Original Research Article

Bipolar release with Z-plasty in correction of neglected congenital torticollis in adolescent patients

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Abstract

Purpose: Neglected congenital muscular torticollis (CMT) is a common presentation in India and other developing countries, coming with cosmesis as the main concern. Very few publications are available regarding management of delayed presentation in adolescent population. The primary aim of this study is to evaluate the results of bipolar release with z-plasty in adolescent population.

Methods: Five Adolescent patients aged from 10-18 years with neglected CMT were treated using bipolar release of sternocleidomastoid muscle with z-plasty lengthening of the sternal end along with skin lengthening using z-plasty proximally. Postoperatively soft cervical collar for six weeks followed by physiotherapy was done. Study was done in NRI Medical College, Chinnakakani and Dr. Pratap Hospital, Vijayawada.

Results: Average follow-up of one year was done, excellent cosmetic results were noted in three patients and good in two patients. Postoperative betterment in cosmesis, range of motion, chin deviation and head tilt were present in all patients. Neither complications nor recurrences requiring redo surgery occurred in these five patients.

Conclusions: Study concludes that patients with neglected CMT presenting in adolescence benefit from surgery. Bipolar release of muscle with skin lengthening using z-plasties is an appropriate method for neglected adolescent patients.

Keywords: Bipolar lengthening, adolescent's torticollis, adult torticollis, congenital muscular torticollis

Introduction

The term 'torticollis' is derived from latin word tortus meaning 'twisted' and 'collum' meaning neck. Congenital muscular torticollis (CMT) is the 3rd most common congenital musculoskeletal anomaly with a reported incidence of 0.3-1.9%. There is a slight male preponderance in cases, with ratio of 3: 2. The right side is more commonly involved.

Patients with CMT present with tilting of the head toward the involved side due to fibrosed and shortened sternocleidomastoid muscle ^[1]. The classical appearance related to torticollis includes recessed eyebrow and zygoma, inferior orbital dystopia on the affected side, commissural canting toward the affected side, deviation of the chin point and nasal tip, distorted craniofacial skeletal structures and inferiorly and posteriorly positioned ipsilateral ear ^[6].

Early diagnosed CMT can be treated conservatively with good or excellent results up to one year of age. In patients older than one year of age, corrective surgery has both aesthetic and functional benefits. Ling *et al.* have stated that the optimal time for surgery is between 1 and

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4 years ^[8]. For patients aged more than five year's efficacy of treatment iscontroversial. Some authors have reported that operative treatment is of little value after this age, and the results are even worse when the operation is done after puberty and may lead to more complications ^[9]

Neglected CMT is common in India, where patients commonly presenting at an age of more than ten years. Little has been published on the management of such patients. The aim of our study was to evaluate the results of bipolar release in this age group.

Materials and methods

Over a period of five years, five patients of adolescent age group were operated on for neglected CMT. Inclusion criteria included age 10-18 years and no history of previous surgery. Birth and family history data were noted. Pathological causes have been ruled out that cause torticollis. Preoperative assessment was done as per the Lee *et al.* scoring system of restriction of neck movements and head tilt and facial asymmetry. Study was done in NRI Medical College, chinnakakani and Dr. Pratap Hospital, Vijayawada.

Release of both the inferior and the superior head of Sternocleidomastoid was performed (i.e., Bipolar release) in all cases. Inferior release was done through an incision 1 cm above the medial third of the clavicle. The clavicular head was released completely while the sternal head was lengthened by z-plasty. The mastoid head was released though an incision just below the tip of the mastoid process. All tight fascial sheaths were released, taking due precaution not to injure any neurovascular structure. It was not necessary to release the carotid sheath in any of our patients. An additional Z-plasty has been done infra auricularly to the skin to gain extra length.

The postoperative protocol included soft cervical collar for six weeks followed by physiotherapy, including both active and passive movement. After six weeks, soft cervical collar was applied during the night only. Patients were reviewed every three weeks for three months, six-weekly for one year. At each follow-up, neck range of movement (ROM), head tilt and craniofacial asymmetry were assessed. At the final assessment, postoperative complications (including scarring, loss of sternomastoid column, and lateral bands) were recorded. A scoring system modified from Lee *et al.* [10], which included function and cosmetic results, was used. An excellent result corresponded to 17-18 points; a good result to 15-16 points; a fair result to 13-14 points; and a poor result to less than 12 points (Table 1).

Table 1: Scoring system for assessment of congenital muscular torticollis; modified from Lee *et al.* [10] (LOR is limitation of rotation)

| 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 |

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Results

03 of the patients were boys while 02 were girls, and their ages at presentation ranged from 10 to 18 years. 4 of the patients had involvement of the right side, while one had involvement of the left side.

The mean follow-up for the patients was around one year. Excellent results were noted in two patients, good in three (Table 2). No surgery-related complications or recurrences requiring surgery occurred in any of the patients.

| Patient | Age (completed years) | Sex | Side | Postop score at final follow-up | Result |
|---------|-----------------------|-----|------|---------------------------------|-----------|
| 1 | 14 | F | R | 16 | Good |
| 2 | 18 | M | L | 15 | Good |
| 3 | 17 | F | R | 17 | Excellent |
| 4 | 15 | M | R | 17 | Excellent |
| 5 | 10 | M | R | 15 | Good |

Table 2: Results and patient details

Postoperatively there was improvement in the functional range of movement in all patients. Restriction of movement was 10– 25° in only one patient. Others had movements within normal limits or a restriction of less than 10° . Cosmetic improvement in the form of reduction in head tilt and chin deviation was present in all patients. Postoperative head tilt was mild or fully corrected in ten of the patients, while none had an unacceptable head tilt postoperatively. The V-shape of the neck at the sternum was retained in all 05 patients, as we performed a z-plasty of the sternal end of the muscle. There was no cosmetically unacceptable scar visible at either of the two surgical sites in any of the patients.

Neglected Torticollis - Never too late for surgical correction? Dr. Pratap Duggirala



Fig 1: Preoperative images of a 14 year old girl with severe torticollis on right side showing shortened and prominent right sternocleidomastoid, head tilt to right, chin deviation to left, right side hemihypoplasia of face, elevation of right shoulder



Fig 2: Intraoperative images - BIPOLAR RELEASE was performed (release of both inferior and

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superior attachments of sternocleidomastoid).

Inferior Release

Skin incision was a transverse incision 1 cm above clavicle region, and for underlying SCM muscle, clavicular head was released completely while sternal head was lengthened by z plasty.

Superior Release

Skin incision was a planned z plasty for lengthening and underlying SCM muscle release was transverse including all fibrous fascia bands.



Fig 3: Postoperative images after 3 weeks showing marked improvement in head tilt, chin deviation, marked improvement in head rotation and correction of the shoulder elevation. Right side hemihypoplasia has no improvement

Discussion

Most cases of CMT resolve completely, either spontaneously within months after birth or following the early initiation of conservative measures such as gentle controlled passive manual stretching exercises on the affected side ^[5,7]. Sonmez *et al.* found that 95% of the patients diagnosed and treated effectively before an age of one year did not need surgical treatment ^[11].

Conservative management is usually tried before an age of one year. Nonoperative therapy after the age of one year is rarely successful ^[12]. The goals of surgical correction for older children are improvement in cosmetic deformity and cervical motion. The timing of surgery is controversial. Reversal of the craniofacial asymmetry is best achieved at an early age, when there is high growth and remodeling potential. Canale *et al.* ^[12] reported that full recovery of facial asymmetry after four years of age is difficult to obtain. Characteristically, there is flattening of the occiput contralaterally and depression of the malar prominence ipsilaterally, with downward displacement of the ear, eye and mouth on the affected side. Provided that the surgery is done while the patient is immature, these skeletal deformities may improve following surgery ^[13].

Late presentation of congenital muscular torticollis is quite common in developing countries like India, where many female patients present with cosmetic concerns just prior to a planned wedding. Lee *et al.* [10] and Minamitani *et al.* [14] reported that late release of the sternocleidomastoid muscle for patients more than six years of age could yield acceptable results. In a group of patients over the age of 26, Ippolito and Tudisco [15] reported that, although there was no resolution in facial asymmetry, there was improvement in the neck movements of all patients, and there were no complications. In contrast, Coventry and Harris [16] reported that the upper limit for good results after surgery for muscular torticollis is twelve years. Ling [17] also maintains that the benefit of treatment is limited over the age of five, and that the complication rate is high.

In our study, we observed functional and cosmetic improvements in all the patients. Our results of bipolar release with z-plasty point indicate that good results can be obtained in patients treated late provided that optimum surgery and rehabilitation are carried out.

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Although there are various surgical procedures for CMT, unipolar and bipolar release are the most popular. Subcutaneous tenotomy is not recommended, as it does not achieve adequate release, while total resection of the sternocleidomastoid muscle: The most effective method in older children-carries the significant risk of injuring the spinal accessory nerve. Bipolar releases are usually used in older children with a severe deformity. Wirth *et al.* [18], in a review of 55 patients with an average follow-up of fifteen years after surgical release, recommended that biterminal release should be performed at the age of 3-5 years in all patients who do not respond to nonoperative treatment. As advocated by Wirth *et al.*, we also believe that bipolar release combined with z-plasty preserves the normal V contour of the sternocleidomastoid and ensures a better cosmetic outcome.

The postoperative immobilization protocol for congenital muscular torticollis is also controversial (Fig. (Fig.4).4). The various techniques utilized include traction, cast, halo vest and collar. In the initial days following surgery, the patient has a tendency to keep the head in its former position in order to reduce pain, and compliance with the prescribed exercises is poor. If the head remains in this position, the released structures will regain their former tightness. Gentle cervical traction given immediately postoperatively ensures sustained correction. Once postoperative pain subsides, the patient can be shifted to a collar, and an exercise protocol can be initiated to ensure a satisfactory outcome. We ensured strict adherence to our postoperative protocol in all patients by providing regular follow-up and motivation to ensure a uniformly good outcome. Though our study does not establish improved outcomes with soft cervical collar, we strongly believe that postoperative cervical collar and strict adherence to physiotherapy protocol play important roles in helping the patient to overcome the abnormally adjusted position of the head and neck.

Facial asymmetry is the most significant factor affecting the cosmetic outcome. We obtained improvements in facial asymmetry in most of our patients. However, the extent of the improvement varied. We also found that the perception of facial asymmetry decreased markedly in the immediate postoperative period, indicating that whatever the bony changes that occur, they usually look compounded by the abnormal soft tissue structures. We believe that surgical bipolar sectioning of the sternocleidomastoid muscle should be considered even in adults with irreversible facial and skeletal deformities. The surgery restores the range of neck motion and resolves the head tilt; it can therefore improve quality of life.

We found that a greater restriction in neck ROM and a more severe head tilt (as seen preoperatively) were associated with a poorer final outcome. This finding is expected, but it should be noted that our results do not contraindicate surgery in such patients. However, there was no correlation between age of the patient or preoperative facial asymmetry and the final outcome. Thus, we believe that surgery should be performed in all cases of congenital muscular torticollis, however old the patient may be at presentation, and however severe the facial asymmetry.

One potential limitation of our study was the subjectivity involved in measuring the preoperative and postoperative variables, such as head tilt, neck ROM, facial asymmetry, scar, etc. We also didn't attempt to measure interobserver variability in the measurement, which is a potential drawback of this study.

To conclude, we believe that bipolar release is a very viable option for correcting neglected congenital muscular torticollis in adolescent. The procedure is much more effective than unipolar release or subcutaneous tenotomy, and is relatively complication-free and safe when compared to total resection of the sternocleidomastoid muscle. Lengthening of the sternal head by z-plasty restores the V shape of the base of the neck, which adds to the cosmesis, especially in a female patient. Postoperative soft cervical collargo a long way toward ensuring good to excellent results.

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