affected only 3 patients.]

Sensory loss in the sacral distribution was observed in 8 patients. The tendon reflexes in the lower limbs were diminished in 7, and an abnormal plantar response, either extensor or absent, was noted in 8 children.]

Urinary Tract Infection. There was evidence of infection in 8 patients and 4 of these had hydro-nephrosis on intravenous pyelography. The infection usually required prolonged antibiotic therapy for adequate control.

Skeletal Abnormality. [Asymmetry of the sacrum or agenesis of some of the sacral vertebrae] occurred in 6 patients. It could usually be detected on rectal examination and confirmed on radiography. In addition all the patients had a spina bifida of varying extent.]

Observations at Operation. [The subcutaneous lipoma varied in size and overlay the sacrum or lower lumbar region. In each case there was a deep stalk-like extension which passed intradurally through the gap of the spina bifida. It was continuous with the

TABLE 2
NEUROLOGICAL INVOLVEMENT ✓

Neurological Involvement	No. Affected	No. Improved by Operation	
		Definite	Doubtful
Motor paralysis . . .	9	5	1
Depressed tendon reflexes . . .	7	2	
Abnormal plantar responses . . .	8	1	
Sensory loss . . .	8		1
Loss of bladder control . . .	9	2	2
Loss of rectal control . . .	3	1

FIG. 1.—Case 4. Operative details. Child prone with head end to left. Shows lipoma attached to conus medullaris by stalk, which is angulated around a fibrous arch. A catheter has been passed under the arch. The dura is exposed after laminectomy.

conus medullaris and intimately connected with the nerves of the cauda equina. The lower end of the cord was never in its normal position but frequently as low as the first or second sacral vertebra. The roots of the cauda equina ran upwards from the conus medullaris and, after a variable course of up to 2 cm. or more, angled acutely downwards to their respective foramina.]

[At the upper limit of the spina bifida there was a firm fibrous or cartilaginous transverse arch around which the cord was acutely angulated. The liability to pressure on the cord at this site] could be demon-

FIG. 2.—Case 8. Operative details. Child in prone position with head end to the left. Shows lipoma attached by stalk to conus medullaris. Cauda equina nerves run in an upward (cephalad) direction.

strated by traction on the lipoma. Complete resection of the lipoma was usually not practicable because of the danger of damage to the cauda equina, but a fairly extensive removal was usually achieved. In addition a laminectomy of two or three vertebrae above the spina bifida, and a vertical incision of the underlying dura were found to be essential to abolish the acute angulation of the cord and to allow it to retract up the spinal canal towards its normal position. After this the cauda equina roots had a more normal downward or transverse course.

Post-operative Course. During the first post-operative week a temporary deterioration in bladder control was frequently observed, but this invariably recovered within a few days. Apart from a tendency for the subcutaneous collection of CSF, which needed aspiration, there were no other post-operative complications.

The follow-up period is in many instances still relatively short for full appraisal of improvement or deterioration. However, an improvement in motor function has occurred in 5 patients and possibly also in a sixth. In one patient (Case 5), the response was dramatic and she regained the ability to walk within a week of operation and subsequently progressed to complete normality of motor function. It is also of interest that though Case 8 was thought to be neurologically normal, there was a definite improvement in motor function after operation. In retrospect, however, his early history does suggest some delay in motor milestones. Depressed tendon jerks became normal in two children, and an extensor plantar response reverted to flexor in Case 1.

Bladder function was improved in only two instances and a temporary improvement occurred in two others. One patient had improvement in rectal control after operation, while another (Case 2) showed deterioration which was very slow to resolve.

Histology. The lipoma consisted of normal adipose tissue, with additional connective tissue and frequently small nerve trunks. There was no sinus tract apparent in any of the specimens, and no teratomatous elements were found.

Discussion

The dangers of this apparently benign lipoma are amply illustrated by the cases from the literature as well as the cases we have described. The chief condition from which one has to distinguish it is lumbar or sacral meningocele. [A lipoma of the cauda equina should be suspected in any lumbosacral swelling with normal skin cover and no associated hydrocephalus.]

[In the majority of cases the onset of motor or sphincter disturbances is fairly gradual and insidious. It may, however, be quite sudden and dramatic] as in two of our patients. One child (Case 3), who was normal at birth, developed severe weakness of one leg at 3 weeks of age. The other (Case 5), a 3-year-old girl, lost the ability to walk in the course of 24 hours.

Once neurological involvement has occurred there is no guarantee that even immediate operation will give complete resolution. The dramatic recovery in Case 5 may well be exceptional. Certainly in Case 3 the improvement was slow and is still not complete. The outlook for bladder function seems worse than for motor function and only two patients have shown a long-term improvement.

[The precise cause of the neurological disturbances is not easily determined. There are several possibilities.

(a) A traction lesion due to the cauda equina and the conus being densely adherent to the fat, which in turn is firmly fixed to lumbar or sacral skin.

(b) Compression of the cord, the conus, or the cauda equina by an extension of the lipoma within the canal.

(c) Pressure due to angulation of the terminal spinal cord around a cartilaginous or fibrous arch at the upper end of the spina bifida.

(d) A type of recurrent trauma due to the cord being dragged around this arch in movements of the trunk, comparable to traumatic ulnar neuritis.]

Direct downward traction of the cord is, in our view, unlikely to be the only important factor; nor is the amount of lipoma within the canal sufficiently great in most cases to cause direct compression. Direct pressure or recurrent trauma due to movement of the cord at the site of angulation seems to us an important aetiological factor.

There are two important aspects of the operation; removal of the lipoma and laminectomy above the lesion. Of the two, the former is the more dangerous and possibly less important. As much lipoma as possible is removed without damage to the cord, the conus, or the cauda equina. A large part of the fatty tissue can be excised without risk, but the central portion is so closely intermingled with neural elements that complete removal is not possible.

In addition to this fatty tissue invading the lower-most neural elements, there is sometimes an upward extension of the lipoma within the canal around the cord, either outside or within the dura. This might be a factor causing compression and it can be removed without difficulty. In fact, the only part of the fatty tissue that is difficult to remove and particularly susceptible to damage is the deep central

portion of the mass which is so densely bound to the lower part of the cord.

The purpose of removing two or three laminae is to release the cord from compression by the lowermost arch of fibrous tissue or cartilage at the upper end of the spina bifida, and to allow plenty of space for the cord to move upwards.

In each case the dura was incised the full length of the laminectomy, allowing the residual prominence of neural and lipomatous elements to lie at the top of the dural opening before closing the dura as far as possible around the remaining mass of tissue.

Clearly there is the possibility of damaging normally functioning nerve elements, and operation is not to be undertaken lightly. It would indeed be tragic to convert a normal child into one with some motor or sphincter weakness and it is certainly not much consolation to know that this paralysis might in due course have occurred spontaneously. Should the operation be deferred, however, the child must be examined at short intervals to detect the slightest neurological impairment. In addition, the parents must be given the strictest instructions to report any muscular weakness of the lower limbs, any tendency to trip or fall, any sign of abnormal gait, and any variation from normal bladder function.

We are convinced that in our hands it is wise to explore these lesions even before there are any signs of neurological involvement. In cases with established neurological abnormality the results of surgery may appear disappointing. However, it must be remembered that this is a progressive lesion. In these circumstances even an arrest of neurological deterioration would in itself justify operation.

We hope that the recognition of the potential dangers of leaving alone these apparently benign lesions will avert in others some of the tragic consequences that we have met in our own cases.

Summary

Twelve cases of lipoma of the cauda equina are described with detailed neurological assessment and post-operative follow-up ranging from 3 to 19 months.

The benign-looking subcutaneous lipoma is readily mistaken for a lumbosacral meningocele with normal skin cover. However, the stalk-like intradural extension of the lipoma is firmly adherent to the cauda equina and conus medullaris, and neurological complications are common. These affect mainly motor and bladder function.

Early diagnosis is imperative and careful operative treatment before the onset of neurological signs may prevent the development of neurological complications. In cases with established neurological

involvement, operation may in some instances produce improvement, while in others it may at least arrest the progression of the neurological deficit.

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Appendix

Case Summaries

Case 1. Trudy T. (born October 24, 1959). A swelling in the lumbosacral region had been present since birth. At 6 weeks of age the swelling was about 12 cm. in diameter and there was no neurological abnormality or hydrocephalus. She was operated on at 9 months of age, and a large lipoma was found in association with a meningocele. About two-thirds of the lipoma was removed. The post-operative course was uneventful, and motor development progressed normally. She was walking and climbing stairs by 14 months.

She was referred to the Children's Hospital, Sheffield, in September 1962 aged 2 years 11 months, with a three months' history of deterioration of urinary control, which had previously been considered completely normal. She had become incontinent and lost bladder sensation. In addition the swelling over the sacrum had recurred and slowly increased.

EXAMINATION. There was a large lipomatous swelling in the sacral region, with an overlying scar. The urinary bladder was grossly distended. Her gait and motor power were normal, but the plantar response was extensor on both sides. Anaesthesia was present over the saddle area. The anal reflex was negative.

After manual expression of her bladder there was still 150 ml. of residual urine on catheterization. Cystoscopy revealed a large trabeculated bladder with wide ureteric orifices. Retrograde pyelography on the right side

showed gross hydronephrosis and ureteric reflux. The left side was not catheterized.

On the basis of the sensory loss, the extensor plantar response and the deterioration in bladder function, a diagnosis of lipoma of the cauda equina was made.

OPERATION. October 29, 1962. A large lipomatous mass extended intradurally and was continuous with the conus medullaris. The subcutaneous part was adherent to the adjoining bone. The roots of the cauda equina were running in an upward direction before angling down towards their foramina. The lipoma was resected as far as possible.

POST-OPERATIVE COURSE. Eighteen days after operation the child asked for her potty and passed urine spontaneously. She repeated this next day and four times in the course of the following 24 hours, and no manual expression was necessary. Following that she developed dribbling incontinence again and only used the potty intermittently. She was only able to pass about 50 to 100 ml. spontaneously, and a further 200 ml. could be expressed. By the time of her discharge (December 3) she only needed expression every four to five hours. She was kept on maintenance sulphadimidine therapy for her persistent urinary tract infection.

FOLLOW-UP. Her motor power remained normal, and the plantar response became flexor. However, there was no real improvement in her urinary control. She was again requiring two-hourly expression. The urinary infection persisted in spite of courses of various antibiotics. She was readmitted in March 1963 for bladder washouts and the urinary infection was again brought under control.

In July 1963, a Y-V plasty of the bladderneck was performed. This did not prove successful but resulted in continuous incontinence. In November 1963, the left

ureter was joined to the right and a ureterostomy was performed. In March 1964 the urine was normal for the first time. Prophylactic sulphadimidine was again given. At her last visit in May 1964, 19 months after the spinal operation, she had not developed any further neurological abnormality and her urine was still free of infection.

COMMENT. A lipoma, present since birth, was partially removed at 9 months. Bladder symptoms appeared at 2½ years. Hydronephrosis was present, probably of longer duration than the symptoms suggest.

Resection of the lipoma led to only temporary improvement in bladder function and she subsequently needed ureterostomy.

Case 3. Michael P. (born August 1, 1963). A sacral swelling was noted at birth. A surgeon advised against operation because of the absence of neurological signs. At the age of 3 weeks his mother noted weakness of the left leg.

EXAMINATION. He was an active alert baby, with a subcutaneous lipomatous swelling over the sacrum (Fig. 3), approximately 7 cm. in diameter. There was a flaccid paralysis of the left leg with almost no active movement. The reflexes were asymmetrical. The withdrawal reflex was normal on the right but absent on the left. The knee-jerk was brisk on the right but absent on the left; the ankle-jerks were absent on both sides. The plantar response was extensor (normal) on the right, and absent on the left. The right leg appeared normal apart from intermittent spasm of the quadriceps and hip flexors, with associated increase in tone.

The tone in the anal sphincter was reduced and the anal reflex was negative. The bladder was not distended. He passed urine intermittently with a good stream, and there was no dribbling incontinence. It was difficult to assess his sensory function.

FIG. 3.—Case 3. Shows lumbosacral swelling.

OPERATION. September 4, 1963. The lipoma was continuous with the lower end of cord which was at the level of the second sacral vertebra. The cauda equina nerves ran upwards for about 2 cm. before angling down to their foramina. Laminectomy of L3 and L4 was performed and the lipoma dissected away as completely as possible. This allowed the cord to ride back up the canal.

POST-OPERATIVE COURSE. When reassessed two months later there was a marked improvement in his left leg. He was able to move the leg as a whole and to bend the knee. The foot, however, was still completely paralysed. Abduction of the hips was limited beyond 60°. The withdrawal reflex on the left was present but still sluggish compared to the right. His general neurological development was normal.

Eleven months after operation there was further improvement in the power of his left leg, but he still had marked paralysis below the knee and an inversion deformity of the left foot. There was no weakness of the right leg and sphincter control appeared normal.

COMMENT. A sacral lipoma was present at birth with no neurological involvement. Severe paralysis of the left leg was present by the third week. An improvement in motor function followed operation.

Case 5. Mary P. (born April 21, 1960). A swelling over the sacrum was noted at birth, with no associated abnormalities.

She was first seen at the Children's Hospital aged 1 month. The sacral swelling, slightly to the left of the midline, was diagnosed as a meningocele. No weakness was noted in her legs. The anal sphincter had normal tone and the urinary stream was said to be good. There was no evidence of hydrocephalus. In view of the absence of any complication operation was not recommended.

Reassessment at 1, 2, and 3 years of age revealed no abnormality apart from some increase in the size of the swelling, and operation was deferred.

She continued well until October 1963, aged 3½, when she quite suddenly lost the ability to walk, though she was still able to stand. Apart from a slight limp the previous day, no abnormality had been noted. She was admitted to the Children's Hospital, Sheffield, three days later.

There appeared to have been some further deterioration and she could only just stand with support. It was

difficult to assess individual muscles but there was a bilateral foot drop and also weakness of the hip extensors, and possibly other hip and knee muscles. There was no loss of sphincter control.

A diagnosis of lipoma of the cauda equina was made and urgent operation recommended.

OPERATION. November 6, 1963. A large subcutaneous lipoma (9 × 5 × 4 cm.) was found, with a stalk passing through a spina bifida defect of L5 and S1 vertebrae. A small meningocele was present in the depths of the lipoma. The lipoma passed through the meninges and was continuous with the lower end of the cord which was tethered down and reached practically to S1 level. The cauda equina nerves were passing upwards before turning acutely down again. After resection of the lipoma and a laminectomy of L4 vertebra, the cord could be freed and moved up at least 2 cm.

POST-OPERATIVE PROGRESS. Neurological examination on the first post-operative day revealed spontaneous movements of both legs. She was able to flex the knees and hips, in response to painful stimulation of her soles. No sensory loss was apparent but the perineal region was difficult to assess. The knee-jerks were absent, the ankle-jerks present but sluggish, and the plantar response flexor.

A week after operation there was a striking improvement. She was able to walk around holding onto things and could also walk with one hand held. She was still unable to stand without support. The reflexes were unchanged. Sphincter control was normal. One week later she was walking well without support, and was discharged.

At follow-up assessment three months later there was further improvement. She walked well for up to 100 to 150 yards before tiring. There was slight inversion of the right foot but no obvious limp. She was able to go up steps and to climb on and off a bed. The knee-jerks were present and fairly brisk, the ankle-jerks were more active than before, and the plantar response still flexor. Sphincter control was completely normal.

Seven months after operation her gait was normal and she was able to run, to climb stairs, and to ride a tricycle. She could stand on one leg without support. The tendon jerks were normal and the plantar response flexor. There was no sensory loss and sphincter control was normal.

COMMENT. A child, unable to walk or to stand without support, returned to normal after operation.