Original Article

Evaluation of Surgical Management of Congenital Muscular Torticollis using Bipolar Releases with Z-lengthening in BSMMU

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Abstract:
Congenital muscular torticollis is not an uncommon problem. We evaluated 14 patients who were operated by bipolar releases with Z-lengthening. Post operatively Halter traction was used for 20 hours for 6 weeks and only at night for another 6 weeks. Out of 14 patients 5 were male, 9 were female with mean age of 9.06 years (2-17 years) and mean follow up period was 3 years and 4 months (3 months to 7 years), with involvement of the right side in all the patients. Mean lateral flexion deficit improved 16° and mean rotational deficit improved 12°. According to modified Lee scoring system 3 patients had excellent, 7 good, 3 fair and 1 poor result. Cosmetic improvement was significant and patients and parents were happy. There were no post-operative complications. Scar was not an issue and one patient had residual band due to poor post-operative rehabilitation as the age of the child was 2 years. Bipolar release with Z-lengthening gives good to excellent results in most patients.

Key words: Congenital muscular torticollis, Bipolar release, Z-lengthening

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Introduction:
Congenital muscular torticollis (CMT) is a rare type of congenital musculoskeletal disorder presented as unilateral shortening of the sternocleidomastoid muscle (SCM)[1]. 'Torticollis' is derived from the Latin words 'tortus', meaning 'twisted' and 'collum', meaning 'neck'. In 1912 it was first defined by Tubby as "a deformity, congenital or acquired in origin, characterized by lateral inclination of the head to shoulder, with torsion of the neck and deviation of the face" [2].

CMT usually found in newborn infants or young children with incidence ranging from 0.3% to 2%. Due to effective shortening of SCM on the involved side there is ipsilateral head tilt and contralateral rotation of the face and chin[1].

Though exact etiology of congenital muscular torticollis is controversial but birth trauma, intrauterine malposition, infection and venous occlusion have all been implicated[1,2]. Treatment that can be offered are observation, use of orthotics, exercise programs, traction and various operations[3,4,6]. Surgical procedure is subcutaneous tenotomy, open tenotomy, bipolar tenotomy, and radical resection of a sternomastoid tumor or the sternocleidomastoid muscle[1,3,5,7-10].

Optimum treatment time is again controversial. Chandler and Altenberg[5] described 1-4 years of age as optimal. Ling[7] supported this idea. Coventry and Harris[6] extended the recommended age of treatment under 12 years. Ippolito[11], Tudisco[12] and Massobrio[13] and Lee, Kang and Bose[14] reported successful treatment on patients under 12 years. Most authors considered preschool age to be the ideal time for surgery because compliance and rehabilitation are easier in this age group. In younger age group, fitting of brace is difficult because of short neck and poor compliance and post-operative exercise is usually difficult in younger patients.

So, the aim of this study was to evaluate the outcomes of bipolar releases with Z-lengthening of CMT in patients and parent's satisfaction, correction of the deformities, per and post-operative complications and relapse of the deformity.

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Materials and Methods:
Total fourteen children of CMT were operated in BSMMU during the period from March 2011 to January 2018. All the patients presented lately at school age, except one who presented at preschool age. Many times, parents were not aware of the problems or neglected the situation. Two patients were lost from the follow up. Ophthalmic and bony pathology of the cervical spines were excluded by anterior-posterior and lateral radiographs. The passive range of movements of lateral flexion and rotation were measured clinically with a goniometer and compared with the opposite side in order to calculate the deficit. Our patients did not have any congenital anomalies like DDH, metatarsus adductus and no patient was recurrent case. No patients had the history of surgery for the same and the cause of torticollis was not determined in any patient.

All patients were operated under general anesthesia and modified Ferkel's procedures (Ferkel D) with bipolar release of the sternomastoid with Z-lengthening of the sternal head[15,16]. The patients were supine with the neck turned to opposite side. A bolster was kept in the interscapular area to keep the neck extended to make taught the sternomastoid. Two incisions were used. A small retro-auricular incision was made just below the mastoid tip and the proximal head of the sternomastoid was identified, tendon was freed from the bone, cut by diathermy after identification of the distal attachment and allowed to slide distally. A second supraclavicular incision was made 1 cm above the medial end of the clavicle, after dividing all tight fascial sheaths, sternal and clavicular heads were exposed. Clavicular head was completely cut with diathermy and Z-lengthening of the sternal head was performed and after gentle manipulation stretching, sterno-mastoid tendon ends were approximated with 3-0 absorbable suture to preserve the cosmetic V shape of the neck. Subcuticular closer were done in layers to reduce the likelihood scar formation. Postoperatively Halter traction was used for 12 weeks (more than 20 hours in a day for initial 6 weeks and then another 6 weeks at night only) and cervical collar in the remaining hours for initial 6 weeks. Gradual stretching, lateral flexion, and rotational exercises, out of collar as comfort permits and continued for 3 months.

Results were evaluated by using modified Lee scoring system[17,18], which assessed both function (movements of the neck) and cosmesis. Cosmesis was assessed by head tilt, scaring and lateral band formation. In Modified Lee scoring system excellent results corresponded to 17-18 points, 15-16 points as good, 13-14 points as fair and <12 points as poor results[18] (Table-I).

The changes in the mean ranges of movements following surgery were compared for statistical significance using paired t-test and p value <0.001 was considered as significant results. Statistical analysis was done using SPSS v22 software.

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subjects of this article.

The researchers were duly concerned about the ethical issues related to the study. Formal ethical clearance was taken from Ethical Review Board of BSMMU before commencing the study. Confidentiality was maintained properly and these data were not used other than the study.

Table-I: Scoring system for assessment of congenital muscular torticollis (modified from Lee et al.[18])

Points	Neck movement	Head tilt	Scar	Loss of column	Lateral band	Facial asymmetry
3	Full	None	Fine	None	None	None
2	<10° LOR or side flexion	Mild	Slight	Slight	Slight	Slight
1	10°-25° LOR or side flexion	Moderate	Moderate	Obvious but cosmetically acceptable	Obvious but cosmetically acceptable	Moderate
0	>25° LOR or side flexion	Severe	Unacceptable	Unacceptable	Unacceptable	Severe

(LOR= Limitation of Rotation)

Results:
Out of 14 patients 5 were male and 9 were female (Figure-1) with a mean age of 9.06 years (2 to 17 years) in the study. Right sides were involved in all the patients. Mean follow up periods was 3 years 4 months (3 months to 7 years). Mean lateral flexion deficit of the neck improved from 23.71° (15°-35°) pre-operatively to 8.5° (5°-15°) post-operatively, demonstrating a mean improvement of 15.21°. The mean rotational deficit improved from 15.57° (10°-20°) preoperatively to 2.5° (0°-5°) post-operatively with a mean improvement of 13.07°. There was improvement in functional range of Motion (ROM) in all patients (Table-II). These improvements were statistically significant (p<0.001). Cosmetic improvement in the form of reduction in the head tilt

& chin deviation were present in all patients (Figure-2). The V shape of the neck at sternum was retained in all patients. According to modified Lee scoring system 3 patients had excellent, 7 good and 3 fair and 1 poor result. The appearance of the scar was excellent in 6, good in 8 patients with no hypertrophy or keloid formation.

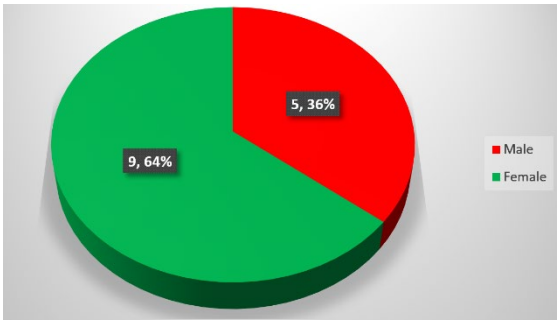

Figure-1: Gender distribution of patients (n=14)

Table-II: Mean improvement of rotational and lateral flexion deficit between pre and post-operative state (n=14)

Deficit	Pre-operative (Mean±SD)	Post-operative (Mean±SD)	P-value
Rotational Deficit (0°)	15.57±3.90	2.5±2.59	<0.001
Lateral Flexion Deficit (0°)	23.71±6.39	8.5±3.61	<0.001

P value reached from paired t-test

Table-III: Outcome of bipolar releases with Z-lengthening surgery of congenital muscular torticollis following modified Lee scoring system

Overall outcomes	Excellent	Good	Fair	Poor
Craniofacial asymmetry	9	4	1	0
Scar	6	6	0	1
Residual contracture	8	5	0	1
Cosmetic satisfaction	8	5	0	1
Functional satisfaction	7	6	0	1

Pre-operative

Per-operative

Pre-operative

1 year Post-operative

Pre-operative

5 years & 7 months Post-operative

Pre-operative

9 months after surgery

Halter traction

Cervical Collar

Pre-operative

3 years Post-operative

Figure-2: Congenital Muscular Torticollis (CMT): Pre- & per-operative view and outcomes after surgery and related procedures

The residual appearance of sternomastoid showed 8 excellent, 5 good and 1 poor result. 1 patient had residual band due to poor post-operative rehabilitation as the age of the child was 2 years at the time of surgery. The level of cosmetic satisfaction was 8 excellent, 5 good, 1 poor result and the level of functional satisfaction was excellent in 7, 6 good and 1 poor result (Table-III). No intraoperative complications like external jugular vein, nerve injuries were encountered in any patients.

Discussion:

The primary goals of surgery for congenital muscular torticollis are improvements in the head tilt, neck movement and cosmesis. Although timing of surgery is controversial, but most authors agreed that the preschool age is the ideal time. Good or excellent results can be achieved in older age group (adolescent) and compliance with post-operative rehabilitation is the most important consideration for optimum results and surgery should be delayed until such complacence is possible.

We made incisions away from the clavicle and used subcutaneous sutures and thus scar was minimum and not a problem in cosmesis, results noted by other authors[5,6,13]. The most popular methods are bipolar release[8,9,16,17] for severe deformity and in older patients[14,15] as we did in all our cases.

Post-operative immobilization of the head and neck in overcorrected position has been claimed to give better results[19] and some authors recommended of cast brace alone with early exercise. Cheng and Au[3] had good clinical results with post-operative bracing and rehabilitation in children older than 2 years. Ippolito et al.[25] suggested that a long-term Minerva cast could prevent recurrence. We used cervical traction (Halter traction) for initial 6 weeks more than 20 hours and another 6 weeks at night only with excellent and good results.

Another controversial issue is Z-lengthening of the sternomastoid. If a bipolar release is performed without Z-lengthening, loss of normal contour of the neck may affect the cosmetic appearance[7]. We did bipolar release with Z-lengthening in all cases with all patients reported improve movements in neck and head tilt and good cosmetic results except one with retaining of the normal contour of the sternomastoid.

Rate of recurrence is high (0.8-20%) in many series[20,21-24] but in our series only 1 patient developed tight sternomastoid post operatively because of poor compliance in post-operative rehabilitation as the age of the child was only 2 years, and other reason could be due to inadequate release of the sternomastoid[25-28].

Limitations of the Study:

Limitations of our study are small sample size, rational and lateral flexion deficit were assayed by a single assessor (Islam MA). We did not specifically measure facial asymmetry but our patients and parents were happy with the improvement in facial asymmetry and overall cosmetic results.

Conclusion:

In conclusion, surgical management of congenital muscular torticollis using bipolar release of the sternomastoid and Z-lengthening is a time tasted safe, effective procedure, which gives excellent clinical, functional and cosmetic results with minimum complications.

Conflict of Interest:

The authors declare that there is no conflict of interests regarding the publication of this paper.

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