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THE SURGICAL TREATMENT OF DYSTONIC TORTICOLLIS

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In a previous catamnestic analysis of 50 patients operated upon for dystonic torticollis at the Department of Neurosurgery of the Serafimer Hospital, Stockholm (Professor H. Olivecrona) during the years 1926–1944, the results of intradural section of ventral cervical roots and/or extradural section of the accessory nerve were presented (*Lundberg* 1946). This analysis suggested certain modifications of the standard procedure. These modifications were later applied to 22 cases treated at the Department of Neurosurgery, Lund, in 1946–1957. The latter cases have been carefully followed up by questionnaires and personal examination.

The main purpose of the present investigation was to assess, on the basis of two relatively large series, the end-results of a surgical technique hitherto generally used in the treatment of torticollis and, secondly, to estimate the value of certain modifications used in the second series.

PATHOGENETIC VIEWS

The condition generally known as torticollis spastica is characterized by a more or less continuous irregular activity of the neck musculature, giving rise to compulsive movements or posture of the head. In addition various types of tremor are not uncommon. It is not correct to call this condition spasticity, dystonia being a more adequate term (see *Herz & Glaser* 1949). The activity usually occurs when the head is held upright and usually disappears when the patient is lying. It is accentuated considerably by mental stress and generally reduced if the patient supports the chin with the hand.

The etiology of torticollis can only be briefly discussed here. The reader is referred to the extensive survey of *Patterson & Little* (1943). In their report of 103 cases of torticollis *Patterson & Little* expressed the view that torticollis is rarely psychogenic and that many cases regarded as being of functional origin do not satisfy the definition of spasmodic torticollis: "An involuntary hyperkinesis manifesting itself by mobile, tonic or clonic spasm of the neck musculature, and producing a more or less stereotyped deviation of the head into an abnormal position,

the chin being rotated to one side or the head bent directly forward (antecollis) or backward (retrocollis)". It is well known that mental strain and tension can contribute considerably to the symptoms of torticollis. This is compatible with *Foerster*'s (1921) opinion that lesions of the extrapyramidal system are often asymptomatic until symptoms are precipitated by mental or other factors.

Foltz et al. (1959) described 7 cases of torticollis in *Macaca Mulatta* produced by stereotactic lesions placed paramedially in the brain stem. All the lesions were placed in the middle of the tegmentum mesencephali at the level of the decussatio brachii conjunctivi involving the brachium conjunctivum, fasciculus longitudinalis medialis and formatio reticularis medialis. Lesions in the last mentioned structure were regarded as the most important in the causation of torticollis. It appears that the torticollis produced by *Foltz* closely resembled that seen in human beings, in contrast to the picture produced by lesions of the brain stem of monkeys by *Kemberling et al.* (1952). It is noteworthy that the dystonic activity often disappeared when the animals were left alone, but could be provoked by measures making the animals restless or uneasy. In addition, the activity decreased when the animals had become accustomed to a situation that had formerly annoyed them, but increased as soon as they were irritated by new measures.

REVIEW OF THERAPEUTIC METHODS

1. *Myotomy.* Myotomy was used as early as 1641 by the German *Isaac Minnius* and was recommended later by *Kocher, Miculicz* and *de Quervain* at the end of the nineteenth century (*de Quervain* 1896). This method has since been abandoned because it does not control the dystonic activity.

2. *Division of peripheral nerves.* Division of the accessory nerve was first described in 1834 by *Bujalski* (according to *Patterson & Little* 1943). *Keen* (1891) severed the posterior rami of the three uppermost cervical segments bilaterally. *Finney & Hughson* (1925) extended this method to include extradural division of the accessory nerve on both sides and improved upon the method of dissecting the nerves.

3. *Intradural division of cervical roots.* The first operation of this kind was performed in 1902 by *Cushing.* He divided the second ventral root on the left side and the corresponding dorsal root on the right (Cushing's comment to *McKenzie*'s article 1924). Later occasional reports of unilateral division of roots were published by *Taylor* (1915), *Foerster* (1933) and *McKenzie* (1924). The first to divide several roots on both sides was *Coleman* (1927).

Unilateral division of *dorsal* cervical roots for spasmodic torticollis was first performed by *Taylor* (1915) and was later suggested by *McKenzie* (1924) and *Frazier* (1930). The theoretical background was the work of *Sherrington* who stressed the importance of the afferent impulses for muscular activity. Only a few cases treated in this way have been published, including one by *Coleman* (1927). *McKenzie* (1955) considered deafferentation useless, on the basis of poor results obtained in two cases.

Dandy (1930) reported the results of bilateral division of the three uppermost ventral and dorsal roots in 8 cases (see Table 3). He concluded that deafferentation was superfluous and suggested bilateral intradural division of the three uppermost ventral roots as a standard procedure, combined if necessary with division of the ac-

cessory nerve. In addition, denervation should in some cases be extended to include the 4th to the 7th cervical segment inclusive, by resection of the posterior rami according to Finney's method.

After the reports of *Dandy* (1930) and *Olivecrona* (1931) the standard method suggested by *Dandy* has been the one most commonly used. *Putnam* (1946) added the proposal that the 4th ventral root could be divided on both sides without impairing respiration.

Five recent reports on fairly large series of operations for torticollis deserve mention. For a summary of the results, the reader is referred to Table 3. Only a few complementary notes will be given here.

Adson et al. (1946) described 21 cases in which they only divided the accessory nerve with no improvement in 4, improvement in 16 and recovery in 1. Intradural section of roots gave partly satisfactory results. In some cases these authors found that denervation caused instability of the cervical spine which required stabilisation by vertebro-occipital fusion. In one case this operation was fatal because the bone transplant slipped and compressed the medulla. Two other deaths were recorded, one due to ligation of a large spinal artery and one where division of the 4th ventral roots had caused paralysis of the diaphragm, which was complicated by pneumonia.

Putnam et al. (1949) pointed out that their 18 cases had all been previously treated by different conservative methods without or with only insignificant and temporary effect.

Poppen & Martinez-Niochet (1951) included in their material 19 patients suffering from other neurological diseases besides torticollis, particularly dyskinesia of other parts. The results were described as poor in 14 cases and satisfactory in 22. One patient died from air embolism. Postmortem examination revealed diffuse cortical necrosis, a focal lesion in the mesencephalon and destruction of nerve cells in the dentate nuclei.

Wycis & Moore (1954) divided the 4th ventral root bilaterally in one case without causing paralysis of the diaphragm. They stated that 8 of their patients made a complete recovery and 1 showed considerable improvement. The follow-up time, however, appeared to be short.

McKenzie (1955) described the anatomy of anastomoses to the first motor root and stressed the importance of not overlooking these anastomoses. His result were given as excellent in 10 cases and not quite satisfactory in 2. Of the former group, at least 2 patients were completely free of symptoms when seen 2 and 9 years respectively after operation.

Törmä & Troupp (1958) followed up 16 of their 29 cases for more than 1 year. Two patients out of 18 treated by intradural division of ventral roots died in association with the operation. One patient died from a ruptured aneurysm of the posterior communicating artery and the other probably from an undiagnosed postoperative hematoma.

As to treatment other than surgical, the reader is again referred to *Patterson & Little* (1943). After an exhaustive survey and discussion of the literature on torticollis, they compared the results of different methods on the basis of a follow-up study of 103 cases. They concluded that surgical treatment is superior to other methods.

SERIES FROM SERAFIMER HOSPITAL

In 1926–1944 58 patients had been operated upon with extradural division of the accessory nerve and/or intradural division of cervical roots in different combinations (only accessory nerve in 11, unilateral root section in 3, bilateral section of ventral roots in 30 cases, bilateral section of ventral and dorsal roots in 6). In 3 cases the 4th ventral root was divided on one side, in all the others the operation included only the 3 upper roots. The operations claimed no mortality. The patients were later reviewed by questionnaires and 50 co-operated. The average follow-up time was 8 years after the operation (at most 16 years, at least 1½ years).

According to the questionnaires, the symptoms of torticollis had abated considerably or disappeared in 80 per cent. If sequelae ascribable to the operation were included, partial or complete improvement was recorded in 66 per cent. Of 46 patients, in whom the accessory nerve had been divided, 46 per cent reported severe or moderate symptoms in the operated shoulder and arm. Working capacity was unimpaired in 30 per cent, moderately impaired in 46 per cent, and 24 per cent of the patients were severely or completely disabled. The loss of working capacity was ascribed partly or entirely to persistent symptoms of torticollis in 31 per cent, insufficiency and pain in the musculature of the neck in 28 per cent, and insufficiency and pain in one shoulder and arm in 41 per cent.

The material did not permit comparisons of the results with regard to different surgical methods. It was concluded that the poor results could probably be ascribed in about equal degree to persistent torticollis, paralysis of the neck muscles after division of ventral roots and paralysis of the trapezius after division of the accessory nerve.

Different possibilities of improving the results were discussed:

1) Treatment in several stages with stepwise denervation of the muscles involved.

2) Deafferentation of the neck musculature alternately to extensive deefferentation as the first measure.

3) Denervation of the sternomastoid muscle without interference with the trapezius innervation.

GENERAL CONSIDERATIONS

The results of dividing nerves and roots in torticollis dystonica must be evaluated with respect both to the effect on the symptoms of torti-

collis and to the sequelae which are a direct consequence of denervation. Persistent symptoms of torticollis may be due to insufficient denervation, to reinnervation and to spread of the dystonic activity to new muscle groups. An attempt was made to assess to what extent these factors might have been responsible for the poor results in the Serafimer series (see above). Decisive conclusions in this respect can, however, only be made on the basis of personal examination of the patients.

The following categories of undesired side-effects can be distinguished.

1. *Symptoms of insufficiency of the neck muscles.* For some time after bilateral division of the 3 uppermost ventral roots it is difficult for the patient to hold his head upright, besides which the mobility may be limited and a feeling of tiredness and pain because of muscular tension occur. As a rule, the patients learn how to balance the head and in favourable cases the symptoms of muscle weakness disappear. But sometimes weakness, stiffness and pain persist.

2. *Secondary changes of the spine:* fixed malposition and spondylosis may develop if denervation interferes with the muscle balance. Torticollis as such, however, may have the same effect.

3. *Paralysis of the trapezius* is often without any therapeutic effect in torticollis and must therefore be regarded as an undesirable complication.[1] According to *Patterson & Little* (1943) the contralateral trapezius is involved in about 8 per cent and the homolateral in about 18 per cent, while the corresponding figures for the sternomastoid muscle is 47 and 19 per cent respectively. On denervation of the contralateral sternomastoid it should therefore be preferable to preserve the innervation of the trapezius in most cases.

4. *Paralysis of the diaphragm* can occur after division of the 4th ventral root, but if this is only done on one side, the risk of respiratory insufficiency appears to be small.

5. *Paralysis of the outer larynx muscles* and the muscles of the hyoid bone can cause postoperative dysphagia. This is favoured by difficulty

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[1] *Nordén* (1946) pointed out that injury to the accessory nerve sometimes gives rise to considerable symptoms because of paralysis of the trapezius. In addition to the well-known symptoms of paralysis which at least among manual labourers may cause a considerable loss of working capacity, pain in the shoulder and the arm was reported in 14 of 16 cases. *Nordén* believed this pain to be due to tension in the plexus brachialis because of a drooping shoulder. The significance of this complication appears to have escaped attention in the literature on torticollis.

in holding the chin forward because of paralysis of the extensors of the neck and the sternomastoid muscle.

6. *Symptoms of anesthesia following division of dorsal roots.*

On examining the patients in the present series it was attemped to assess to what extent the results of operations were influenced by the abovemenioned complications.

PRESENT MATERIAL

In the Lund series the cases were selected according to the following criteria:

1) History of more than half a year (except in a few very severe cases).

2) Conservative treatment had proved unsuccessful.

3) At the first examination the patient was informed of the risks involved in the operation and of its mutilating nature. Only those patients, who after due information definitely requested operation, were accepted, usually not until after they had been observed for some time.

4) Old patients with signs of cerebral arteriosclerosis were not accepted for operation.

5) Neither were any patients accepted with marked dystonia or dyskinesia of other types; one of the patients developed such symptoms later.

None of the patients showed any neurological signs apart from the torticollis. One reported slight weakness of the right hand for a day or so before the onset of the torticollis and one had had transient facial paresis.

As regards diseases of possible causal importance, one patient had had a fracture of the base of the skull 1 year before the onset of the torticollis, one had had severe pertussis 1 month before the onset and one had had meningo-encephalitis in association with parotitis 3 years before the onset. Two of the patients reported torticollis in other members of the family: one had a brother and a son with moderate and severe torticollis respectively, the other had a sister with this disease. In addition 2 sisters with typical torticollis dystonica were operated upon, but they are not included in the present material because they have not been followed up long enough.

The main reason why the patients wanted to be operated upon was the unnatural position of the head. It was moderate in 9 cases and severe in 12. In 4 the involuntary movements of the head were moderate and in 5 they were severe. The malposition or movement of the head was predominantly rotation in one direction in 16 cases (to the right in 10, to the left in 6). Alternating turning of the head in both directions was observed in 2 cases. The head was bent backwards (retrocollis) in 1 case and mainly tilted to one side in 2 cases.

The patients' ages when first admitted were 20–65 years (average 41 years). 13 of the patients were females and 10 were males. The patients had had the disease for 4 months to 13 years (average 5 years) at the time of the first operation.

The overall average duration of hospitalization of patients was 40 days and postoperative convalescence 9 months.

The interval between the operation and the review was on the average 6½ years (range 1 to 13 years). In 3 cases it was less than 2 years.

METHODS

Operative methods. The main principle was to denervate the sternomastoid muscle on one side first and if that failed to give relief, intradural division of cervical roots was done in a second stage.

In 18 cases the sternomastoid muscle was denervated in the following way: After local anesthesia the incision was made along the anterior edge of the muscle. The accessory nerve was exposed to the edge of the trapezius and all branches to the m. sternomastoideus were extirpated in their entirety. To prevent regeneration, in 14 cases a 3–4 cm. long cylinder of tantalum foil (0.02 mm. thick) was placed around that part of the accessory nerve medial to and behind the sternomastoid muscle.

In 18 cases intradural root section was done after laminectomy of the 3–4 uppermost cervical vertebrae under general anesthesia with the patient in the prone position. In 9 of these cases not only the 3 uppermost motor roots were divided but also the 4th motor root on that side to which the head was turned and in 3 cases the cephalad half of the 4th root bilaterally. As a rule, the first ventral root could be approached without extirpation of the occipital bone. Division of dorsal roots and denervation of the sternomastoid muscle was done in 3 cases (C_2–C_4 in 2 cases, C_1–C_4 in 1 case). Denervation of the sternomastoid was the only operation in 4 cases.

In one case in which troublesome dystonia persisted despite root section and denervation of the sternomastoid muscle, it was decided to try to divide the rami posteriores below C_4 on one side. A search of the literature failed to reveal any description of the operative technique. After preparatory studies on corpses the following procedure was used: The skin was incised just behind the anterior edge of the trapezius and carefully dissected from the superficial fascia in order to avoid injury to the accessory nerve. The accessory nerve was identified by electric stimulation and after an incision had been made in the fascia parallel to the nerve, the latter could be prepared and held aside. The transverse processes were approached between the splenius and levator scapulae; the transverso-occipitalis (= semispinalis capitis) was divided exposing the loose layer between this muscle and the semispinalis cervicis. The posterior tubercles of the transverse processes were used as orientation points. Medially and posterior to these tubercles 3 nerves were encountered and identified as probably being rami posteriores 4–6. They were divided and the proximal nerve endings were covered with small plastic caps, which were fastened with silver clips.

Questionnaires. The patients were re-examined by questionnaires formulated to yield data on persistent or recurrent symptoms of dystonia and insufficiency of the denervated musculature. The answers obtained were compared and supplemented by data from the annual reports of their condition, which the patients send regularly to the department, and with results of the objective findings obtained at the follow-up examinations. The symptoms of muscular insufficiency were then grouped according to whether they were due to division of cervical roots (disturbed mobility and pain in the neck and dysphagia) or symptoms due to paralysis of the trapezius (disturbed mobility and pain in one shoulder and arm); only symptoms that were not present before the operation were included.

Objective re-examination included functional examination of the neck and shoulder musculature. 20 patients were examined according to a standardized scheme and by the same examiner and within a period of 1 year. The strength of

the different groups of neck muscles was tested according to *Kendall & Kendall* (1949). The active and passive mobility of the neck was also examined. The results were recorded according to a 4-grade scale.

RESULTS

There was no operative mortality. Neither were there any serious postoperative complications, apart from those due to neck muscle paresis.

Transient complications.

Paresis of diaphragm. The 4th ventral root was divided completely or partially in 9 cases. Postoperative roentgen examination of the diaphragm in 5 cases showed paresis in 2. In no instance were the symptoms or signs of respiratory insufficiency severe permanent.

Difficulty in balancing the head was common during the early postoperative period, but the patients soon learned to hold the head upright.

Dysphagia of a transient nature occurred in 4 cases during the postoperative period.

Permanent complications.

Permanent slight swallowing difficulties were noted in 5 cases and moderate to severe difficulties in 2. One of these patients has to hold the chin with the hand to be able to drink, and to tilt the head to one side every time she swallows. The other patient finds it difficult to swallow liquid food. Both these patients have signs of slight paresis of the infrahyoid musculature and decreased lordosis of the neck with indrawn chin. The latter complication was observed in 9 patients (only 2 of whom had no swallowing difficulties at all).

Paresis of the cervical muscles is a desired effect of the operation. If very severe, *i.e.*, if the patient cannot balance the head, it is an undesirable complication which did not occur in any of our cases. Inability to raise the head against gravity was noted in no case on backward bending of the neck (which is the most important function in this connection), in 1 case on forward bending of the head and in 1 on sideward tilt. Moderate weakness was noted in 4 cases in backward bending, in 10 on forward bending and in 4 on tilting of the head to one side and 2 on tilting to both sides. This degree of weakness was further noted in 7 cases on rotating the head (5 to both sides, 2 to one side).

Atrophy in the posterior musculature of the neck was moderate in 6 cases (unilateral in 2) and severe in 1 (unilateral). Differences in atrophy between right and left were not related to the number of roots divided. The atrophy of the sternomastoid muscle after division of the accessory nerve corresponded to the paresis (see below). This also applies to the trapezius muscle. None of the patients found the atrophy disturbing from a cosmetic point of view.

Scoliosis occured postoperatively in 6 cases. It was severe in 2 and moderate in 4. Two of these patients had been examined roentgenographically before operation and in neither had examination revealed any abnormality of the cervical spine.

The mobility of the neck was normal or practically normal in all directions in 9 patients. In 5 mobility was assessed as slightly and in 6 moderately reduced, *i.e.*, about half of normal mobility (3 of them could not turn the head in one direction and 2 could not bend it back at all). The passive mobility was practically equal to the active mobility.

Pain occurred after operation in 5 of the patients. The pain involved the neck or back of the head and as a rule, it was strikingly correlated with the dystonic muscular activity, which was preserved in varying degree in this group.

Symptoms from the shoulder and arm after division of the accessory nerve (total in 2 and partial in 18) could be explained by weakness of the trapezius muscle, droop of the shoulder and decreased range of active abduction. The strength of the trapezius was moderately decreased after partial division of the accessory nerve in 8 cases and after total division in 1. All these cases had a slight to moderate droop of the shoulder. The range of active abduction against gravity was slightly limited in 2 patients after partial, and in 1 after total division of the accessory nerve (160–170° with the arm straight).

Shoulder pain occurred in 2 patients with moderate weakness of the trapezius after partial division of the accessory nerve. The pain was moderate. Two of the above-mentioned 9 patients reported that the symptoms interfered with their working capacity. In one of these 2 the accessory nerve had been completely divided.

Loss of sensibility in the 3 patients who had undergone division of dorsal roots was moderate in 2 cases and corresponded to the denervated segment. The patients were not troubled by the hypesthesia. In the 3rd case no decrease in sensibility could be demonstrated (C_2–C_4 divided bilaterally).

Persistent dystonia.

At the re-examination 14 patients showed signs of persistent dystonia; 11 patients found this symptom embarrassing. In 2 cases palpation and inspection revealed dystonic activity despite the absence of involuntary movements of the head or other symptoms of torticollis.

The malposition of the head was severe in 1 patient, moderate in 4 and slight in 9, while in 7 the position of the head was normal. Involuntary movements were severe in 1 patient, moderate in 1 and slight in 5, while 13 were free from this symptom. Tremor was severe in 2, slight in 8 and absent in the other 10 patients.

Dystonic activity of the neck muscles was observed in 9 patients but produced no symptoms in 4 of them. The remaining 5, 4 of whom had undergone division of ventral roots, had both symptoms and signs of dystonia, which were slight in 4 and moderate in 1.

At the re-examination considerable dystonic activity of the sternomastoid muscle was observed in 9 patients, but produced no symptoms in 4 of them. Of the other 5, all of whom had undergone partial division of the accessory nerve, 4 had moderate and 1 severe symptoms of dystonia.

In 6 of the 14 patients with persistent dystonia, dystonic activity of the trapezius was noted after partial division of the accessory nerve. In 3 of them the picture was dominated by dystonia of other muscles, while such activity of the trapezius appeared to cause moderate or slight distress.

Two of the 3 patients who had undergone division of dorsal roots had signs of slight persistent dystonia but all 3 reported symptoms of tension from the neck muscles, which were generally hypertrophic.

Recurrences.

Six of the patients described in the preceding section had clear-cut recurrences of torticollis after initial improvement.

Reinnervation of the sternomastoid muscle was the main cause of recurrence in 2 cases. 12 years before the re-examination 1 of them had undergone partial division of the accessory nerve without insertion of a tantalum sheath. He was troubled by severe dystonic activity of the muscle in question, which was very strong and hypertrophic. He nevertheless declined reoperation. The other patient also had symptoms of torticollis due partly to dystonia of the sternomastoid although the

muscle was moderately weak after denervation with insertion of a tantalum sheath 7 years previously.

Some function of the sternomastoid muscle was noted in 11 of the 16 cases where the muscle had been selectively denervated. Of the 4 operated upon without insertion of a tantalum sheath, 2 had to be reoperated upon 1 year and 7 years respectively after the operation; the 3rd of these patients had refused reoperation (see above), while in the 4th the muscle was of normal strength 12 years after operation. After denervation with insertion of a tantalum sheath the strength of the muscle was normal in 1 patient, moderate in 4 and greatly redused in 8.

In 3 cases with recurrences the deterioration was due mainly to dystonic activity having reappeared in the neck muscles. All 3 patients complain of a troublesome pull of the neck muscles, but they have full working capacity. In 1 of these cases an unsuccessful attempt to secure relief by extradural division of the rami posteriores (see page 107) was made. This patient differs from the remainder in that the dystonia tended to migrate from one muscle group to the other though without involving more caudal segments.

Propagation to caudal segments. The patient in whom the poorest result was noted at the re-examination had begun to show signs of spread in caudal direction of the disease. He complained of "pulling in all directions" in one half of the body.

Evaluation of the results.

The patients' opinions of the effect of the operation on the torticollis as a whole (symptoms of muscular insufficiency included) were: no improvement in 1 case, moderate improvement in 5 cases, considerable improvement in 14 cases and complete recovery in 2 cases. As to working capacity, judged by the examiner, the results were: none in 1 case, severely impaired in 2 cases, moderately impaired in 8 cases and normal in 11 cases. See also Tables 1 and 2.

An attempt to treat the results numerically showed no significant difference between the operative methods used, the groups compared being too small.

Postoperative physical therapy.

In most cases the patients received educational exercise and massage after the operation while they were in hospital, and several of the patients also received physical therapy after discharge. The treatment

TABLE 1
Patients' estimate of the result.

	No improvement	Moderate improvement	Considerable improvement	Complete recovery (symptomfree)
Serafimer Hospital ...	8 cases (= 16 %)	9 cases (= 18 %)	25 cases (= 50 %)	8 cases (= 16 %)
Lund	1 case	5 cases	14 cases	2 cases
Total (72 cases)	9 cases (= 13 %)	14 cases (= 19 %)	39 cases (= 54 %)	10 cases (= 14 %)

TABLE 2
Working capacity.

	None	Severely impaired	Moderately impaired	Normal
Serafimer Hospital ...	3 cases (= 6 %)	9 cases (= 18 %)	23 cases (= 46 %)	15 cases (= 30 %)
Lund	1 case	2 cases	8 cases	11 cases
Total (72 cases)	4 cases (= 5 %)	11 cases (= 16 %)	31 cases (= 43 %)	26 cases (= 36 %)

and the results of this treatment were not studied systematically. The purpose of the treatment was to teach the patient to balance and keep the head upright as soon as possible and to prevent decrease in mobility due to contracture and malposition of the spine. Relaxation exercises in 2 cases were unsuccessful. In 2 other cases who complained in a late stage of neck stiffness, physical therapy was followed by a marked subjective and objective improvement.

DISCUSSION

The results of operation will, of course, vary with the criteria used in the selection of patients for surgery. Differences in these criteria also make comparison between different series difficult. Occasionally torticollis dystonia is only part of a more extensive dyskinetic syndrome or a symptom of generalized cerebral arteriosclerosis. The acceptance of such cases for operation will, of course, reduce the overall results. On the other hand, the acceptance of early cases observed for only a short time before operation and of mild cases might give a too favourable impression of what can be expected from operation.

TABLE 3

	No. of cases	Op. mortality	Unimproved	Improved	Cured or symptom-free
Finney & Hughson, 1925. Bilateral resection of rami posteriores	31	0	3 (10 %)	16 (52 %)	12 (38 %)
Dandy, 1930. Bilateral resection of ventral and dorsal roots	8	1		2	5
Patterson & Little, 1943. Non-surgical treatment	197	0	122 (62 %)	75 (38 %)	
Section of accessory nerve	19	0	7	11	1
Bilateral section of cervical roots (with or without acc. nerve section)	8	0	0	8	0
Lundberg, 1946. (Olivecrona's series). Section of accessory nerve	11	0	3	7	1
Bilateral section of cervical roots (with or without acc. nerve section)	36	0	5 (13 %)	24 (67 %)	7 (20 %)
Putnam et al., 1949. Bilateral section of ventral roots with or without accessory nerve section	16	0	2	12	2
Poppen & Martinez-Niochet, 1951. Bilateral section of dorsal and ventral roots (22 cases), section of accessory nerve (22 cases) excision of sternomastoid (11 cases)	37	1	14 (38 %)	22 (59 %)	0
Wycis & Moore, 1954. Mainly bilateral section of ventral roots and section of accessory nerve	9	0	0	1	8
McKenzie, 1955. Bilateral section of ventral roots, accessory nerve section	12	0	0	10	2
Törmä & Troupp, 1958. Bilateral section of ventral roots, intradural section of accessory nerve	18	2	3	10	3
Denervation of sternomastoid	5	0	5	0	0
Lundberg & Svantesson, 1960. Denervation of sternomastoid	4	0	0	4	0
Bilateral section of cervical roots (with or without acc. nerve section)	18	0	1	15	2

Table 3 summarizes the results given by the authors cited in our historical review. The classification used here may tend to give a somewhat wrong impression of the original reports. It was not possible to include the report of *Adson et al.* (1946, see page 103) in Table 3.

No reliable data are available on the frequency of spontaneous remissions in torticollis dystonia or at what interval after the onset remissions might be expected. Of 43 cases described by *Herz & Glaser* (1949) spontaneous improvement occurred in 6 patients 6–10 years after onset and in 3 the symptoms disappeared spontaneously 1.5–6.5 years after onset. In 3 cases short remissions also occurred. Those cases in which spontaneous remissions were observed had been followed up for at least 3 months and on the average for 6.5 years.

In view of these figures and the somewhat mutilating nature of the operation it might be questioned whether the preoperative observation period of 6 months used in the present series was not too short.

Sequelae impairing the results. In the report on the Serafimer series it was suggested that the results were impaired by three main groups of symptoms (see page 104), which according to the questionnaire answers seemed equally important as causes of reduced working capacity. In the Lund series, where the patients' reports could be checked at personal examinations by one and the same examiner, reduction of working capacity was found to be due mainly to persistent dystonic activity, while division of ventral roots appeared to have only a slight invalidating effect.

In the Serafimer series paralysis of the trapezius was considered to cause distressing symptoms and to impair working capacity in about half of the cases. In the Lund series there was no instance of reduced working capacity because of paralysis of the accessory nerve. This appears to imply a considerable advance. It is true that selective denervation of the sternomastoid muscle implies a greater risk of reinnervation with recurrence of dystonic activity in this muscle. In 2 cases of the present series recurrence of considerable dystonic activity in the denervated sternomastoid muscle occurred. Reoperation is, however, a minor procedure and was considered indicated in these cases. Extirpation of the sternomastoid muscle as used *e.g.* by *Poppen & Martinez-Niochet* (1951) might also be indicated in such cases.

The erratic appearance of dysphagia, which was prominent in only 2 out of 22 cases, must be due to variations in the innervation of the swallowing muscles. This complication can hardly be eliminated by modifying the method of operation, but suitable exercise in an early stage may be of benefit.

Choice of operative method. The two series do not permit any statistical comparison regarding the results obtained by different combinations of the 3 procedures, *i.e.*, division of the accessory nerve, ventral

roots and dorsal roots respectively. However, observations discussed in the preceding paragraph argue definitely against resection of the entire accessory nerve as a first measure and suggest that one should always avoid denervation of the trapezius, unless it is obvious that this muscle actively contributes to the dystonia. Such denervation should be preceded by transient blockage of the nerve by injection of alcohol or crushing of the nerve. Selective denervation of the sternomastoid muscle requires only a minor operation without risk of complications and may be chosen as the initial measure in suitable cases, particularly in those with only mild symptoms of dystonia. This operation may give the patient some relief during the expectancy before root section is decided. If a patient has had severe disabling dystonia for a considerable time, there is no reason why the treatment should not be started by root section. Section of the accessory nerve may then turn out to be unnecessary.

Our interest in division of the dorsal roots was prompted mainly by the impression that many of the postoperative complications were due to muscular insufficiency after division of the ventral roots. This, however, does not seem to be the case. In addition, the distressing symptoms after dorsal root division in 3 cases were, if anything, more severe than what was usually found after division of ventral roots, and the effect on dystonia, as judged by objective examination, was not better. Judging from the present investigation, there is no reason to prefer division of the dorsal roots in the surgical treatment of torticollis dystonica.

At present the following procedure appears to offer the best results and should be used as a standard method: on that side to which the head is rotated the 4 uppermost ventral roots are divided, and on the other side the 3 uppermost roots. If necessary a sternomastoid muscle showing dystonic activity is subjected to selective denervation.

SUMMARY

Fifty cases of dystonic torticollis operated upon at the Serafimer Hospital in 1926–1944 were reexamined by questionnaires. 22 cases treated in the neurosurgical department in Lund during 1946–1957 were reexamined by questionnaires and 20 of them personally by one and the same examiner. The average observation period for the first series was 8 years (at least 1½ years) and for the second 6 years (at least 13 months). There was no operative mortality or serious postoperative complication.

The two series contained a total of 40 cases operated upon according to the following standard method: division of the accessory nerve (complete or partial) + bilateral division of the 3–4 uppermost ventral roots. These 40 patients described the results as follows:

No improvement	Moderate improvement	Considerable improvement	Symptom-free
5 cases (12.5 %)	8 cases (20 %)	21 cases (52.8 %)	5 cases (15 %)

The results for the whole series (including cases operated on by division of the accessory nerve only and division of dorsal roots) were roughly the same. The results in 9 cases treated by bilateral section of dorsal roots (alone or in combination with ventral root section) did not support the suggestion that this operation might be preferable.

In the first series paralysis of the trapezius muscle after division of the accessory nerve appeared to be responsible for a considerable part (roughly ⅓) of the poor results. In the second series the accessory nerve was only partly divided, the innervation of the trapezius being preserved and reinnervation of the sternomastoid muscle prevented by a sheath of tantalum foil around the nerve. Paresis of the trapezius was in no case responsible for impaired working capacity in the second series. Working capacity was found to be reduced mainly because of persistent dyskinetic or dystonic activity, disability from paresis due to root section being relatively insignificant.

Bilateral division of the upper ventral roots, if necessary completed by selective denervation of the sternomastoid and followed by adequate physical therapy appears to be the best treatment as yet available for severe torticollis.

RESUME

Cinquante cas de torticolis dystonique opérés à l'Hôpital Serafimer dans la période 1926 – 1944 ont été réexaminés sur la base d'un questionnaire. 22 cas traités par le Service neurochirurgical de Lund entre 1946 et 1957 ont également été réexaminés sur la base d'un questionnaire, 20 d'entre eux ayant toutefois été examinés personnellement par le même examinateur. La période moyenne d'observation dans la première série a été de 8 ans (au moins 2 ans et demi) et pour la seconde de 6 ans (au moins 13 mois). Il n'y a pas eu de mortalité opératoire ou de complications post-opératoires graves.

Les deux séries de cas comprennent au total 40 cas opérés d'après la méthode standard suivante: division du nerf accessoire (complète ou partielle) + division bilatérale des 3–4 racines ventrales supérieures. Ces 40 malades décrivent comme suit les résultats obtenus:

Pas d'amélioration	Amélioration modérée	Amélioration considerable	Disparition des symptômes
5 cas (12,5 %)	8 cas (20 %)	21 cas (52,8 %)	5 cas (15 %)

Les résultats pour les deux séries (y compris les cas opérés seulement par la division des nerfs accessoires et la division des racines dorsales) ont été sensiblement les mêmes. Dans 9 cas traités par section bilatérale des racines dorsales (uniquement ou en combinaison avec la section des racines ventrales) les résultats n'ont pas confirmé l'idée que cette opération était préférable.

Dans la première série, la paralysie du muscle trapèze après la division du nerf accessoire s'avère être responsable d'une partie considérable (en gros ⅓) des maigres résultats. Dans la seconde série, le nerf accessoire n'a été que partiellement divisé, l'innervation du trapèze ayant été conservée et la réinnervation du muscle sterno-mastoïdien ayant été empêchée par une gaîne d'un feuillet de tantale autour du nerf. La parésie du trapèze n'a été dans aucun cas responsable d'une capacité de travail diminuée dans la seconde série. La capacité de travail s'est trouvée réduite principalement par suite d'une dyskinésie ou d'une dystonic persistante. L'incapacité causée par une parésie due à la section de la racine était relativement insignifiante.

La division bilatérale des racines ventrales supérieures, si nécessaire complétée par dénervation sélective du muscle sterno-mastoïdien, suivie d'une physiotherapie appropriée semble être le meilleur traitement que l'on connaisse jusqu'ici pour les cas graves de torticolis.

ZUSAMMENFASSUNG

Fünfzig Fälle von dystonischem Torticollis, die am Serafimer Krankenhaus in den Jahren 1926–1944 operiert worden waren, wurden mittels Fragebogen nachuntersucht. 22 Fälle, die an der neurochirurgischen Abteilung in Lund von 1946–1957 behandelt worden waren, wurden mittels Fragebogen und 20 Fälle wurden persönlich von ein und demselben Untersucher nachuntersucht. Die durchschnittliche Beobach-

tungszeit für die erste Untersuchungsreihe war 8 Jahre (zumindest 1½ Jahre) und für die zweite 6 Jahre (zumindest 13 Monate). Kein operativer Todesfall oder ernstliche postoperative Komplikation trat auf.

Diese beiden Untersuchungsreihen umfassten insgesamt 40 Fälle, die gemäss folgender Standardmethode operiert worden waren: Durchtrennung des N. accessorius (volständig oder teilweise) + doppelseitige Durchtrennung der 3–4 obersten ventralen Wurzeln. Diese 40 Patienten beschrieben das Ergebnis folgenderweise:

Keine Besserung	Mässige Besserung	Bedeutende Besserung	Symptomfrei
5 Fälle (12,5 %)	8 Fälle (20 %)	21 Fälle (52,8 %)	5 Fälle (15 %)

Die Ergebnisse in sämtlichen Fällen (einschliesslich der Fälle, die nur mittels Durchtrennung des N. accessorius und der dorsalen Wurzeln operiert wurden) waren ungefähr die gleichen. Die Ergebnisse in 9 Fällen, die mittels doppelseitiger Durchtrennung der dorsalen Wurzeln (allein oder in Kombination mit Durchtrennung ventraler Wurzeln) behandelt waren, unterstützen nicht die Annahme, dass diese Operation vorzuziehen sei.

In der ersten Reihenfolge schien die Lähmung des M. trapezius nach der Durchtrennung des N. accessorius zum grossen Teil (ungefähr ⅓) für die schlechten Ergebnisse verantwortlich zu sein. In der zweiten Reihenfolge wurde der N. accessorius nur teilweise durchtrennt indem die Innervation des M. trapezius bewahrt wurde und die Reinnervation des M. sternocleidomastoideus mittels einer Scheide von Tantalumfolie um den Nerven verhindert wurde. Parese des M. trapezius war in keinem Falle die Ursache herabgesetzter Arbeitsfähigkeit in der zweiten Reihenfolge. Die Arbeitsfähigkeit war hauptsächlich wegen fortgesetzter dyskineticher und dystonisher Aktivitet verringert indem die Störung, die als eine Folge der Wurzeldurchtrennung auftrat relativ unbedeutend war.

Doppelseitige Durchtrennung der oberen ventralen Wurzeln, wenn notwendig kombiniert mit einer selektiven Dennervation des M. sternocleidomastoideus und einer adäquaten physikalischen Behandlung scheint zur Zeit die beste Behandlungsart für schweren Schiefhals zu sein.

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