Short report

Remission in spasmodic torticollis

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SUMMARY In 26 patients with spasmodic torticollis followed up for a median period of 12 years, the frequency of sustained remission was 23%, the median duration of remission was 8 years and duration of torticollis before remission 3 years. These results are more favourable than stated in the literature and should be taken into consideration before recommending surgical treatment.

Spasmodic torticollis is currently regarded as an adult-onset focal dystonia usually commencing between the fourth and seventh decades.^{1,2} Some patients may also have other movement disorders such as tremor, writer's cramp or facial dystonia and other members of the family may be similarly affected.³ Secondary physical and psychological morbidity is common. A few patients respond well, but inconsistently, to oral medications; the majority remain refractory and recommended surgical procedures carry a significant inevitable morbidity without predictable benefit.

Patients and results

Between 1968 and 1983, 26 patients (16 male, 10 female) with spasmodic torticollis were seen at University College Hospital who were available for long term follow-up. Mean age at onset was 41.7 years (range 21-68) and the mean length of follow-up was 13.7 years (2-33). Ten patients (38%) underwent remission after a median period of 3 years (2-17) and of these in six (23%) remission has been sustained for a median period of 6.5 years (2-12); the duration of remission for those who relapsed was 8 years (1-13). The remission rate at different periods of follow-up is shown in the table. All the patients were assessed at the end of the follow-up period to ensure that remission had persisted. In two cases very mild residual torticollis was apparent of which the patient was quite unaware. Of the 26 patients, none gave a history of significant psychiatric incapacity but in two who recovered, the onset of torticollis had been immediately preceded by marital disharmony.

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Illustrative case reports

Patient 1 A stoical and singularly un-neurotic 46-year-old former pilot with a history of neck injuries developed spasmodic torticollis in 1959 when he found he was unable to prevent his chin rotating towards his left shoulder and also developed facial grimacing. His condition slowly deteriorated over the next 10 years and was complicated by cervical spondylosis and brachial neuralgia. Many medications were unsuccessful and the patient declined surgery. In 1976 after a mass healing service in Los Angeles there was sudden improvement. Complete resolution occurred over the next ten days and this has been maintained for seven years.

Patient 2 In 1979 a 51-year-old woman developed spasmodic torticollis to her right; the involuntary movements were exacerbated by walking or turning. She had left her husband because of his violent behaviour shortly before the onset of her symptoms. Her condition remained unaltered for three years and in 1981 she became so depressed that a psychiatric admission was required. The following year when getting off a bus she noticed a dramatic absence of torticollis and over the ensuing six months her condition gradually resolved and she has been symptom free for over a year.

Patient 3 A 21-year-old man presented with spasmodic torticollis in 1970. He also had writer's cramp and his mother suffered from Meige's syndrome. His condition deteriorated with severe neck pain and was unresponsive to medications. Levodopa was thought to produce benefit, but improvement was maintained after a placebo switch. One morning having been the previous night in a state of panic and agitation over the intractable nature of his condition, he awoke to find that neck postures were fuller and less painful. Over the ensuing six months torticollis

Table Actuarial percentage of cumulative remission rate in spasmodic torticollis

Years of follow up	1	2	3	5	7	10	15	20
Number of patients	26	26	24	21	19	17	9	6
% Rate of remission	0	15	21	24	31	29	22	16

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appeared only when walking and complete remission gradually occurred and has persisted for ten years.

Patient 4 A 58-year-old spinster who had been previously given methyldopa for hypertension in 1970 developed severe torticollis to her right which gradually became more disabling over six years causing neck pain and severe depression. Haloperidol 1.5 mg a day induced gradual improvement in her torticollis over three months; deterioration occurred on placebo, but she failed to benefit subsequently from haloperidol. By 1978 she was in complete remission and has remained well without medication for five years. Examination revealed a slight non-disabling neck torsion at the end of the follow-up period.

Patient 5 A 44-year-old man developed incapacitating torticollis in 1954 and after two years his condition spontaneously recovered. He remained well until 1965 when on the day of his fourth marriage torticollis recurred—previously it had been to the left and was now to the right—and his condition has remained static for eighteen years. His torticollis had not been amenable to treatment although he obtained slight relief from 5 mg of diazepam daily.

Patient 6 A 32-year-old man who had viral encephalitis in childhood, at the age of thirty-two developed writer's cramp and then spasmodic torticollis. His condition was unaltered for three years and then gradually improved to complete remission while undergoing hypnosis. Thirteen years later torticollis recurred and apart from increasingly severe degenerative cervical spondylosis his condition has remained static.

Discussion

In this study the frequency of remission (38%) is higher than previous reports (10–27%)^{4,5} and the frequency of sustained remission is also higher at 23% (3–12%) despite a median follow-up period after remission of 7 years. However with relapses at

eight and thirteen years in patients 5 and 6, the possibility of late recurrence must remain. Remissions occurred later after onset (3 years) compared with other studies⁶ with none in the first year and one after seventeen years. There were no evident clinical features—neurological or psychiatric—which facilitated prognosis. Only one remission could be related to medication (patient 4). Onset of improvement was invariably sudden, but only occasionally immediately complete, more often there was a gradual cessation of involuntary movement over weeks or months. In individual patients there was a similarity between speed of onset and suddenness of remission. To what extent these findings are compatible with the currently accepted notion of an organically determined focal dystonia remains speculative but whatever the explanation, the more favourable natural history would seem to commend patient and conservative management.

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