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OPERATIVE TREATMENT OF CONGENITAL MUSCULAR TORTICOLLIS: LONG TERM RESULTS OF MYOTOMY

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Abstract. Myotomy was performed in 40 patients under 10 years of age with typical congenital muscular torticollis. Follow-up studies were carried out on 29 out of 40 cases. The patients without muscle relief of the affected sternomastoid muscle after myotomy had good range of motion of the neck. On the other hand, the patients who had undergone muscle release had, to a greater or lesser extent, disturbances of the range of movement. For complete release of contracture, total excision may be the only operation in younger cases.

A considerable amount of research has been reported on the pathogenesis of congenital muscular torticollis. There is, however, no uniformity of thought as to its etiology or treatment. Pathological studies have demonstrated that the sternomastoid muscle undergoes degeneration of muscle fibers and proliferation of the connective tissue among muscle fibers. At the final stage of the typical wryneck, the muscle fibers, as observed macroscopically and microscopically, change into a hard tendinous cord and this scar tissue spreads over the whole length of the sternomastoid muscle, and are permanently shortened and inelastic (1, 2). It can, therefore, be said that the typical wryneck is due to contracture of the sternomastoid muscle, caused by such scar tissue. The authors consider that it can only be improved by surgical release of the contracted muscle.

From the 19th century, several methods of myotomy have been recommended as the first choice by Stromeyer (3), Volkmann (4), Lange (5) and Hellstadius (6). Mikulicz (7), however, recognized the inadequacies of these methods and reported a series of patients upon whom he did complete excision of the sternomastoid muscle. This did not become popular in Japan because of its technical complexity and disappearance of the muscle relief after the operation.

In the treatment of the typical wryneck, the aim of the surgeon is to correct the distorted appearance of the wryneck rather than to improve the limitation of neck movements. This means that the purpose of the treatment is cosmetic rather than functional, so factors to be taken into consideration include minimizing the operation scar and preserving the muscle relief of the sternomastoid muscle after the operation. Despite the various reports on the technical proce-

dures of operative treatment of wryneck, there have been few publications of follow-up studies of such cases. The present report was a follow-up of such a series and an analysis of the extent of preservation of muscle relief, the role of facial scoliosis and deformity of the skull, and the recurrence of contractures.

MATERIALS AND METHODS

Follow-up studies were carried out in 29 of 40 myotomy cases under ten years of age who were treated in the orthopedic clinic of Okayama University Medical School from June 1954 to May 1965. Twelve males and 17 females were re-examined in 1967 and also in 1973. The longest follow-up period was 18 years and the shortest was 5 years.

RESULTS

Preservation of muscle relief post-operatively. As shown in Table 1, muscle relief was completely lost in most of our patients without limitation of neck movements. Even in the few cases where muscle relief existed, it was a thin and cordlike relief and apparently different from the normal figure. Moreover, in the patients with muscle relief of a considerable size, a greater or lesser contracture of the sternomastoid muscle and limitation of the neck movements was noticed.

TABLE 1. CLINICAL FINDINGS IN PATIENTS TREATED BY MYOTOMY AT FINAL FOLLOW-UP

Case	Age/Sex	Ope. yrs	Follow up yrs	Facial Scoliosis		EMD		Plagio-cephaly		ROM		Relief		Comments
				Preop.	Postop.	Preop.	Postop.	Preop.	Postop.	Rot.	Tilt	St.	Cl.	
1	18/F	5/12	18	+	−	45 50	66 65	−	−	→	→	—	thin cord	with CDH
2	18/M	5/12	17	−	−	45 45	74 73	+	+	→	→	thin cord	—	
3	5/F	6/12	5	+	−	58 58	65 66	+	+	→	→	norm.	thin cord	
4	8/M	8/12	8		−	50 60	66 63		+	→	→	—	—	
5	16/F	9/12	15	−	−		67 67		−	→	→	—	—	
6	12/F	9/12	11	+	−		72 70	+	+	→	↓	—	cord	
7	8/F	10/12	7	−	−	52 52	60 60		+	→	↓	—	cord	with CDH Re-ope
8	9/F	10/12	8	−	+		62 68	+	+	↓	↓	cord	cord	Re-ope
9	16/M	1	15		−	53 55	74 73	+	−	→	→	norm.	thin cord	with CDH
10	16/F	1	15		−	60 60	72 70	−	−	→	↓	—	cord	
11	13/F	1	12		−		73 73		+	→	↓	—	cord	
12	6/M	1	5	+	+		65 60			↓	↓	cord	cord	Re-ope

TABLE 1 Continued

Case	Age/Sex	Ope. yrs	Follow up yrs	Facial Scoliosis Preop.	Facial Scoliosis Postop.	EMD Preop.	EMD Postop.	Plagiocephaly Preop.	Plagiocephaly Postop.	ROM Rot.	ROM Tilt	Relief St.	Relief Cl.	Comments
13	15/M	2	12	+	+		76 82		+	↓	↓	cord	—	
14	13/F	2	11	+	—	60 60	75 75		—	→	→	—	—	
15	13/F	2	11	—	—		70 65		+	→	↓	—	cord	Re-ope at other hosp. in 1967
16	16/F	3	13	+	—	65 60	72 74		+	→	↓	—	cord	
17	16/M	4	12	+	—		78 77		—	→	→	thin cord	thin cord	
18	11/F	5	6	+	+		64 61		+	→	→	—	—	
19	15/M	5	9		—		72 71	+	—	→	→	—	—	
20	24/M	6	18		—		76 75		—	→	→	—	—	
21	23/F	6	17	+	—		75 75		—	→	→	—	thin cord	
22	25/M	7	18		+	70 65	80 75		—	→	↓	—	cord	
23	26/F	8	18		—		67 66		—	→	↓	—	cord	
24	23/F	8	15		—	64 68	70 70		—	→	→	—	thin cord	
25	14/M	8	6	+	—		73 73		—	→	↓	—	cord	
26	18/F	9	9	+	—	63 66	70 72		—	→	→	—	—	
27	23/F	9	13	+	—	60 65	70 72		—	→	→	—	thin cord	
28	22/M	9	12	+	—		80 80		—	→	→	—	thin cord	
29	16/M	9	7	+	—	69 67	73 72		+	→	→	thin cord	thin cord	

Abbreviations used in Tables 1 and 2

Ope···operation
EMD···eye mouth distance
Preop. ···before operation
Postop···after operation
ROM···range of motion
Rot. ···rotation of the head
Tilt. ···tilting of the head
St. ···sternal portion of the sternocleid muscle
Cl···clavicular portion of the sternocleid muscle
→···normal range of motion
↓ ···disturbance of motion
CDH···congenital dislocation of the hip

Case 19 is a 15 year old male whose muscle relief of the right sternomastoid is completely lost after ten postoperative years (Volkmann's method) (Fig. 1).

Case 2 is an 18 year old male with right wryneck who has a thin cord remaining in the right sternomastoid muscle without limitation of the neck movements at 17 postoperative years (Hellstadius' method). A small muscle relief is visible on his right sternomastoid muscle but it is different from a normal one (Fig. 2).

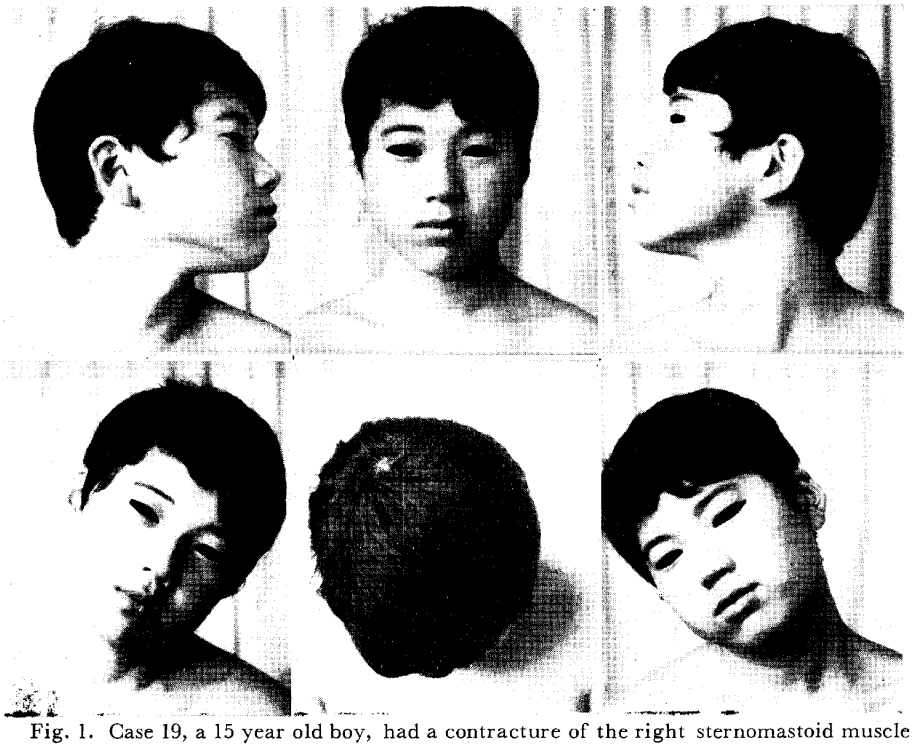

Fig. 1. Case 19, a 15 year old boy, had a contracture of the right sternomastoid muscle which was treated by myotomy (Volkmann's method). 10 years postoperatively, there was no relief of sternomastoid muscle on his right side neck.

Case 10 is a 16 year old female with a right wryneck who was treated by myotomy (Volkmann's method) and, when re-examined at nine and 15 postoperative years, had no subjective complaint. While relief of the clavicular portion of the sternomastoid muscle is cord-like, the relief of the sternal portion has disappeared completely. The release of contracture itself is still inadequate (Fig. 3).

As is obvious from Table 1, there was no case in which muscle relief and range of motion of the neck was normal. On the other hand, a slight limitation of neck movement was present in most cases where muscle relief was preserved. Five of the cases who underwent operation under the age of three had recurrent wryneck.

Postoperative estimate of the end results of myotomy. The cases who visited our clinic for both examinations in 1967 and 1973 comprised to 15 cases as shown in Table 2. No patient had any worsening of local findings in the period between 1967 and 1973, even patients over seven years old and over two postoperative years in 1967. Moreover, no worsening in clinical symptoms was observed after

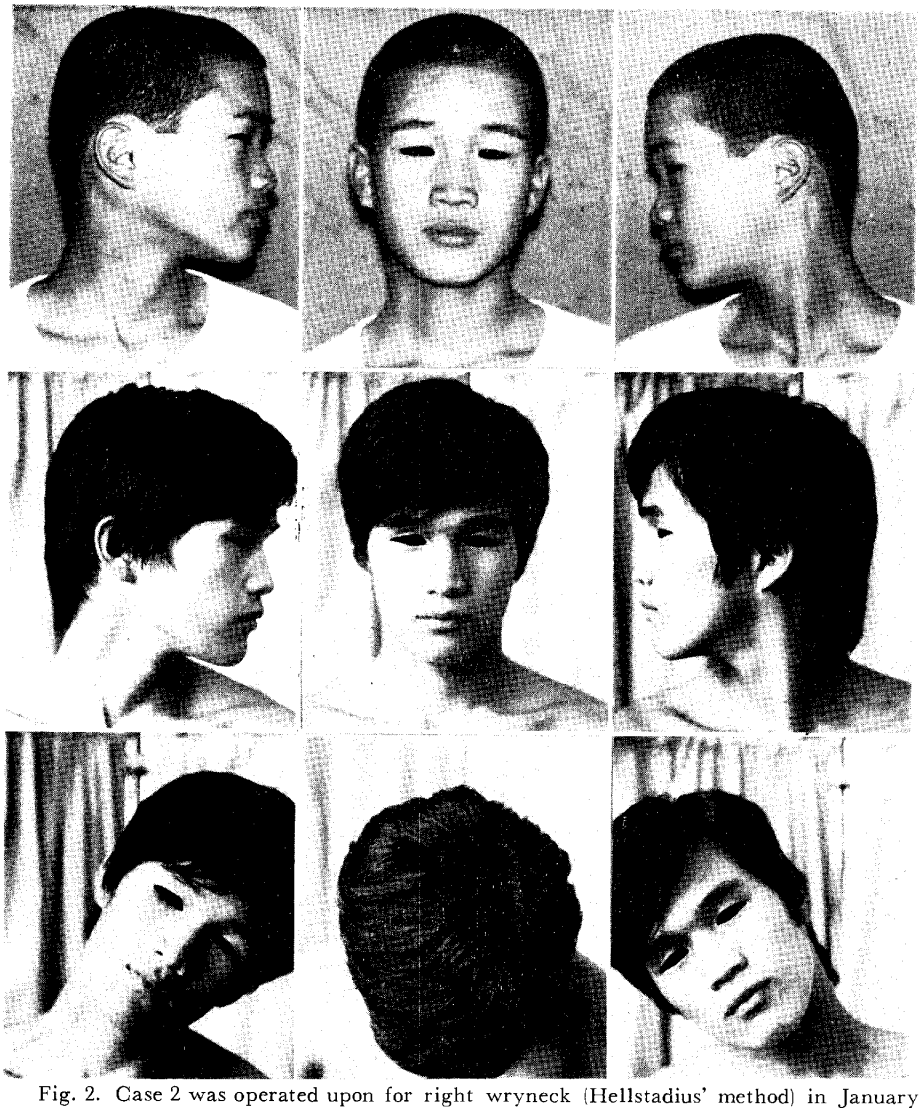

Fig. 2. Case 2 was operated upon for right wryneck (Hellstadius' method) in January 1956 and was referred in November 1967 and October 1973 for re-examination. Top lining photos were taken at 12 years of age and middle and lower at 18 years of age.

the age of 10 years even if the contracture still remained postoperatively. This result may be related to the annual increment of sternomastoid muscle in length (8).

Facial scoliosis. Facial scoliosis was improved in our patients following the elimination of the sternomastoid muscle contracture, while patients with a recurrence of the contracture had apparent facial scoliosis.

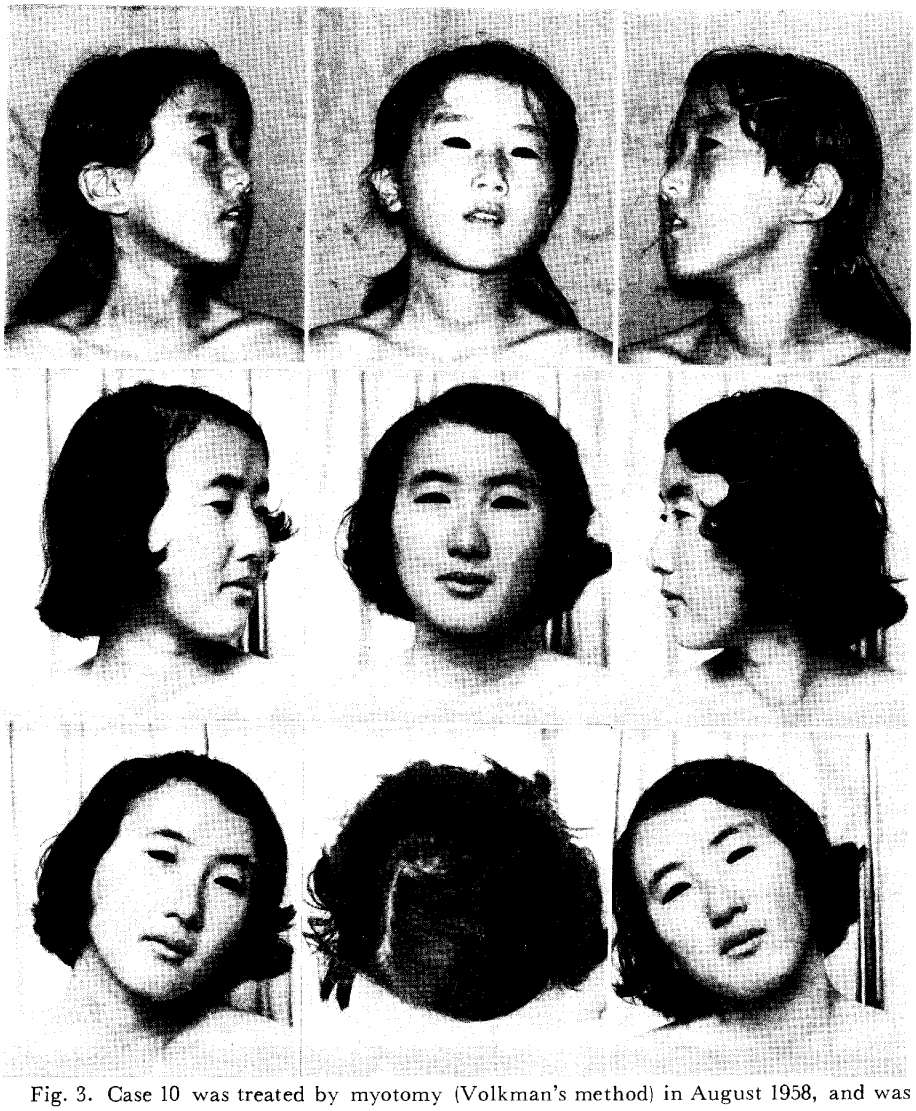

Fig. 3. Case 10 was treated by myotomy (Volkman's method) in August 1958, and was re-examined in August 1967 and December 1973. Top lining photos were taken at 10 years of age and middle and lower at 16 years of age.

Case 8 is a 14 year old female who underwent operation at 10 months of age (Lange's method). At re-examination at eight years of age, she had a recurrence of the deformity. At 9 years of age, she was treated by total excision of the right contracted sterno-mastoid muscle. Four years after the second operation, her facial scoliosis had improved (Fig. 4).

Case 13 is a 15 year old male who underwent operation at one year of age

TABLE 2. COMPARISON OF CLINICAL FINDINGS IN PATIENTS FOLLOWED UP FROM 1967 TO 1973

Case	Age/Sex	Ope yrs	Follow up yrs.	Facial Scoliosis	EMD	Plagiocephaly	ROM		Relief		Comments
							Rot.	Tilt	St.	Cl.	
1	18/F	5/12	11	—	62-63	—	→	→	—	thin cord	with CDH
			18	—	66-65	—	→	→	—	thin cord	
2	18/M	5/12	11	—	69-69	+	→	→	thin cord	—	
			17	—	74-73	+	→	→	thin cord	—	
6	12/F	9/12	5	—	63-61	+	→	↓	—	cord	
			11	—	72-70	+	→	↓	—	cord	
7	16/F	10/12	7	—	58-61		↓	↓	cord	cord	with CDH
		8	8	—	65-65	+	→	↓	—	cord	Re-ope
8	14/F	10/12	8	+	62-68	+	↓	↓	cord	cord	Re-ope
		9	4	—	69-72	+	→	→	—	—	total excision
9	16/M	1	9	—	65-64	+	→	↓	norm.	thin cord	with CDH
			15	—	74-73	—	→	→	norm.	thin cord	
10	16/F	1	9	—	68-70	—	→	↓	—	cord	
			15	—	72-70	—	→	↓	—	cord	
11	13/F	1	6	—	63-64	+	→	↓	—	cord	
			12	—	73-73	+	→	↓	—	cord	
13	15/M	2	7	+	63-68	+	↓	↓	cord	—	Re-ope was recommended
			12	+	76-82	+	↓	↓	cord	—	no treatment
14	13/F	2	6	—	66-67	—	→	→	—	—	
			11	—	75-75	—	→	→	—	—	
19	15/M	5	2	—	65-60	+	→	→	—	—	
			9	—	72-71	—	→	→	—	—	
21	23/F	6	11	—	75-74	—	→	↓	—	thin cord	
			17	—	75-75	—	→	→	—	thin cord	
23	26/F	8	12	—	69-67	—	→	↓	—	cord	
			18	—	67-66	—	→	↓	—	cord	
24	23/F	8	9	—	69-69	—	→	↓	—	thin cord	
			15	—	70-70	—	→	→	—	thin cord	
27	23/F	9	7	—	67-69	—	→	→	thin cord	—	Right eye is blind
			14	—	70-72	—	→	→	thin cord	—	

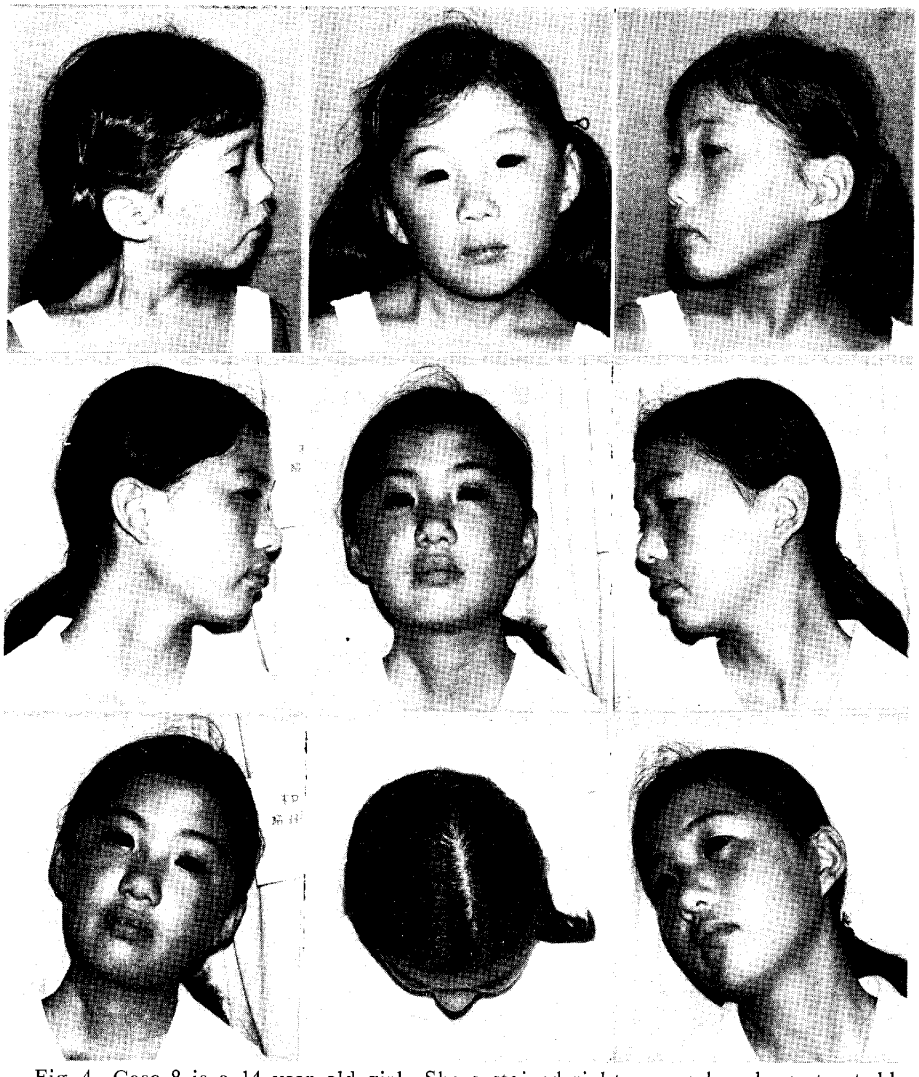

Fig. 4. Case 8 is a 14 year old girl. She sustained right wryneck and was treated by myotomy (Lange's method) in May 1959. In August 1967, she was referred for examination and had a contracted right sternomastoid muscle. In January 1968, she was treated by total excision of a contracted muscle. In August 1972, there is no muscle relief on her right side neck. Top lining photos were taken before re-operation and middle and lower after re-operation.

(Hellstadius' method). Six years after operation, he had a recurrence of the deformity, especially with a contracted sternal portion of the right sternomastoid muscle. Twelve years after operation, he had limitation of neck movement and facial scoliosis. However, these conditions have not shown any degeneration since 1967 (Fig. 5).

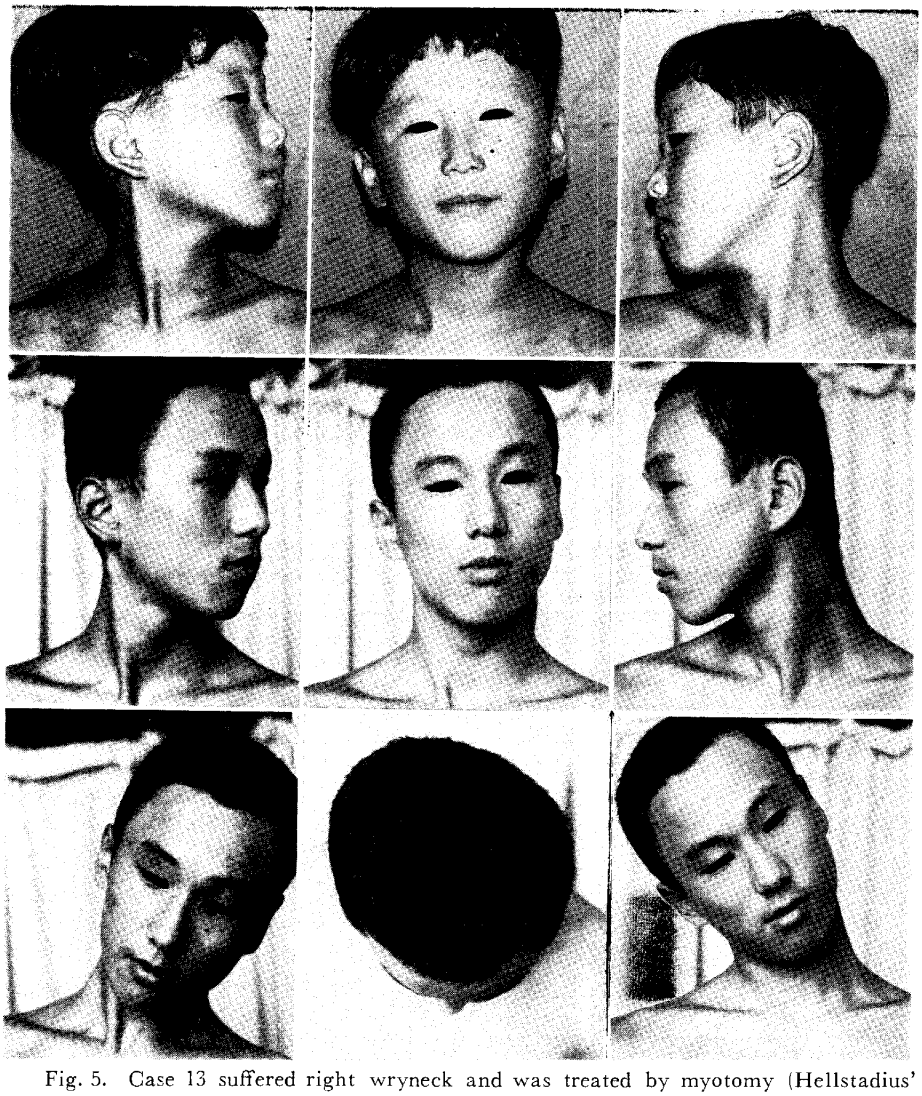

Fig. 5. Case 13 suffered right wryneck and was treated by myotomy (Hellstadius' method) in December 1960. In August 1967, he was re-examined and had a contracted right sternomastoid muscle. This boy was recommended re-operation but had no treatment. In August 1973, his local findings were the same as previously. Top lining photos were taken at seven years after operation and middle and lower at twelve years after operation.

Deformity of the skull. This deformity of the occipital skull is called plagiocephyaly. In most of our patients, plagiocephaly still remained after release of the contracture. Those undergoing operation under three years of age had plagiocephaly more frequently than those operated on over three years old as seen in Table 1. The degree of wryneck did not seem to be related to the degree

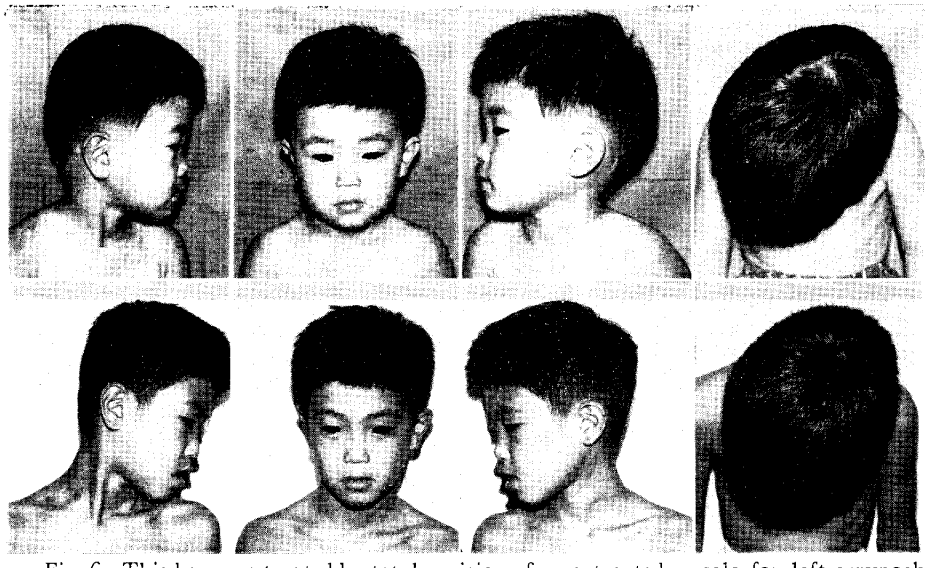

Fig. 6. This boy was treated by total excision of a contracted muscle for left wryneck at one year of age. He was re-examined in August 1967 and 1972. The upper photos were taken at one year postoperatively and lower at six post-operatively.

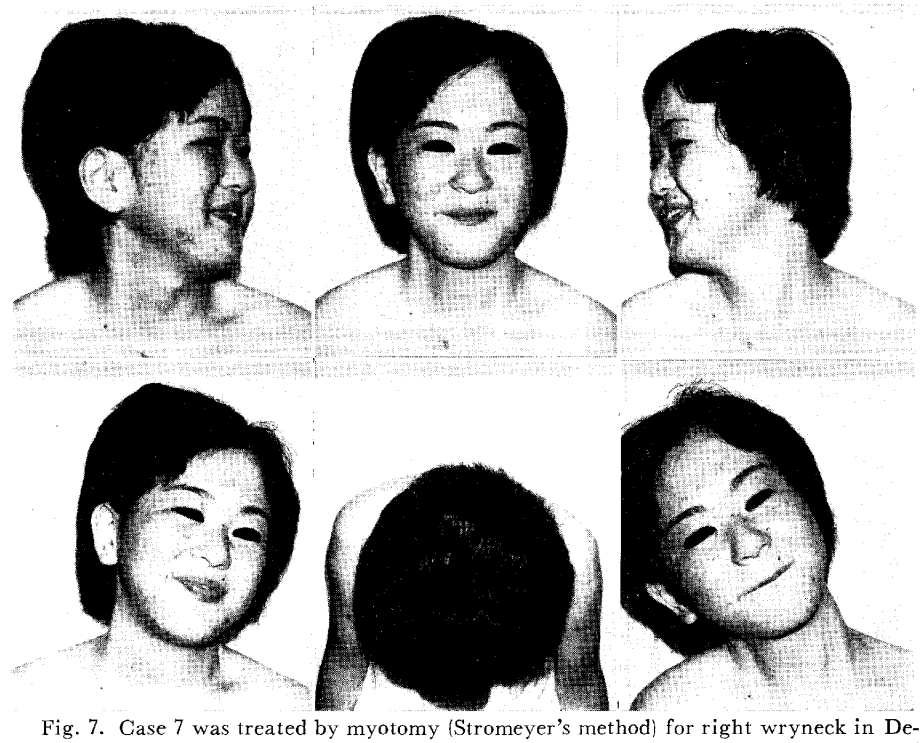

Fig. 7. Case 7 was treated by myotomy (Stromeyer's method) for right wryneck in December 1957 at 10 months of age. In May 1965, she was treated by myotomy (Hellstadius' method) for recurrent wryneck. In July 1973, the lateral flexion of her neck to the left was disturbed slightly.

of plagiocephaly. Post-operatively, this deformity may not improve as easily as is generally considered.

The patient showed in Fig. 6 is an eight year old male who was operated upon at age of one year. In spite of complete improvement of the contracture, his occipital deformity remains six years after operation.

Recurrence of contracture after myotomy. The complete elimination of contracture after myotomy was seen in some cases as mentioned above. The myotomy operation gives, however, unsatisfactory results in terms of complete release of contracture, even if there is no need of re-operation.

Case 7 is a 16 year old girl who was treated by myotomy twice for wryneck. First operation was done in December 1957 and second operation in May 1965. Eight years after the second operation the contracture of the right sternomastoid muscle remained (Fig. 7).

DISCUSSION

Since Volkmann, it has been well demonstrated that a considerable number of cases lose muscle relief after myotomy. To preserve the muscle relief, Fränkel (9) and Springer (10) devised a rational plan for the myotomy operation and Imai (11) recommended that the operation should be performed in the nursing period under one year old. It seems desirable but difficult to remove the contracture while preserving the muscle relief at such a young age. On the other hand, it is generally accepted that most cases of wryneck in early infancy heal spontaneously without any treatment. For this reason, the present authors do not perform surgical operation on patients under one year of age. Taking spontaneous heal ing into consideration, the early stage operation recommended by Imai cannot be considered justified.

Opinions differ on the question of whether or not the deformity usually seen in the typical wryneck is caused by the abnormal pulls caused by contracture in the sternomastoid muscle during the growing period of the child. There is also a difference of opinion as to the recovery of facial scoliosis after release of the contracted sternomastoid muscle (12, 13). The facial scoliosis is usually designated as the discrepancy of the distance between the outer corner of the eye-lid and that of the lip on both sides, but this does not appear to correspond to the subjective impression.

Flattening of the occiput on the side opposite the neck deformity is observed in most patients with wryneck. According to some papers (14, 15) the degree of skull deformity is related not to that of wryneck, but to the asymmetrical head position and the consistency of the skull.

Some workers suggest that contracture remains and leads to recurrence, especially in cases operated on in the youger age groups, as in our series. After

myotomy, scar tissue may proliferate from the muscle end to the clavicle, and the contracture may reocur later. It is, therefore, necessary to separate the cut end of the muscle as far as possible, but such procedures are quite difficult with children of young age.

Our analysis of the above considerations in the long term follow-up period offers some guide-lines for the treatment of wryneck in the future.

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