EXPERIMENTAL SPASMODOIC TORTICOLLIS*

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SPASMODOIC torticollis is an involuntary hyperkinesis involving the muscles of the neck primarily on one side. Characteristically, paroxysms of moderate to severe contractions of the muscles occur which may be painful, and the resulting deformities of rotation of the head and flexion of the neck may be functionally incapacitating. The muscles of the neck mainly involved in this abnormal involuntary movement are the sternocleidomastoid, the trapezius, the splenius, and the scalenes,\textsuperscript{1,7} though almost all the ipsilateral muscles are involved to some degree.\textsuperscript{13} The bizarre posturing of the head that results is often striking and disabling, consisting of strong clonic-tonic lateral flexion of the head, rotation of the head with occiput toward the same side combined with torsion of the head toward the side of flexion. It has been shown that there is usually bilateral involvement of the muscles of the neck,\textsuperscript{13,25} and at times strong elements of retroflexion of the head are also present. This descriptive definition eliminates from this presentation other types of so-called torticollis.\textsuperscript{6,15,18,20}

The term spasmodic has been applied primarily to this affliction to denote the paroxysmal nature of the complex contractions of muscles. It has long been recognized that environmental stimuli, particularly in the psychic and emotional sphere, will precipitate or accentuate these paroxysmal contractions. In a nonstressful, secure environment, the deformity may be barely discernible and yet become strikingly apparent when the individual is in a state of stress. These paroxysms do not seem to be related directly to vestibular posturing mechanisms. Later in the course of the disease, more fixed positions of head and neck may occur because of secondary structural changes in muscles of the neck and cervical spine.

The lesion in the central nervous system of man that causes spasmodic torticollis has never been established and disagreement continues as to whether the basic etiology is actually organic\textsuperscript{2,9,15,20} or purely psychogenic.\textsuperscript{1,10,27} Some clinicians feel that the organic substrate for the disease is definite, but that the expression of the syndrome is strongly associated with psychogenic factors.\textsuperscript{2,9,12,15,20}

Human studies have not shown convincing focal pathological lesions as

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the cause of the torticollis, though the neostriatum, basal ganglia, vestibular apparatus, putamen, extrapyramidal and vestibular pathways, and diffuse chronic changes in ganglion cells have all been implicated.

Spasmodic torticollis was fortuitously produced in 7 monkeys in which lesions were produced in the mesencephalic tegmentum during the course of other experiments in our laboratories. Since the lesion in the central nervous system responsible for torticollis appears to be unknown and since a psychogenic etiology has been widely cited in the literature, these monkeys with spasmodic torticollis of proven organic etiology were studied.

METHODS

Experiments were carried out on 7 monkeys (Macaca mulatta). Monkeys #1 through #6 in this series, weighing 2.2–3.7 kg., were operated on under sterile conditions by the stereotactic method to produce electrolytic lesions in the medial reticular formation of the mesencephalon. Fine nichrome electrodes, 0.5 mm. in diameter and insulated with Formvar except for a bare tip, 0.5 mm. long, were introduced into the calvarium through a small trephine opening centered over the sagittal sinus in the frontal region. The electrodes were angled 30 degrees from the vertical zero plane and oriented to traverse down the axis of the brain stem at 2 mm., 2.5 mm., or 3 mm. from the midline. The electrode was introduced to the mesencephalic-pontine junction, where an electrolytic lesion was made using the Grass Stimulator, Model 120, with a current of 5 ma., 60 cycles or more, 10–20 volts, for 30 seconds. Five similar lesions were made at 1-mm. intervals as the electrode was withdrawn along the tract of entry. The opposite side was then done in a similar manner since all animals had bilateral lesions. Five of the animals had only one such operative procedure, but monkey #2 had two such operations 3 weeks apart.

All animals were carefully observed immediately following operation for neurological deficits, and upon recovery from anesthesia were observed at least twice daily for several weeks and observations were recorded daily.

In monkey #7, an implanted electrode, 0.35 mm. in diameter, was placed stereotactically into the medial reticular formation at the mesencephalic level 1 mm. lateral to the midline. The electrode was fixed to the skull by means of a plastic cap device under sterile conditions. Immediately upon recovery from the anesthesia, no neurological deficits were present. The next day the rostral tip of the implanted electrode was no longer visible above the scalp and roentgenograms of the skull showed that the electrode tip was now beneath the calvarium (Fig. 1B). Presumably the animal had somehow pushed the electrode farther into his brain. At this time a slightly dilated left pupil was present together with typical spasmodic torticollis (Fig. 1A).

Photographs and a short movie strip were made to document the torticollis and its spasmodic nature.

At the end of the observation period, the brain of each animal was perfused with normal saline followed by 10 per cent formalin and the brain was removed intact. Subsequently, serial sections of the brain were made and stained by the Weil and thionin techniques.

RESULTS

I. Experimental Studies. The spasmodic torticollis produced in the 7 monkeys was striking in its similarity to clinical spasmodic torticollis, and