ON A REMARKABLE CASE
OF
EXTREME MUSCULAR ATROPHY
WITH
EXTENSIVE DISEASE OF THE SPINAL CORD.

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NOTES OF THE CASE BY DR. JOHNSON.

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JAMES CORMACK, æt. 32, a peasant, was admitted into Kilkenny County Hospital, May, 1867. Obiit October, 1867. The appearances then presented were these. The muscles of the shoulder and arm had almost altogether disappeared. The muscles of the forearm remained apparently unaffected. The muscles of the ball of each thumb were almost altogether gone, while the muscles of the fingers continued to be well developed. The muscles on the dorsum of each scapula were nearly gone also.

The various organs of the pelvis and the lower extremities

were well developed. The virile organs were, as the man's aspect indicated, of full proportion, complete and perfect. The thighs and legs were muscular and full, *especially the thighs*, the muscles of which were particularly well developed. Whatever cause had affected the nutrition of other parts did not appear to have extended to them. His height was about 6 ft. 2 in.

The heart's action was extremely perturbed and irregular, and very feeble. It jumped and fluttered in a very remarkable manner, and any exertion, such as the ascent of stairs or incline, was attended with dyspnœa. The disturbance appeared to be functional, as there was no sound indicative of organic change or lesion of the heart. Notwithstanding the attenuated state of the arm, the brachial artery could not be found, while the radial was easily felt at the wrist.

The history of his case, so far as it could be ascertained, was, that he had been inoculated with the natural pock when about a year old. That he had been a very fine and healthy infant up to this time. That he suffered much after the inoculation and screamed at night, and gave other evidences of severe indisposition, and that, when the eruption was at about its height, the left arm was observed suddenly to droop and become powerless, and soon afterwards the right; and that both hung by his sides powerless for some time. It was a considerable time before he appeared to acquire any power in them. From this period the muscles already mentioned ceased to be developed, or fell into a state of progressive atrophy. *At the same time* the heart's action became irregular and perturbed, and continued so ever after.

Such was the account given by the patient himself. I had doubts as to his accuracy; but, *after his death*, his mother corroborated all his statements in a manner so clear, *positive*, and circumstantial, that I am inclined to think her representations not unfounded. She asserts most positively that he was free from all defects up to the time of the inoculation. That he was then a remarkably fine healthy child, with full use of his arms. That they suddenly became powerless and drooped helplessly by his sides, and that at the same time she at once

became conscious of the excited and irregular action of the heart, which she had never observed before.

On opening the thorax, the pericardium contained a considerable quantity of serum, but the heart was of about normal size, and the lungs crepitating, and apparently perfectly healthy. It is much to be regretted that the autopsy, which was made under circumstances of peculiar difficulty, was unavoidably hurried.

It seems remarkable that the same cause which arrested the nutrition of certain pairs of muscles did not affect the development of parts farther from the nervous centres; *e. g.* the lower extremities, as well as the pelvic organs, were wholly unaffected; and it may be asked, why did the muscles of the forearm and those of the fingers escape the blight which affected, in so remarkable a manner, the muscles of the thumb as well as those of the scapula and humerus? or why did not all parts *below* the seat of lesion suffer?

It would almost seem as if nature had expended the powers arrested in the upper extremities upon the development of the lower limbs. These were muscular and well nourished to more than an ordinary degree, while the *length* of the femur and tibia was out of proportion to the trunk. The thorax was small, and the chest narrow. The left side of thorax was larger than the right, causing a difference of height in the shoulders. The impression conveyed, *primâ facie*, was that the right side had fallen in, after the absorption of empyema, but he denied ever having had any illness which would indicate such a condition; and on opening the chest there was no trace, either on pleura or in lung, of past empyema, or disease whatever.

The abdomen was of ordinary proportion and development, and presented no indications of disease or arrest of due nutrition. The genital organs were fully proportioned. Had the development of the entire frame been equal to that of the lower part, I should say the man would have been 6 ft. 6 in. or 7 ft. high.

There was no curvation of spine. The difference in height of shoulders was caused by difference in size in the opposite

sides of the thorax. This, I think, must either have been congenital, or resulted from unequal nutrition, as he advanced to maturity; as I find no traces of lesion or disease to account for it.

It is obvious that the cause or causes which arrested the growth of certain muscles (for his mother asserts positively that they never became developed from the time the arms first fell powerless) did not arrest the development of other organs, certainly not of the bones, and especially not of the long bones.

The case seems to present many points of interest, and some difficult of solution.

Examination of the spinal cord, with remarks.

By MR. LOCKHART CLARKE.

The greater part of the cervical and dorsal regions of the cord was placed in the museum of the College of Surgeons of Dublin, by Dr. Robert McDonnell, at the request of Dr. Johnson. In this part it appears that the *anterior* nerve-roots were in a state of extreme atrophy. Dr. Robert McDonnell has kindly furnished me with the following observations:—"The posterior nerve roots were normal; the anterior nerve-roots were in a state of extreme atrophy. The atrophy was quite marked and unmistakeable to the naked eye, but was made very striking on placing the portion of the cord examined side by side with a similar portion of a healthy cord. In the part examined microscopically by myself there were few nerve-tubules remaining in the roots."

I regret that only the lower third of the cervical enlargement was sent to me for examination. In this portion, however, I found the most unequivocal evidence of extensive lesions in both the white and grey substances. At its upper end the surface of the posterior columns was softened, and infiltrated with granular exudation, so that the posterior roots of the nerves were more or less destroyed, while the pia mater was thickened and also infiltrated with fluid exudation.

When this part of the cord was carefully hardened in

chromic acid, and transverse sections were examined, it was not difficult to perceive that extensive lesions and displacements of the grey substance had taken place. In Plate XI, fig. 1, the posterior horn on the left side has preserved its natural shape, with the gelatinous substance (*a*) forming a thin layer of paler grey substance at its extremity. On the right side, however, it was drawn backward into a long streak, between *a* and *a'*, toward the softened surface of the corresponding posterior white column. On either side, the cervix cornu (*e*) or neck of the posterior horn was more or less damaged in different sections, by a streak of disintegration; and in the anterior grey substance there was an extensive area of softening and disintegration. On the left side, this was confined chiefly to the central parts (*f*), where the paler areas indicate the advanced and more transparent state of disintegration, while the dark isolated dots indicate the masses of tissue which are not yet reduced to that condition, but which, nevertheless, are damaged in a greater or less degree. On the right side the lesions in the *central* parts of the anterior grey substance are somewhat less conspicuous, but we find a dilated blood-vessel (*b*) with its branches, bordered by a thin layer of exudation, and by the surrounding tissue in a state of disintegration. The anterior median fissure (*d*), which always transmits a large blood-vessel, was dilated and filled with granular exudation, together with the *débris* of fibres at the inner borders of the anterior columns, resulting from disintegration.

A little higher up the grey substance was still further damaged and displaced, as represented in fig. 2. Here we find the same kind of exudation and *débris* of nerve-fibres filling up the anterior median fissure (*d*), and an area of the same nature surrounding a blood-vessel on each side of the anterior commissure. On the left side nearly the whole central portion of the anterior grey substance (*f*) was in a state of softening and disintegration, and a streak of the same kind extended backward along the middle of the posterior horn. On the right side the grey substance was very much altered in shape as well as damaged in structure. The

posterior horn was misshapen, the gelatinous substance being divided into two separate parts, *a á*. Between the anterior horn *b* and the head of the posterior horn *a*, the grey substance (*f*) was drawn out into the lateral column and extensively damaged by softening and disintegration of its tissue. The *tractus intermedio-lateralis, c*, was elongated in the same direction.¹

Although some of the nerve-cells retained their normal size and appearance a great number were found to be in different states of atrophy and disintegration. In many instances the large cells of the anterior grey substance were reduced to at least the one-eighth or one-tenth of their ordinary size, and even to mere granular points; while in other instances, although not much reduced in size, they had lost their sharpness of outline and consisted of an uneven aggregation of granules. Dr. Robert McDonnell, who had also examined some parts of the cord, informs me that he "found abundant evidence of atrophy of the cells, although in some portions they were good."

In the ganglion which was sent me, belonging to the posterior nerve-roots, there was no apparent deviation from the normal state.

Remarks.—This is the eighth case of muscular atrophy in which I have examined the nervous centres supplying the affected muscles. In every case I have found unequivocal evidence of lesions of those centres, accompanied frequently by atrophy of particular parts of the grey substance, including the nerve-cells. Only one of these cases remains unpublished.² The various kinds of lesions and other abnormal appearances have been seen in my preparations by so many competent witnesses, both English and foreign, that no one now entertains the slightest doubt with regard to their

¹ The *tractus intermedio-lateralis*, which, when I discovered it in 1851, I so named from the fact of its being situated at the lateral boundary of the grey substance, and intermediate between the anterior and posterior horns, is a pale tract of grey substance connected with the spinal accessory nerve.

² The rest are as follow, with the dates and places of publication :

1. A remarkable case of muscular atrophy, with disintegration of the

existence. But there are some who think it not improbable, and there are others who are much inclined to believe, that these lesions and atrophies of the nervous centres and nerves are not primary affections, but consequences of the atrophy of the muscles which they supply. I cannot think that there is any good ground for this opinion. If the alterations in the nervous centres consisted only of *atrophy*, or diminution in size either of the grey or white substance, it might then, *à priori*, be justly considered as possible, or not improbable, that those alterations were the consequences of atrophy of the muscular tissue; and this supposition would be supported, to a certain extent, by the results of observation on cases in which limbs have been lost for a great number of years. Dr. Vulpian, of Paris, has recently published a very valuable article on this subject in its relation to the lesions and alterations which I have discovered in the nervous centres in cases of muscular atrophy.¹ He describes and represents the appearances that he found in the spinal cords of two persons in whom amputation had been performed at a very remote period. The first was a woman who at the age of grey substance of the spinal cord. Plate. Beale's 'Archives of Medicine,' No. IX.

2. On a case of muscular atrophy, with disintegration of the grey substance of the spinal cord and atrophy of the cells. Engravings. 'British and Foreign Medico-Chirurgical Review,' July, 1862.

3. Examination of the spinal cord in a case of "wasting palsy." Displacement, atrophy, and disintegration of the grey substance of the cord. Ibid., October, 1863. Engravings.

4. A case of muscular atrophy, with extensive displacement and disintegration of the grey substance of the cord, and degeneration of nerve-cells. Five Plates. Beale's 'Archives of Medicine,' No. XIII.

5. A case of muscular atrophy, with disintegration of the grey substance of the cord, and enormous dilatation of the blood-vessels. Plate. 'Medico-Chirurgical Transactions,' 1866.

6. Another case, with disintegration of the grey substance and lateral white column, and remarkable atrophy of the nerve-cells. Ibid., 1867. Plate.

¹ Influence de l'abolition des fonctions des nerfs sur la région de la moëlle épinière qui leur donne origine.—Examen de la moëlle épinière dans des cas d'amputation d'ancienne date, par A. Vulpian, 'Archives de physiologie normale et pathologique,' No. III, Mai—Juin, 1868.

twelve years suffered amputation of the right leg, and who died of cerebral apoplexy at the age of fifty-nine, that is, forty-seven years after the operation. All parts of the cord were examined, but scarcely any appreciable difference was observed between the two lateral halves, either in the cervical or dorsal region, or even in the superior part of the dorso-lumbar enlargement. In these regions there was at most only a very slight diminution of the white substance of the right half; but in the *lower* part of the lumbar enlargement, nearly to the end, the right half of the cord was evidently smaller than the left. The grey substance and anterior column were the parts most atrophied; and the *posterior* column seemed to be the only portion of the right side that escaped; but the nerve-cells retained their normal appearance, nor was there any new formation of connective tissue either in the grey or white substance. Neither by the naked eye nor by means of the microscope could any appreciable difference be discovered between the nerve-roots of the two sides.

The second case was a woman who died of pleuro-pneumonia of the left side in 1868, and whose left leg was amputated, in 1848, a little above the ankle. The leg and thigh of that side were more slender than those of the right. The nerves were below their normal size, but no alteration of their fibres was observable under the microscope. On external examination there was no visible alteration in the cord, and both the posterior and anterior nerve-roots were similar on the two sides. On making sections of the cord, however, it was evident that the left side was smaller than the right in every region. This alteration was very slight at the lower extremity of the dorso-lumbar enlargement. A little higher up, but still below the middle of that enlargement, the diminution of the left side was much more conspicuous; while at its middle or thickest part the left side resumed its regular form. The anterior horn and white column, however, were a little smaller than those of the opposite side. In the dorsal region the only diminution of size was in the left anterior column; "but a more attentive examination," says

M. Vulpian, "shows that in the anterior grey substance on the left side there are three areas filled with a transparent substance, which in some places was a little granular,—areas which are evidently produced by an alteration of structure similar to that which Mr. Lockhart Clarke has discovered in different diseases, and amongst others, in progressive muscular atrophy." In the inferior part of the *cervical* enlargement there was diminution only of the left anterior column; but in the *middle* of that enlargement the whole of the white substance of that side had undergone a remarkable reduction in size, while the grey substance was but little changed.

The first opportunity I had of ascertaining whether amputation of a limb was followed, after many years, by atrophy of that portion of the cord from which it derives its nerves, was afforded me by my friend Dr. Dickinson, of St. George's Hospital, who very kindly gave me a portion of the lumbar enlargement of the cord of a seaman, aged seventy-four, whose left thigh had been amputated fifty-three years ago, in consequence of a cannon shot received in an action with the Danes in 1812. The man died in 1865. In the portion of the cord which I examined there was no difference in the size of the two lateral halves, as may be seen in fig. 3, Plate XI, which is an exact outline of the section taken by means of a camera lucida. The only difference was a very slight diminution of the left lateral grey substance (*g*); and the cells it contained were certainly less numerous and less distinct; but there was an entire absence of any alteration resembling the lesions and disintegrations which I have shown to exist in muscular atrophy. Dr. Dickinson, who also examined this portion as well as other portions of the cord, found no alteration in the lower lumbar region; but higher up in the lumbar and in the dorsal region there was marked diminution of the left posterior column, with an increase of connective tissue. The sciatic nerve, which he also examined, terminated at the stump in a large bulbous extremity, but the trunk of the nerve presented to the naked eye little or no alteration. From transverse sections of both sciatic nerves, which he prepared according to my method, and which he was kind enough to lend me, I

have made the drawings represented in figs. 4 and 5, Plate XI. Fig. 4 represents the ultimate tubules or fibres of the nerve in the sound leg, magnified 670 diameters; and fig. 6 represents those of the opposite stump. In the latter the originally large tubules were, for the most part, reduced to a number of small irregular areas surrounded by a close network of connective tissue, which contained a multitude of granules, resulting apparently from the *débris* or atrophy of the medullary sheaths or white substance.

Another case collected by Dr. Dickinson was that of a seaman who had his left arm amputated, two inches from the shoulder-joint, in the year 1842, and who died in 1865. From preparations which Dr. Dickinson kindly lent me I made the sketches represented in figures 6 and 7, Plate XI. In the cervical enlargement (fig. 6) no appreciable difference was observable in the dimensions either of the grey substance or the antero-lateral column (*h h'*) of the left side; nor was there any trace of lesion or disintegration; but the posterior column (*i i'*) was reduced to about two thirds the size of its fellow of the opposite side, and the fissure which, in this region, divides the column into two portions, contained an unusual quantity of connective tissue. Similar alterations were observed in the medulla oblongata, on a level with the lower part of the decussation of the anterior pyramids, as shown at *i i'*, fig. 7. Above this point the posterior column resumed its normal dimensions.¹

Since, therefore, both the portion of the cord and the nerves which supplied a lost limb will undergo, after a number of years, a very decided atrophy—although, apparently, in different places, to a very different extent—it is not improbable that *atrophy* of muscles, progressing through a lengthened period, might give rise, through partial abolition of the functions of the nerves and nervous centres which supply them, to *simple atrophy* of these parts. But it is highly improbable that muscular atrophy is the cause of

¹ In the forthcoming number (November, 1868) of the 'Journal of Anatomy and Physiology,' a full account of Dr. Dickinson's investigations will be published.

those lesions of structure, of those morbid exudations, those remarkable areas of transparent fluid or granular disintegration, or of that enormous dilatation of blood-vessels, which I have discovered in the cords of so many cases of this disease; and the absence of such lesions in cases of lost limbs is strongly in favour of this conclusion. It is true, as I have already shown, that M. Vulpian detected in one solitary spot what appeared to him to be three small areas of disintegration. This, however, not only seems to be an exceptional instance, but was found far distant from that portion of the cord which supplied nerves to the lost limb. It is evident that M. Vulpian laid no stress upon this fact, for he concludes his examinations in the following words: "De plus, et c'est ce que M. Lockhart Clarke a déjà mis en évidence, il n'y a aucune analogie entre les lésions qu'il a décrites dans l'atrophie musculaire progressive et cette simple diminution de volume des diverses parties de la moelle constatée dans des cas d'amputation d'ancienne date. *Nous pouvons donc conclure que les lésions médullaires de l'atrophie musculaire reconnaissent une autre cause que la seule paralysie des muscles et des nerfs.*"

There is abundant evidence to prove that a morbid condition of nerves depending on pressure, wounds, or some other kind of irritation, is the cause of various diseases of the skin, and even ulceration of the subjacent tissues. The writings of Henle,¹ Rayer,² Baerensprung,³ Notta,⁴ Swan,⁵ Parrot,⁶ Charcot,⁷ Paget,⁸ Earle,⁹ and others,

¹ Henle, 'Handbuch Ration. Pathologie.'

² Rayer, 'Maladies de la Peau.'

³ Baerensprung, 'Annalen der Charité Krankenhaus zur Berlin.'

⁴ Notta, "Mémoire sur les lésions fonctionnelles qui sont sous la dépendance des névralgies," 'Archives Generales de Méd.,' 1854.

⁵ Swan, 'Diseases and Injuries of the Nerves.'

⁶ Parrot, "Consideration sur le zona," 'Union Médicale,' Mars, 1856.

⁷ Charcot, "Note sur quelques cas d'affection de la Peau dépendant d'une influence du système nerveux," 'Journal de Physiologie,' tome deuxième, 1859.

⁸ Paget, 'Lectures on Surgical Pathology.'

⁹ 'Medico-Chirurgical Transactions,' vol. vii.

record numerous instances of this kind. It is well known how commonly neuralgia is followed and accompanied by *herpes*, which sometimes takes the course of particular nerves, as the intercostal or lumbar, and Dr. Anstie has shown that it is sometimes followed by erysipelas. Dr. Hugh Ley long since remarked that leech bites induce erythema and ulcers in the skin of neuralgic subjects. Rayet records a very interesting case in which traumatic neuralgia, arising from a gunshot-wound in the thigh, was accompanied at intervals by herpetic eruptions on the skin. Rouget has related a similar case.¹ In like manner the process of nutrition in muscles may be perverted and impaired by disease or injury of their nerves. Every practitioner of experience must have met with cases in which groups of muscles have wasted, sometimes to a remarkable degree, from this cause alone. Two striking instances recently came under my own observation. One was a boy, who slipped from the edge of a high stool on which he was rocking himself, and fell heavily to the ground on one side of the sacrum and of the lower part of the spinal column. For some days he complained very little; but, soon after, the glutei muscles, as well as those of the loins on that side, rapidly wasted to an extraordinary degree, so as to cause lateral curvature of the spinal column by the superior force of the corresponding muscles of the opposite side. The other case was a gentleman who was knocked down by a cab, one of the wheels passing over the *front* of his shoulder. The swelling for some time was very great, but when this subsided the muscles of the shoulder rapidly wasted. The deltoid of this patient, who was a powerful and muscular man, almost wholly disappeared, so that the outer part of the shoulder was perfectly flat, and even somewhat sunk below the acromion process, as in dislocation of the head of the humerus into the axilla. Both these cases rapidly recovered by the application of blisters to the affected and tender parts.

Now, if functional derangement or irritation of nerves will

¹ See also a paper by Dr. Woakes, in the 'Journal of Cutaneous Medicine,' 1867.

give rise to atrophy of the muscles which they supply, it is reasonable to infer that some kind of functional derangement of the nervous centres, which give origin to those nerves, may be followed by similar effects ; so that although there are the strongest grounds for believing that the lesions of *structure* which I have described in the nervous centres of cases of muscular atrophy are the cause of that atrophy, it does not therefore follow that every case of muscular atrophy is the consequence of those *structural* lesions, or even of *functional* disturbance of the nervous centres, since it may be due to affections of the nerves alone.

In the history of the singular case which forms the subject of this communication we learn that after inoculation, at the age of twelve months, the child screamed at night and gave evidence of severe indisposition ; that when the eruption was at its height the left arm was observed suddenly to droop and become powerless ; that, soon after, the right arm followed the same course ; and that it was long before the patient seemed to acquire any power in them. At the same time the heart's action became irregular and continued so ever after. These symptoms were followed by atrophy of the muscles of the arms and shoulders. If these statements be true, they point in a very remarkable manner to disease of the spinal cord as the cause of the muscular atrophy ; for it is evident, 1st, that the pustules of inoculation on the cutaneous surface of the arms could have had no share in paralysing the muscles, except by inducing some morbid change in that portion of the cord from which their motor nerves were derived ; 2nd, that they could have had no share in causing *atrophy* of those muscles, and of the muscles about the chest and back, except through the same channel.

M. Vulpian performed some experiments to ascertain whether any effects were produced on the nutrition of muscles by injuries inflicted on the grey substance of the spinal cord. He found that these injuries were not followed by atrophy of the muscles, which receive their nerves from the injured part ; and he therefore concludes that if there be a close relation of causality between the lesions of the

cord and those of the muscles, in muscular atrophy, we may presume that the influence of the spinal lesions depends on their peculiar irritating nature. The observations of Brown-Séquard confirm this opinion. In commenting on M. Charcot's cases of affections of the skin depending on the influence of the nervous system, he shows the difference of effects between *simple section*, or *cessation of action*, of nerves, and their *irritation*. Simple section of a nerve is not followed by any other alteration of nutrition than slow atrophy of the paralysed parts. It is the same with the spinal cord. He has seen animals live for months after section of the spinal cord without presenting any other lesion of nutrition than a slow atrophy of the paralysed parts; whereas, in two cases in which exostoses formed, at the point where the cord was divided, and compressed its lower end, considerable atrophy of the muscles supervened in the course of five or six *days*, with gangrenous ulceration over the sacrum and some parts of the thigh.¹

In making these remarks on the cause of muscular atrophy, I have been influenced solely by the great preponderance of evidence in favour of one view rather than the other. I have not been contending for a favorite theory; and as my object is solely to arrive at the truth, I would strongly urge a searching investigation into every case that may seem capable of throwing light on this important subject, until every question of doubt be definitively settled.

¹ 'Journal de Physiologie,' tome deuxième, 1859, p. 112..

EXPLANATION OF PLATE XI.

FIG. 1. Transverse section of the grey substance of the cord in the cervical enlargement; *a a'*, the gelatinous substance of the posterior cornu; *b b*, the anterior cornua; *c*, the tractus intermedio-lateralis; *d*, granular exudation filling up the anterior median fissure; *e*, the cervix cornu or neck of the posterior horn; *θ*, a dilated blood-vessel in the middle of the anterior grey substance; disintegration of the central part of the grey substance of left side; the lighter parts on the opposite side indicate the same kind of alterations.

Fig. 2. The same letters indicate the same parts; on the right side the lateral grey substance (*f*), and the tractus intermedio-lateralis (*c*) are drawn out into the lateral column.

Fig. 3. Transverse section of the lumbar enlargement of the spinal cord of a seaman who suffered amputation of the left leg fifty-three years before his death. Only a trifling degree of atrophy of the left anterior grey substance was discovered at *g*.

Fig. 4. Part of a transverse section of the great sciatic nerve of the sound limb, from the same man, showing the size and general appearance of the cut-ends of the nerve-tubes with the axis cylinders in their centres, and united by connective tissue; magnified 670 diameters.

Fig. 5. Part of a similar section from the great sciatic nerve in the stump of the amputated leg, showing how the originally large nerve-tubes have been reduced to a number of small irregular spaces, with intervening granules; magnified 670 diameters.

Fig. 6. Transverse section of the cervical enlargement of the spinal cord, from a seaman who had suffered amputation of his left arm two inches from the shoulder twenty-three years before his death. The only part atrophied was the posterior column on the left side, *i i'*.

Fig. 7. Transverse section of the lower part of the medulla oblongata, a little above the points of the anterior pyramids from the same man; *b b*, anterior grey cornu; *k, k*, caput cornu posterioris or grey tubercle of Rolando; *l*, central grey substance; *m, m*, anterior pyramids, with their decussating fibres between the anterior cornua, *b b*; as in preceding figure, the left posterior column *i i'* is the only part atrophied.