International Clinical and Medical Case Reports Journal
Case Report (ISSN: 2832-5788)

Case Description of a Child with Right-Sided Torticollis and Severely Limited Active Rotation in the Cervical Spine toward the Right Side due to Hypoplasia of the Sternocleidomastoid Muscle on the Left Side

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Citation: Anna Öhman. Case Description of a Child with Right-Sided Torticollis and Severely Limited Active Rotation in the Cervical Spine toward the Right Side due to Hypoplasia of the Sternocleidomastoid Muscle on the Left Side. Int Clinc Med Case Rep Jour. 2025;4(11):1-7.

Received Date: 22 November 2025; **Accepted Date:** 24 November 2025; **Published Date:** 25 November 2025

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ABSTRACT

At referral at the age of 1.5 years, the child showed an extreme difference in rotation of the head between active and passive motion toward the right side. He did not actively rotate his head more than slightly over a neutral position toward the right side but he had full active range of motion (AROM) toward the left side. Passive range of motion (PROM) was excellent in both rotation and lateral flexion bilaterally.

He tilted his head toward the right side, and had stronger lateral flexor muscles on the right side. As there was no limited PROM, this indicated a right-sided postural torticollis. However, the extreme problem of actively rotating toward the right side was disconcerting.

At the age of six years, hypoplasia of the sternocleidomastoid (SCM) muscle on the left side was revealed. The small amount of muscle mass on the left side explained the extremely limited active rotation toward the right side. The SCM muscle on the left side rotates the head toward the right side.

Conclusion: When torticollis with good PROM is combined with unusually large problems with AROM in rotation, hypoplasia/aplasia of the SCM muscle must be investigated.

Keyword: Hypoplasia; Sternocleidomastoid muscle; Infant; Child; Cervical active rotation

ABBREVIATIONS

AROM: Active range of motion

CMT: Congenital muscular torticollis

SCM: Sternocleidomastoid

MFS: Muscle function scale

PROM: Passive range of motion

PT: Physical therapy

MRI: Magnetic resonance imaging

INTRODUCTION

Torticollis is a sign not a diagnosis, meaning asymmetrical posturing of the head and cervical spine.[1,2] The SCM muscle is a two-headed neck muscle where the upper part is attached to the mastoid and the two lower parts are attached to the sternum and the clavicle. Asymmetric function of the SCM muscle can give an asymmetrical head posture. The most common cause of torticollis among infants is congenital muscular torticollis (CMT).[3] In CMT the SCM muscle is contracted or tense and has stronger muscle function on the affected side.[4,5,6] Rotation and lateral flexion can be measured with a big protractor[7,8,9,10,11] and muscle function examined using the muscle function scale (MFS).[12,13] However, torticollis can have other causes than a contracted or tense muscle.[14]

Muscular hypoplasia or aplasia is a rare phenomenon and is very unusual in the SCM muscle.[15-18] In some cases a part of the SCM muscle is missing and in some cases the whole SCM muscle is missing.[15] An MRI is used to confirm muscle aplasia.[19] Aplasia of the SCM muscle has been discovered in newborns[20,21] and in adulthood for others (Singh 2014) or discovered at dissection after death.[22,23] When hypoplasia or aplasia of the SCM muscle occurs, there is an imbalance in the neck that causes the head to tilt to the side with a normal SCM muscle, with the head turning to the opposite side. This head position can be interpreted as CMT if hypoplasia/aplasia is not discovered. In CMT the head is tilted toward the abnormal muscle and rotated to the opposite side.

The most known muscle when it comes to aplasia is the pectoral muscle.[24] Aplasia of the pectoral muscle is characteristic of Poland syndrome.[25] Aplasia of the trapezius muscle has been seen in three family members supporting the notion of a genetic link.[19] Absence of the whole SCM muscle or part of it has been seen in an infant, his father and grandfather, also pointing toward a genetic link.[26] Lung herniation due to aplasia of SCM has been reported in two infants.[20,27] An MRI is used to examine the absence of muscles.[18]

CASE

A child referred to physical therapy (PT) due to torticollis at the age of 1.5 years. He had a mild tilting of his head with lateral flexion toward the right side and a small rotation toward the left side. There was excellent PROM in both rotation and lateral flexion bilaterally. The right side was stronger than the left side, on the MFS scale he got scores of 3 and 1 (Table 1). He was diagnosed as having right-sided postural torticollis. However, it was extremely difficult for him to rotate actively toward the right side, when rotating his head toward the right side he turned his whole torso.

Table 1: Measures and estimates of PROM, AROM and MFS of case at age 1.5 years

Function	Right	Left
Passive rotation	>90°	>90°
Active rotation estimated	5-10°	65°
Passive lateral flexion, ear to shoulder	yes	yes
MFS scores	3	1

Due to a misunderstanding it was believed that an MRI had been carried out on his cervical spine. Later it was clear that the MRI had been carried out on the head to exclude craniosynostosis, although he seemed to have only minor developmental plagiocephaly. This was not evident when he got older, his head shape was in the normal span.

A lot of exercises for active rotation toward the right side were tested; this was a challenge for all as it was very confusing why it was so hard for him to perform active rotation toward the right side when there was no problem at all with PROM. At the age of 2.5 years, the lateral flexors on the left side had become stronger but there was still a minor head tilt toward the right. Strength training of the left side was continued. At four years of age, his head was in midline and he could rotate his head better to the right but not as well to the left. He was given a break from PT and was followed up again at five years of age. At the follow-up, he tilted his head toward the right again and had obvious problems rotating his head toward the right side without rotating his torso. It could first be surmised that the SCM muscle had not grown in length but he still had excellent PROM in his cervical spine. The problem of rotating his head to the right side had been a mystery from the first start of PT and different approaches to exercises, adjusted to his age, were adopted. In passive motion there was no problem at all to rotate his head 90° bilaterally. He could lie in supine position on the examination table with his head over the edge, with his head resting in the examiner's hands to unburden the weight of the head. He could rotate his head to about 90° by himself bilaterally.

At six years of age in April 2024, muscle hypoplasia was discovered as there was no visible sternal head attachment of the SCM muscle on the left side (Figure 1).

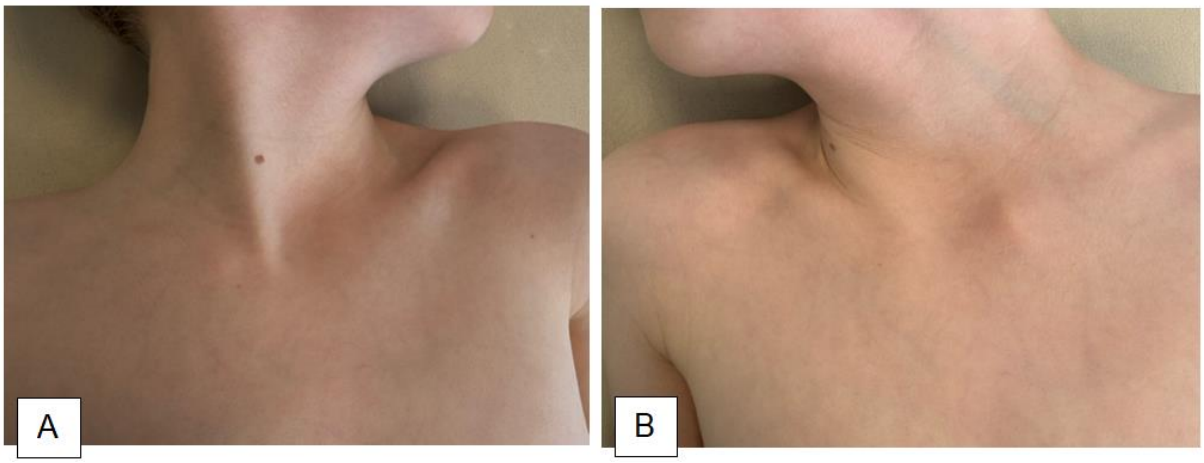

Figure 1: At the age of six years lying on an examination table, when rotating the head toward the left side there was a clearly visible sternal muscular attachment on the right side (A). When rotating the head toward the right side there was no visible muscular attachment on the left side (B).

A contact was taken with a plastic surgeon with long experience of torticollis. On parental agreement, a referral for examination was made. An MRI confirmed hypoplasia of the SCM muscle on the left side. Report after the MRI: "It was hard to follow the proximal attachment of the SCM muscle at processus mastoideus." Attachment at the clavicle and sternum differed from the right side, the muscle was about 4 mm on the left side compared

with 10 mm on the right side. An MRI was also done as it was important to rule out the absence of other neck muscles than the SCM. The other muscles in the neck were found to be symmetrical. If there had been hypoplasia of more muscles, it would have been necessary to rule out other problems.

DISCUSSION

Hypoplasia of the SCM muscle on the left side explains his problems with rotation toward the right side. The sternal part of the SCM muscle on the left side rotates the head to the opposite side. Less muscle mass on the left side affects the ability to rotate the head toward the right side. As muscle hypoplasia is rare, and very rare in the SCM muscle,[15-18] its occurrence is unexpected. Cases with hypoplasia/aplasia may look different. From the current case it can be learned to keep hypoplasia/aplasia of the SCM muscle in mind when there is an extreme discrepancy between AROM in rotation and no limited PROM in the neck at all.

The discovery of hypoplasia of the muscle helps to set reasonable expectations and goals that are achievable. A realistic plan of what needs to do be done to keep the head in midline and get the best possible function. Regular periods of training are probably functionally beneficial, keeping the left side as strong as possible because of the lower muscle mass in the left SCM muscle. Strategies for situations where less rotation could affect a situation could be implemented. For example, on a bicycle holding the handlebars, when looking to the right it may be a risk to turn the torso as this may turn the bicycle involuntarily. Individuals may have to learn to compensate for this. At a young age they can stop the bicycle and look, at an older age they can use mirrors on the bicycle to see people and cars etc. on the limited side.

Some articles describe the discovery of aplasia of the SCM muscle after death during anatomy dissections.[22,23] For others it has been discovered during the first days or months of life.[15,21] It is unknown if the cases discovered after death had any problems; a discussion probably not entered into with the families as it would have presented an ethical dilemma. However, photos could have revealed if they had a tilting toward the opposite side of the absent muscle or a rotation toward the missing muscle. It is unknown if some can keep the head in midline even with aplasia of the SCM muscle. Neglected CMT exists[28] and it can be speculated that there are probably some mild cases of neglected hypoplasia/aplasia of the SCM muscle without obvious problems. Singh at al found a 56-year-old with the absence of the SCM muscle. This man according to himself experienced no problems, and he was not willing to participate in further examination.[29]

It is important to exclude other problems when aplasia of SCM is discovered. Haroon and Beverley found herniation due to aplasia of SCM in an infant one hour after birth,[20] and Bayne et al found it in a young infant that needed surgery at the age of 4 months.[27]

Kumar et al described a four-year-old boy where the parents preferred PT as the mode of treatment.[15] The current case is too young to make his own decisions, and for the moment the parents prefer to continue with PT. With training and strategies to compensate the limited active rotation, this may be the best solution.

Only one case that underwent surgery release on the contralateral SCM muscle to correct torticollis is found in the literature, all other known cases seem to have been resolved well enough with PT.[30] The postoperative treatment received by the child that underwent surgery is not described.

The current case trained strength in cervical lateral flexion on the left side with hypoplasia of the SCM muscle. When he was doing a lot of training his head was in midline. If not doing enough his head tilt reoccurred and training had to be intensified again. It is preferable if surgery can be avoided but training will still be needed after surgery. The SCM muscle is about 4 cm in infanthood and about 14 cm at the age of 13 years.[31] If we accept more than a marginal head tilt, there may be a risk that the healthy muscle will undergo secondary shortening over time giving problems in the future. Vajramani et al followed a child from six months of age to seven years and did not recommend surgery as he adjusted to a mild torticollis with no functional problems.[32]

CONCLUSION

It is important to check that both sternal and clavicular attachment of the SCM muscle are visible. Although the absence of a muscle or parts of a muscle is not expected, rare cases do exist. When torticollis with extreme discrepancy between AROM and PROM in rotation in the cervical spine exists, hypoplasia/aplasia of the SCM muscle must be investigated.

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