

BILATERAL TRIGEMINAL NEURALGIA

BY CHARLES H. FRAZIER, M.D., Sc.D.

OF PHILADELPHIA, PA.

FROM THE NEUROSURGICAL CLINIC OF THE HOSPITAL OF THE UNIVERSITY OF PENNSYLVANIA

FOR the most part, contributions touching on the subject of major trigeminal neuralgia for many years have dealt with methods of treatment, surgical and otherwise. Perhaps one should apologize for bringing before this Association a topic which at first sight appears to lack originality. Granted the principles underlying the treatment of this fifth nerve neuralgia are generally understood and quite universally accepted, there are many other aspects that might properly be designated as problems.

In the Neurosurgical Clinic of the University Hospital the material for study and observation has been abundant and has imposed many obligations that I am quite conscious have not been fulfilled. On my records are 2,198 cases of what I have designated as major trigeminal neuralgia, known to the laity as *tic douloureux*, and for supplementary study there are 479 cases, some of which are border-line and some unquestionably "atypical." Altogether, 875 operations have been performed on sensory the root or Gasserian ganglion.

Yet with this unlimited opportunity for observation we know no more now than 200 years ago of the etiology. Theories there may be aplenty, but facts are wholly lacking. Why should the character of the painful paroxysm vary so? Why should there be a trigger zone in some and not in others? Why should a patient with this torturing ailment take his punishment for twenty years or more before he is willing to capitulate? Why does the victim of this disease suffer as much from the dread of an impending paroxysm as from the paroxysm itself? Many have acknowledged this to be true. Why is morphine not the answer to the "tic" patient's pain and why have we never seen a drug addict among subjects of this disease? Does the motor root carry afferent as well as efferent fibres, and what do we know about the diencephalic root? Is there an anatomical connection between the trigeminal system of either side? These are but a few of the questions that one might ask with no thought of drawing an acceptable reply.

It is my purpose at this time to confine my remarks to bilateral trigeminal neuralgia. To speak of bilateral trigeminal neuralgia seems an anachronism, for, you say, major trigeminal neuralgia is essentially a unilateral disease.

Essentially this is true, but there are notable exceptions. Depending altogether upon ratification by the patient and not upon a systematic canvass of the 2,198 cases, only twenty-three instances of genuine bilateral major trigeminal neuralgia have been brought to my attention—a morbidity rate of about 1 per cent.

BILATERAL TRIGEMINAL NEURALGIA

As to the incidence of bilateral trigeminal neuralgia, there are no available statistics upon which may be based an exact statement. In 1932, one observer (Glaser: *Arch. Neurol. and Psychiat.*, vol. 28, p. 418, 1932) stated that up to that time but fifty-three cases had been referred to in literature. The first case to be recorded in detail was that of Winslow in 1896 (*ANNALS OF SURGERY*, vol. 24, p. 748, 1896), although, since his patient was only thirty-one years of age, one might question the diagnosis, and ten years later Bullock (*Lancet Clin.*, vol. 54, p. 226, 1905) described a case, and this, curiously enough, in a colored woman. I say curiously enough since the Negro seems to enjoy almost an immunity.

Harris (*Neuritis and Neuralgia*, Oxford Medical Publications), who has had a liberal experience with this disease, estimates the incidence of bilateral cases as 5 per cent. Altogether he had seen in 1926 thirty-six instances and if these represented 5 per cent. of his entire series, his total experience would include 729 cases. This percentage is so much in excess of the number that have developed in my own series that I wondered at the discrepancy. Upon investigation of Harris' experience, I found that he includes in his series what he describes as "typical chronic trigeminal neuralgia major" in patients suffering with some form of chronic spastic paraplegia, usually disseminated sclerosis. In fact, he says that cases of disseminated sclerosis in association with trigeminal neuralgia are numerous, too numerous to be accounted for by any theory of chance. The percentage of bilateral cases in my series is about 1 per cent. and I cannot recall any case of major neuralgia in a patient with disseminated sclerosis. Here again our individual experiences are widely at variance.

In his article on bilateral major neuralgia Olivecrona (*Arch. f. klin. Chir.*, vol. 164, p. 196, 1931) records but two cases in his clinic. He, too, refers to the frequent association with disseminated sclerosis in which disease he says bilateral cases are relatively frequent. If this be true, we must assume that insofar as the etiology is concerned we must be dealing with two separate and distinct expressions of the disease. We are convinced that in tic douloureux, unassociated with organic disease, there is no organic lesion as the etiological factor, whereas when associated with disseminated sclerosis, especially when the neuralgia is bilateral, we must postulate that the neuralgia is of central origin.

It is difficult for me to include in the same category these two neuralgias of radically different origin. It is difficult to explain the paroxysmal character of true tic douloureux as due to a central organic lesion and I am disposed to believe that were a comparison drawn between the clinical expression of these two forms of neuralgia one would find they varied from one another in many particulars.

The incidence in my series more nearly approaches that of Cushing, who, in 1920 (*Am. Jour. Med. Sci.*, vol. 160, p. 157, 1920) had seen but two in a series of 322 patients, or a percentage of 0.62. In 1926, Adson (*Ann. Otol.*,

Rhinol., and Laryngol., vol. 35, p. 601, 1926) reported thirteen in a series of 839 cases, or 1.5 per cent.

It is rather important that we should have a fairly accurate conception of the incidence of bilateral neuralgia since so often this pertinent question is asked by the patient. What are the chances of the neuralgia developing on the opposite side? From the figures at hand I should be inclined to answer about 1 per cent., disregarding the 5 per cent., as quoted by Harris, in whose series there are a number of cases associated with disseminated sclerosis.

In order that there may be no confusion as to the subject under discussion, let it be understood that when we speak of a case of bilateral neuralgia we imply one in which the disease began on one side and continued on that side, be it right or left, for many years before it made its appearance on the contralateral side. In my experience and in that of the majority of observers, this is the almost universal experience. We admit occasional exceptions, but I was surprised to read in Adson's series (*loc. cit.*) that the neuralgia generally appeared simultaneously on both sides.

When the patient has been relieved by operation of pain on the side on which it originated, the sudden onset of pain on the other side, often after a lapse of many years, is a dreadful shock. Having reveled in the relief for so many years and never having been told, as he should not be, of the remote possibility of an explosion on the free side, the experience undermines the patient's morale. It is a pathetic visitation. In my entire series I have seen but one case of true major trigeminal neuralgia which from the onset alternated from one side to the other. Occasionally a patient will tell you of the experience of a relative or friend who had the same disease first on one side and then on the other in different years, or at various periods in the same year. But true major trigeminal neuralgia never disappears for good spontaneously nor is it, except in the rarest instances, an alternating affair.

Because it was an exception to the rule and because it illustrates many of the experiences, the trials and the tribulations of the "tic" patient and because, too, it illustrates many interesting features of the bilateral case and the bilateral operation, the following case is introduced and here described in some detail.

First Hospitalization.—S. MacM., aged fifty-three years (File No. 22057) was referred to the Neurosurgical Service, June 24, 1931, by Dr. W. L. Nance, of Georgia, with the following history:

Beginning in 1914, following some dental trouble, he developed what seemed to be an attack of typical major neuralgia on the *left* side of his face. At first the third division (tongue, angle of jaw) were involved and later the maxillary division (infra-orbital distribution). The pain was controlled for a while by repeated alcoholic injections.

As the pain decreased on the *left*, it developed on the *right* side (mandibular division). In 1924, he had all of his teeth extracted but without relief. As the alcoholic injections on the right side began to lose their effect he came to our clinic prepared for operation.

BILATERAL TRIGEMINAL NEURALGIA

First Operation.—Subtotal section of the sensory root, right, June 25, 1931. Ether anaesthesia. The ganglion was approached by our conventional temporal approach. It was described as very thin and it was recorded that the whole ganglion, including its mesial margin, was exposed. A perfect view of the sensory root was obtained; one outer and two of the mesial fasciculi were isolated and the intervening fasciculi divided. After securing perfect hemostasis the wound was closed with tier sutures without drainage. After an uneventful convalescence the patient was discharged from the service seven days after the operation.

Second Hospitalization.—Not long after the sensory root was divided on the right side (1931) the pain reappeared on the left side. It was the typical "tic" pain, paroxysmal, aggravated by talking and swallowing and eating, sharp, shooting, and was referred both to the maxillary and mandibular divisions. The patient had had all his teeth extracted, as recorded above, he had had several sinus operations, thirty osteopathic treatments and a number of alcoholic injections. Finally, he became demoralized and returned to our Neurosurgical Clinic June 19, 1934, just three years after his first operation.

Examination.—At this time it was observed that the right masseter muscle (side of first radical operation) contracted although there was a slight deviation of the jaw to the left upon opening the mouth, but the left masseter did not contract. Probably the motor distribution of the trigeminus had been damaged by the repeated alcoholic injections (left). A careful sensory examination on the right side revealed total anaesthesia to pain (Fig. 1), as one would expect, in the mandibular and maxillary divisions (right). There appeared to be a left Horner's syndrome. The patient apparently had reached the limit of endurance and craved relief by operation.



FIG. 1.—The area of anaesthesia after subtotal section of the sensory root.

Second Operation.—Subtotal section of the sensory root, left, June 22, 1934. In this case the function of the masseter was preserved on the side of our previous radical operation but as a result of multiple alcoholic injections elsewhere, was lost on the side we were about to operate upon. There was, therefore, no special reason for concern about the motor root. But since it is our practise to do so as routine, the motor root was avoided. The outer two-thirds of the sensory root were divided and the wound closed without drainage.

Comment.—There must be exceptions to all rules as there was in this case. Rarely does the pain alternate, first on one side, then on the other. In this case it did unquestionably. It began on the right, later involved the left side and later subsided only to reappear with increasing severity again on the right.

As to sex of the twenty-three cases of bilateral major neuralgia, nineteen were female and four male; and as to the age, one was under forty, five between forty and forty-nine, seven between fifty and fifty-nine, four between sixty and sixty-nine, and five were between seventy and seventy-five.

There is usually a lapse of one to many years between the original onset on one side and the development of the disease on the other. In my own

series there were intervals varying from one to twenty-two years. In my experience and in that of others the pain which appears later on the opposite side is usually of much less intensity. This is a matter of practical significance since if the contralateral pain is not severe the performance of a major operation may not be justifiable. And so, in my series of twenty-three cases the radical operation was performed on both sides in only four instances. That an alcoholic injection was all-sufficient to control the pain on the second affected side may be assumed from the fact that in only one instance did the patient return for a second injection, that is for the pain on the contralateral side.

That trigeminal neuralgia is in an overwhelming majority of cases right- or left-sided might offer a clue as to its etiology. At once this unilaterality suggests a unilateral focus of irritation. Yet today no one will venture to attribute this pain syndrome to any peripheral lesion. Certainly, not to any infection process, not to sinusitis and not to dental sepsis. Sinuses have been drained freely and every tooth often has been extracted.

One returns ever to the thought that the pain may be due to vasospasm. The trigeminal system is so richly endowed with sympathetic plexuses and ganglia, that this hypothesis would seem to have a reasonable foundation. Can one draw an analogy between Raynaud's disease and trigeminal neuralgia? In the former, vasospasm is admitted to be the causal factor of pain but Raynaud's disease is almost invariably bilateral in its distribution and trigeminal neuralgia, with these few exceptions, unilateral. After drawing this analogy one comes to the end of the road of speculation. In favor of vasospasm as the cause of pain are: first, the fact that the pain is paroxysmal in character; and, secondly, that no matter how long the duration of the disease, one can find absolutely not a vestige of structural damage to the trigeminal system from its peripheral filaments, through its chief divisions, through the ganglion, through the sensory root to the central nuclei.

However one may speculate as to the origin of bilateral trigeminal neuralgia or however the incidence of the bilateral true paroxysmal neuralgia may vary in the statistics of various clinics, the practical aspect of the disease in question relates only to the treatment.

Why should the treatment of bilateral neuralgia differ from that of unilateral neuralgia? It is not a question as to whether the pain can be relieved on both sides but whether in the treatment employed the motor root can be conserved. The motor root supplies the pterygoid, the masseters and the temporal muscles, the muscles of mastication. Not only would the patient not be able to masticate were the motor root sacrificed on both sides, but the patient would not be able to keep her mouth shut, actually not figuratively speaking, acknowledgedly a lamentable predicament. And so in all discussions of the treatment of bilateral neuralgia one's attention is focused on the motor root. The importance of conserving the motor root was appreciated long ago. In that oft-quoted article (Tiffany, *ANNALS OF SURGERY*, vol. 24, p. 575, 1896) Tiffany, in 1896, wrote:

BILATERAL TRIGEMINAL NEURALGIA

"The expediency of attempting to save the motor fibres accompanying the third division may be considered. Usually I have not taken the pains to do so, yet I thought that I recognized them. In one of my cases the muscles of mastication were made to contract again and again by passing a tenaculum under the third branch close to the bone. Because of the case reported by Winslow comes the suggestion that perhaps an operation may be expedient on both Gasserian ganglia, and if so, *a bilateral paralysis of the muscles of mastication would be unfortunate* (italics mine). While the necessity for a bilateral operation is remote, yet the Hartley-Krause operation will be more complete if the motor root is spared, and I think it can be."

As a matter of fact, it is generally acknowledged that in resections of the Gasserian ganglion, either unilateral or bilateral, the motor root would be sacrificed. However, since excisions of the ganglion have been abandoned, this plan of treatment can be omitted from our discussion.

Of the radical methods of treatment there remain only to be considered alcoholic injections of the ganglion or the conventional operation on the sensory root. Wilfred Harris (*loc. cit.*), an outstanding exponent of the injection treatment, states that paralysis of the masticating muscles follows the injection immediately, but with *rare exceptions* the muscles commence to recover within a few months and eventually complete power returns. In this connection I might refer to the case above reported in which after repeated injections of the third division motor root paralysis persisted. He acknowledges, however, that there are exceptions, and, in this connection, Olivecrona (*loc. cit.*) says in his experience regeneration of the motor root is not so universal that one can be sure of it. While I have had no experience with bilateral injections of the ganglion, admission of the uncertainty of functional recovery of the motor root should stamp this procedure as unacceptable if not prohibitive.

If excisions of the Gasserian ganglion and alcoholic injections of the ganglion cannot be performed with absolute assurance that the motor root will be conserved, what can be said of the conventional operation on the sensory root? In 1919, I first called attention to the possibility and feasibility of conserving the motor root in the radical operation on the sensory root (*J.A.M.A.*, vol. 87, p. 1730, 1926). We had not discussed the question before but it then became evident, first, that the motor root could readily be identified and, secondly, that it could be separated from the sensory root. To be sure it lies in close contact with the sensory root, directly behind it, and in some instances its separation is more difficult than in others. We have called attention to the fact that in some cases the individual fasciculi of the sensory root are not, as in the average case, easily counted and separated one from the other. In the exceptional case the fasciculi are matted together as though there had been some inflammatory process, an arachnitis if you choose. In such instances the motor root may be with greater difficulty isolated.

But though there seems to be some skepticism on the part of certain authors, it will now be acknowledged by all who have had more than a casual

experience with sensory root operations that conservation of the motor root in the conventional operation on the sensory root is readily accomplished. One of the skeptics said he could well understand why even the most experienced surgeon would hesitate to perform the radical sensory root operation on a patient whose motor root had been sacrificed at a previous operation on the opposite side. He had in mind my experience with a patient upon whom, for the first time, the radical sensory root operation, first on the one side and later on the other, had been performed. It so happened in this case that I had sectioned the left sensory root in 1917. Up to this time conservation of the motor root had not been attempted. In 1926, the patient reappeared in the clinic with major neuralgia on the opposite or right side. There was complete paralysis of the muscles of mastication on the left side. At first we proposed an alcoholic injection. But knowing the transitory effect of the injection treatment and coming as she had from a great distance (Lima, Peru), the patient insisted upon the major operation. Although we had been in the habit of conserving the motor root for seven years, this was the first occasion on which I was confronted with a case in which one would have declined to operate, unless confident of one's ability to protect the only remaining motor root. I welcomed the opportunity to make this demonstration. This introduction is perhaps too long for the recital of what was at that time in our clinic a routine practise. The operation was performed and the function of the motor root was conserved. To be sure in this case there was a transitory paralysis due to the trauma incidental to the operation but on the seventh day the function of the muscles returned. I say routine practise, our plan of procedure is as follows: assuming the ganglion has been reached, the dura is separated from the surface of the ganglion until one can see on its mesial side the bluish, pulsating arachnoid overlying the sensory root. The arachnoid is incised just mesial to the ganglion and at right angles to the root. Thus, and in this way only, is a free exposure of the root secured. We assume a subtotal section of the root is contemplated and, in most cases this implies an accurate section of the outer two-thirds of the root. To accomplish this we proceed first to elevate the outer third of the root; this third is encircled on a special hook and divided. Now the motor root lies behind the mesial portion or middle section of the sensory root. This portion of the sensory root is then elevated from the base of the skull and at once the motor root will be seen. It can readily be identified, first because it appears quite separate and apart from the sensory root, and secondly because of its passage behind the ganglion. After thus isolating the motor root, the mesial portion of the sensory root is encircled on a hook, as with the outer third already divided, and sectioned.

Sometimes, be it remembered, the motor root may be composed of two fasciculi. It is well to bear this in mind because one may fear lest one of these two fasciculi may belong to the sensory root. And, be it remembered

BILATERAL TRIGEMINAL NEURALGIA

too, that there is a great variation in the size of this motor root; in one case its diameter may be twice that of another.

It is not in every case of bilateral neuralgia that the radical operation must be performed on both sides. In the first place, one must always consult the wishes of the patient. This is my invariable practise in all cases of neuralgia whether unilateral or bilateral. It is for the patient to decide whether he prefers an alcoholic injection or an operation. In the second place when in bilateral cases the pain appears on the opposite side, it is usually of not nearly the same violent intensity and is usually confined to a single division. Under these circumstances an alcoholic injection will be all-sufficient. In only one instance did the patient return for a second injection. In the entire series of twenty-three cases were there five bilateral major operations; there were nine major operations on one side with alcoholic injections on the other; there were six major operations on one side, and, though pain developed on the opposite side, the patient did not return for treatment; there were three bilateral alcoholic injections.

Thus, in a discussion of the treatment of bilateral neuralgia, it is the motor root alone that commands our attention. The hazards of the operation have been so reduced that a fatality is a rare exception. Our mortality rate is .8 of 1 per cent. Many years ago surgeons dreaded the thought of a bilateral keratitis and its possible effects upon vision. With the subtotal section introduced by us and practised since 1915, we have found an effective way of avoiding corneal complications.

Since my original demonstration in 1926, I have performed the radical operation in four other cases. The interval between the first and second operations in the five cases was one, two, three, five and eight years, respectively. In two of the four cases the outer two-thirds of the sensory root was divided; in two the outer two-thirds was divided on one side and the middle third on the other; in one the outer two-thirds of the sensory root was divided on one side and the outer two-thirds of the ganglion was resected on the other.

From this series it may be concluded when, for any reason, the radical operation on the sensory root is indicated, one may perform the operation with the assurance that the muscles of mastication will not be paralyzed.

DISCUSSION.—DR. WILLIAM JASON MIXTER (Boston, Mass.).—Doctor Frazier's experience is so great in this field that there is little that can be added to what he has said in his paper.

It has been my fortune to deal with fifteen of these cases. Four of them were subjected to bilateral section of the posterior root, two of the four having been operated on for the first time by my father while he was still alive.

The interesting thing in that group of four cases to me was that the anterior root could be preserved with good function of the muscles of mastication on one side, but that most patients had great difficulty in feeding themselves after both posterior roots had been cut. They lost the sense of position of their own mouths to such an extent that all of them had to learn to eat over again with a mirror. Otherwise they were sticking their noses and their tongues with their forks and having a terrible time.

CHARLES H. FRAZIER

Therefore we must admit that there is a very definite disability following bilateral section of the posterior roots. However, in cases with extreme pain there is no question in my mind but that it is an operation of very great value for the relief of these patients.

DR. MARTIN BUEL TINKER (Cornell Heights, Ithaca, N. Y.).—It seems to me Doctor Frazier deserves great credit for improving the modern operation by dividing the sensory roots rather than the entire root as was formerly done.

This is impressed upon me especially by reason of the fact that my early training was with Doctor Keen in the last part of his work in Philadelphia, where he had at that time an unusual series of thirty operations. It was also my privilege to assist Harvey Cushing in his early work at Johns Hopkins. In those days the motor root was always sacrificed, and those who have seen the results realize the disadvantages to these patients.

Doctor Frazier's safeguarding the eye is also highly important. The trophic disturbances in some patients are serious indeed. Some of you have seen an eye lost as a result of trophic disturbances. Doctor Frazier largely has been responsible for eliminating these two serious dangers in the modern Gasserian operation.

People in the smaller cities and country districts also have trigeminal neuralgia, and some of them haven't the price of carfare to Philadelphia.

Some younger members of this Association ought to go some place and learn this work; it isn't impossible or necessarily serious. I haven't lost any of my patients and I have succeeded in relieving many people who suffered great distress, as all know who have had much experience, in the treating of such cases.