STERNOMASTOID TUMOUR AND MUSCULAR TORTICOLLIS

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Torticollis due to contracture of the sternomastoid muscle is a well recognised entity, but there seems little agreement as to etiology, treatment or even terminology. The deformity is largely cosmetic and the results of treatment gratifying. The greatest interest lies in the causation of this condition, with reference in particular to sternomastoid tumour. The pathogenesis remains uncertain but appears unique, there being no known clinical or histological parallel in any other disease process. The objects of this paper are to clarify the relationship of sternomastoid tumour to torticollis, to present some observations on the two conditions and to discuss the results of treatment.

ETIOLOGY AND PATHOGENESIS

Despite several comprehensive reviews, notably by Chandler and Altenberg (1944) and Lidge, Bechtol and Lambert (1957), there is little agreement about the etiology of sternomastoid tumour or muscular torticollis. Chandler and Altenberg regarded the progression from one to the other as so certain that they recommended excision of all tumours at the earliest age. By contrast, Hulbert (1950) and Coventry and Harris (1959) contended that the majority of tumours resolve completely without active treatment. Gray (1935) and Bianco (1958) claimed that most children presenting with torticollis give no history of a sternomastoid tumour, but Coventry and Harris (1959) stated that in half their cases the presence of a tumour was missed by parents and only detected on routine paediatric examination.

It is now well established that a sternomastoid tumour is frequently preceded by a breech, forceps or primiparous birth (Witzel 1883, Fitzsimmons 1933, Chandler and Altenberg 1944, Hulbert 1950, Coventry and Harris 1959). It is tempting to assume that the common factor is trauma. Stromeyer (1838) postulated that rupture of the muscle during parturition produced a tumorous haematoma, and that fibrous replacement caused subsequent torticollis. Of fifteen neonatal or stillbirth necropsies with sternomastoid haematoma, Spencer (1893) found that twelve were by breech or forceps delivery. Sanerkin and Edwards (1966) demonstrated extensive post-mortem muscle fibre damage, haemorrhage and ischaemic change after a breech delivery. Fitzsimmons (1933) reviewed fifty-four cases of Erb's palsy and found an associated sternomastoid tumour in eleven. This he construed as further evidence that the tumour was due to birth injury. Experimentally, Middleton (1930) produced histologically similar tumours by ligation of veins draining the sartorius muscle in dogs, and inferred that venous occlusion during parturition was the cause in babies. However, the striking histological feature of sternomastoid tumour, described as early as 1875 by Taylor, is profuse fibrous replacement of muscle (Figs. 1 and 2) without a trace of haemosiderin, even in specimens excised as early as three weeks after birth (Chandler 1948). Reye (1951) examined four necropsy specimens at ages between four and twenty weeks, and concluded that the appearances suggested almost an overgrowth of tendon at the expense of muscle. Kiesewetter, Nelson, Palladino and Koop (1955), in a study of thirty-two surgical specimens, found very small amounts of haemosiderin in eight only. These lesions were not consistent with massive haematoma, and the tumour much more closely resembled fibrosarcoma, fibromatosis of palmar or plantar fascia, or desmoid tumours. Finally, birth injury cannot have been a factor in the now numerous published instances of sternomastoid tumour following Caesarian section (Schloessmann 1911, Stern 1924, Rossi 1928, Chandler and Altenberg 1944, Kiesewetter et al. 1955).
It is conceivable, as suggested by Coventry and Harris (1959), that intra-uterine torticollis predisposes to a high incidence of breech or forceps deliveries. The subsequent sternomastoid tumour could then be a form of muscle dysplasia, or indeed a response to trauma of already abnormal muscle. Van Roonesen (1670) first postulated abnormal uterine pressure as a cause of torticollis. Sippel (1920) and Chandler and Altenberg (1944) demonstrated a correlation between the breech position and the side of a subsequent tumour. Schmidt (1890) and Chandler (1948) noted asymmetry of the pinna and face together with torticollis immediately after birth and before the development of a tumour. Aside from purely environmental factors, genetic influence as first postulated by Joachimsthal (1905) cannot be dismissed. Von Lackum (1929) quoted a family of three siblings with torticollis, and Stevens (1948) published an unusual case of a right-sided torticollis symmetrically affecting identical twins.

A curious accompaniment of established torticollis is the widespread facial asymmetry or hemiatrophy so succinctly described by Golding-Bird (1890) though he mistakenly ascribed it to incipient poliomyelitis. Middleton (1930) documented extensive bony changes in the skull and facial bones, and it is difficult to account for all the observed changes purely on the basis of relative shortening of the sternomastoid muscle during growth. No really satisfactory explanation of this phenomenon exists.

The developmental and anatomical peculiarity of the muscle has received insufficient emphasis. Straus and Howell (1936) emphasised that the sternomastoid muscle is unique in its double innervation, and developmentally it may be primarily visceral in origin with later somatic components added. Last (1963) pointed out that the muscle really consists of four parts, and Jahns (1936) differentiated between "sternal" torticollis and "clavicular" torticollis, relating the distinction to the results of treatment. Formerly the blood supply of the muscles was believed, by Nové-Josserand and Viannay (1906), to be segmental via end arteries, occlusion of which resulted in segmental infarction. In applying to humans his experimental evidence from dogs, Middleton (1930) assumed that the venous drainage of the muscle followed an identical pattern. However, subsequent injection studies, notably by Chandler and Altenberg (1944), have shown that there is an abundant arterial and venous supply which follows no regular or segmental pattern, and this casts doubt on both these hypotheses.

TERMINOLOGY AND CLASSIFICATION

The term "congenital" torticollis, or wry neck, is widely used but seems inappropriate since the deformity is seldom, if ever, present at birth. Caput obstipum (Holloway 1931), an
old title used in much of the German literature, emphasises the obstetric background but otherwise has little modern significance. The condition has been called “infantile” by Adams (1967) though cases may present at any age through childhood or indeed in adolescence. Anderson (1893) referred simply to “sternomastoid” torticollis, and on the whole the term “muscular” torticollis (Chandler 1948) seems preferable as it underlines the basic pathological process.

Apart from differences in terminology, doubts have been raised as to the uniformity of all cases. Anderson (1893) distinguished between torticollis of a truly congenital nature and that arising solely as a result of birth trauma. Hulbert (1950) described a transient “postural” type, and a “muscular” type presumed due to a previous sternomastoid tumour. Finally, both Hulbert (1950) and Coventry and Harris (1959) showed that most sternomastoid tumours resolve completely and did not progress to muscular torticollis. This has led some to question whether the two are not in fact separate pathological entities.

CLINICAL MATERIAL

Between 1950 and 1965, 152 children with torticollis were seen at the Bristol Hospital for Sick Children, the Bristol Royal Infirmary and the Winford Orthopaedic Hospital. There were three groups of patients. Twenty-six babies presented at or shortly after birth with transient postural torticollis which settled with a minimum of conservative treatment. Fifty-one babies presented within the first weeks of life with a sternomastoid tumour. Seventy-five children presented at various ages with muscular torticollis.

Of the fifty-one babies with a sternomastoid tumour, one was associated with multiple congenital deformities and was left out of the series. Of the remainder, thirty-one were followed up and examined personally, and a further nineteen case notes were available, thus making fifty in all available for study.

Of the seventy-five children presenting with muscular torticollis, twenty-nine were excluded because of inadequate documentation. One was associated with Engelmann’s disease and was therefore left out. Of the remaining forty-five, thirty-four were followed up and examined personally and the case records were available in a further eleven. To this number were added seven patients from the sternomastoid tumour group who subsequently developed muscular torticollis. This made fifty-two cases available for study.

The twenty-six babies with postural torticollis were not considered relevant to this study and were not included. In these cases the etiology was not clear, but in many cases positioning “in utero” was suspected. The early onset at or soon after birth precluded an ophthalmic cause which, according to Duke-Elder (1949), is not seen until eighteen months of age when it is commonly associated with a non-concomitant squint.

PATIENTS PRESENTING WITH A STERNOMASTOID TUMOUR

In fifty babies a sternomastoid tumour (Fig. 3) was palpable between one and fifteen weeks of age, the mean being three weeks. Thirty were in boys and twenty in girls. The birth history is summarised in Table I. Over half the children had breech or forceps birth, and of the remainder over half again were firstborn. One case occurred in a child with an identical twin (unaffected) and in two others a second degree relative was known to have had torticollis.

The tumour occurred on the right side in three-quarters of all cases, and when the birth had been by the breech, the preponderance was even higher (Table II). Many of the babies had associated deformities, mainly minor in nature (Table III). Nearly one-quarter showed transient torticollis which subsided with the tumour. Plagiocephaly was recorded in approximately one-fifth of all cases.
Most of the babies received no treatment except for simple instructions to the mother. A few were given stretching exercises, supervised by a physiotherapy department. No other measures were taken.

Two-thirds of the patients were followed up for between two and sixteen years with an average of six years, but the length of follow-up in one-third was one year or less. The clinical state at follow-up is summarised in Table IV. Seven out of fifty patients subsequently developed muscular torticollis. The remainder had no complaint but on careful examination minor degrees of asymmetry or tightness could be demonstrated in approximately one-quarter (Fig. 4).

**PATIENTS PRESENTING WITH MUSCULAR TORTICOLLIS**

Fifty-two children presented with muscular torticollis (Fig. 5). Twenty-nine were boys and twenty-three girls. Their ages are shown in Figure 6. The children presented at all ages with a fairly even spread through the first decade, after which sporadic cases were seen in adolescence. However, retrospective assessment of the age of onset of the contracture revealed that approximately one-third commenced in the first year of life.

In ten cases there was a history of proven or probable sternomastoid tumour at birth. Moreover, the birth histories of the whole group (Table V) showed a remarkably similar pattern to those of babies with sternomastoid tumour. Over half had been born by breech or forceps delivery and, of the remainder, approximately one-half were firstborn. One child had an identical twin who was not affected. Two were singly affected members of binovular twins. Four had similarly affected first degree relatives and four had similarly affected second degree relatives.
TABLE I  FIFTY BABIES WITH STERNOMASTOID TUMOUR: BIRTH HISTORIES

<table>
<thead>
<tr>
<th>Birth history</th>
<th>Number</th>
</tr>
</thead>
<tbody>
<tr>
<td>Breech</td>
<td>17</td>
</tr>
<tr>
<td>Forceps</td>
<td>12</td>
</tr>
<tr>
<td>Primigravida normal</td>
<td>12</td>
</tr>
<tr>
<td>Multigravida normal</td>
<td>5</td>
</tr>
<tr>
<td>Unknown</td>
<td>4</td>
</tr>
</tbody>
</table>

TABLE II  FIFTY BABIES WITH STERNOMASTOID TUMOUR: SIDE OF LESION

<table>
<thead>
<tr>
<th>Number of babies</th>
</tr>
</thead>
<tbody>
<tr>
<td>Right</td>
</tr>
<tr>
<td>-------</td>
</tr>
<tr>
<td>Whole series . 37</td>
</tr>
<tr>
<td>Breech births . 15</td>
</tr>
</tbody>
</table>

TABLE III  FIFTY BABIES WITH STERNOMASTOID TUMOUR: ASSOCIATED DEFORMITIES

<table>
<thead>
<tr>
<th>Associated deformities</th>
<th>Number of cases</th>
</tr>
</thead>
<tbody>
<tr>
<td>Transient torticollis</td>
<td>12</td>
</tr>
<tr>
<td>Plagiocephaly</td>
<td>9</td>
</tr>
<tr>
<td>Cephalohaematoma</td>
<td>2</td>
</tr>
<tr>
<td>Pyloric stenosis</td>
<td>1</td>
</tr>
<tr>
<td>Congenital dislocation of hip</td>
<td>1</td>
</tr>
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</table>

TABLE IV  FIFTY BABIES WITH STERNOMASTOID TUMOUR: AVERAGE FOLLOW-UP WAS SIX YEARS

<table>
<thead>
<tr>
<th>Outcome following sternomastoid tumour</th>
<th>Number</th>
</tr>
</thead>
<tbody>
<tr>
<td>Normal</td>
<td>22</td>
</tr>
<tr>
<td>Minimal tightness or band</td>
<td>5</td>
</tr>
<tr>
<td>Minimal asymmetry or plagiocephaly</td>
<td>7</td>
</tr>
<tr>
<td>Torticollis</td>
<td>7</td>
</tr>
<tr>
<td>Too recent or unknown</td>
<td>9</td>
</tr>
</tbody>
</table>

TABLE V  FIFTY-TWO CHILDREN WITH MUSCULAR TORTICOLLIS: BIRTH HISTORIES

<table>
<thead>
<tr>
<th>Birth history</th>
<th>Number</th>
</tr>
</thead>
<tbody>
<tr>
<td>Breech</td>
<td>22</td>
</tr>
<tr>
<td>Forceps</td>
<td>9</td>
</tr>
<tr>
<td>Primigravida normal</td>
<td>14</td>
</tr>
<tr>
<td>Multigravida normal</td>
<td>4</td>
</tr>
<tr>
<td>Unknown</td>
<td>3</td>
</tr>
</tbody>
</table>

TABLE VI  FIFTY-TWO CHILDREN WITH MUSCULAR TORTICOLLIS: SIDE OF LESION

<table>
<thead>
<tr>
<th>Number of babies</th>
</tr>
</thead>
<tbody>
<tr>
<td>Right</td>
</tr>
<tr>
<td>-------</td>
</tr>
<tr>
<td>Whole series . 31</td>
</tr>
<tr>
<td>Breech births . 14</td>
</tr>
</tbody>
</table>

TABLE VII  FIFTY-TWO CHILDREN WITH IDIOPATHIC TORTICOLLIS: FACIAL ASYMMETRY

<table>
<thead>
<tr>
<th>Facial asymmetry</th>
<th>Number</th>
</tr>
</thead>
<tbody>
<tr>
<td>Present</td>
<td>36</td>
</tr>
<tr>
<td>Absent or very slight</td>
<td>7</td>
</tr>
<tr>
<td>Unrecorded</td>
<td>9</td>
</tr>
</tbody>
</table>

TABLE VIII  FIFTY-TWO CHILDREN WITH TORTICOLLIS: SITE OF CONTRACTURE

<table>
<thead>
<tr>
<th>Site of contracture</th>
<th>Number</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mainly sternal head</td>
<td>8</td>
</tr>
<tr>
<td>Mainly clavicular head</td>
<td>19</td>
</tr>
<tr>
<td>Both heads</td>
<td>25</td>
</tr>
</tbody>
</table>

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The contracture was more frequent on the right side, and in children born by the breech this preponderance was approximately two to one (Table VI). Facial asymmetry (Fig. 5) was associated with most of the contractures but was by no means invariable (Table VII). The site of the contracture within the muscle was noted in most cases: the clavicular head was usually involved, often principally (Table VIII). In one case this was associated with a bony anomaly within the clavicular attachment.

DISCUSSION

There is a strong similarity between the association of early sternomastoid tumour in children born by the breech, after forceps delivery, and in the firstborn, and the association of children presenting later with torticollis after similar obstetric events. On the other hand, only one baby in seven with a sternomastoid tumour subsequently developed muscular torticollis. In the remainder the tumour resolved, though careful examination in some children later in life revealed intermediate residua, such as a band in the muscle or slight restriction of movement. The relationship is further underlined by the striking right-sided preponderance of both lesions following breech births, an observation previously recorded by Coventry and Harris (1959). It suggests that the cause of both lesions may be related to antenatal position rather than to birth trauma. This is supported by the finding of plagiocephaly in one out of five babies with sternomastoid tumour, an incidence twice that found in the normal population under one year of age by Wynne-Davies (1968). Familial factors, although not preponderant, may play a part. In nine of a combined 100 cases a first or second degree relative was affected. One patient with sternomastoid tumour suffered also from congenital pyloric stenosis. Three such cases were recorded by Chandler and Altenberg (1944).

The relationship between the two conditions is therefore complex. Whatever the change in the muscle it seems likely that it is present at or before birth and has three inconstant and variable sequelae. It may resolve completely; it may clinically become manifest as a sternomastoid tumour: or it may remain clinically latent, subsequently undergoing a variable degree of cicatrisation to produce torticollis.

In established torticollis the contracture is more common in the clavicular head as described by Jahss (1936). In one case this was associated with a bony anomaly in the clavicular
head, like that previously recorded by Middleton (1930). Facial asymmetry was generally, though not invariably, present but it was not possible to correlate its severity with that of the torticollis as suggested by Hough (1934). The relationship is not necessarily a direct one.

TREATMENT OF MUSCULAR TORTICOLLIS

According to Hulbert (1950) subcutaneous tenotomy was practised in Roman times, but the first recorded instance was that by Isaac Minus in 1685. The method has been advocated subsequently by Howell (1929), Elmslie (1943) and Hulbert (1950). William Cheselden (1749) was the first to describe an open operation, a technique subsequently used by Meyerding (1921) and Hough (1934). Jahss (1936) described a variation involving medial transplantation of the clavicular belly. Hellstadius (1927) and Soeur (1940) reported the results of a double approach using a lower subcutaneous tenotomy and an upper open division. Chandler and Altenberg (1944) advised block excision at the earliest stage of frank sternomastoid tumours or cicatrised muscle. The use of a plaster collar to maintain correction after operation was advocated by Meyerding (1921), Howell (1929), Hough (1934) and Soeur (1940).

Of the fifty-two children with muscular torticollis, thirty-six examined at follow-up had been treated by open division of the lower attachment of the muscle (Fig. 7). Two-thirds of these operations were performed by one surgeon and the remaining third by a number of surgeons using the same technique. The age at operation and the length of follow-up are shown in Figure 8.

Operative technique—In all cases a low transverse incision was used dividing skin and platysma. In most instances both heads of the muscle were divided, and in many the underlying deep fascia (Table IX). Closure was performed by suturing the platysma and the skin in two layers, though occasionally skin only was closed. After operation a dressing was applied and immediate physiotherapy begun. In only five cases were stretching exercises continued for longer than four months, and in many the duration was much less.

![Diagram of the age of operation in thirty-six cases of muscular torticollis, and the time of follow-up in years in each case.](image)

RESULTS

All patients were pleased with the correction and none presented for further treatment. However, at follow-up stringent criteria were used to reveal minor cosmetic blemishes (Table X).
Musculo-fascial bands—These were generally only demonstrable at extremes of rotation and lateral flexion of the neck (Fig. 9). They stretched from the occipital attachment of the muscle to the clavicle and were invariably attached well lateral to the normal insertion of the clavicular head. The bands sometimes consisted of tight fascia and skin only, but more frequently they contained actively contractile muscle. They appeared to result from anomalous reattachment of the severed clavicular head as suggested by Jahss (1936). Although present in two-thirds of all cases, they were seldom an important cosmetic defect, though in general they caused some terminal restriction of movement.

Restriction of movement—This occurred only at the extremes of rotation or lateral flexion. It seldom affected more than the last 15 degrees of range. Usually, though not invariably, it was due to a musculo-fascial band.

Absence of the sternomastoid column—Absence of the normal vertical contour of the sternomastoid muscle in the neck resulted in a quite noticeable cosmetic defect (Fig. 10) in just over one-third of cases.

Reverse torticollis—In one case (Figs. 11 and 12) the sternomastoid column was completely absent and a reversal of the torticollis had occurred, presumably due to muscular imbalance.

Disfiguring scar—The neck scar constituted a cosmetic defect to some extent in all cases, particularly in the girls. The scars frequently showed broadening of the medial end (Fig. 10), but in two cases only did this constitute a disfigurement sufficient to warrant further surgical measures.

Residual facial asymmetry—Facial asymmetry of varying degree was recorded before operation in twenty-five of the thirty-six cases. At follow-up residual asymmetry was detected in twelve, that is in approximately half, and the details of these cases are shown in Figure 13. Residual facial asymmetry was generally unnoticed by patient and parent alike (Fig. 7). It was most readily detected from the position of the eyebrow. It is noteworthy that in all but one of the twelve cases with residual asymmetry there were other residua of the torticollis (Fig. 13). Three of the patients were treated in the first year of life yet still retained asymmetry when seen seven, nine and ten years later. It seems likely that the persistence of bands after surgery in the young child may prevent complete restoration of symmetry. Further tenotomy of these bands is therefore probably indicated.

**DISCUSSION**

In the treatment of this largely cosmetic problem many methods are open to the surgeon and almost all claim equally satisfactory results. The major deformity is not difficult to correct...
and attention must be focused on the relatively minor cosmetic defects that precede or follow surgery. The most common criticism of the open operation is the resultant scar. In this series only two patients were considered to have a disfiguring scar. On the other hand, the method was safe and posed no threat to the neurovascular structures in the neck. It could be relied on to correct the principal deformity without the use of subsequent splintage, but anomalous lateral reattachment of the severed clavicular head led to a high incidence of musculo-fascial bands. These were previously noted by Hough (1934), and to prevent this complication...
There was some evidence.

Jahss (1936) described a variation of the technique involving medial transplantation of the clavicular belly. In this series these bands were seldom a serious cosmetic blemish, though in general they caused some terminal restriction of movement. There was some evidence, however, that their persistence was related to the persistence of slight degrees of facial asymmetry. Hough (1934) and Soeur (1940) stated that correction of asymmetry could be expected if surgery was performed under the age of fourteen years. In this series, however, resolution did not always occur, even after treatment in the early years of life. In all but one of these residual cases bands were present.

The most noticeable cosmetic defect was loss of the normal contour of the neck, also previously noted by Hough (1934). In one case division of the muscle had been so complete that reverse torticollis resulted, presumably from muscular imbalance. It was worth noting that in this case the facial asymmetry disappeared completely over a follow-up period of three years (Fig. 12).

**SUMMARY AND CONCLUSIONS**

1. A parallel study has been made of fifty patients presenting with a sternomastoid tumour and fifty-two patients presenting with muscular torticollis.
2. In the birth histories of both these groups there was a high incidence of breech, forceps and primiparous births. The distribution of each was strikingly similar.
3. Sternomastoid tumours were right-sided in three-quarters of all cases and in an even higher proportion of the breech births. There was twice the expected incidence of plagiocephaly. Only one in seven proceeded to muscular torticollis, but in some of the remainder minor residua could be detected.

4. Muscular torticollis presented at any age, but one-third commenced in the first year of life. Only one in five gave a history of previous sternomastoid tumour. The contracture showed a predilection for the clavicular head, and was generally associated with some degree of facial asymmetry.

5. Nine of a combined 102 cases had a first or second degree similarly affected relative.

6. It is concluded that whatever the condition in the muscle at birth, it has three inconstant and variable sequela. The torticollis may resolve completely; it may become clinically manifest as a tumour; or it may remain clinically latent, subsequently undergoing a variable degree of cicatrisation to produce torticollis.

7. The treatment of established torticollis by open division is described and the follow-up in thirty-six cases recorded.

8. This operation can be relied on to cure the principal deformity, but is accompanied by a number of minor cosmetic defects. Of these the most striking are tight bands apparently due to anomalous reattachment of the clavicular head, and loss of the sternomastoid column of the neck.

9. The method could not be relied on to cure facial asymmetry completely, even in the early years of life. However, there was some evidence to suggest that persistence of asymmetry was allied to persistence of other residua of the torticollis, for example fascial bands.

My thanks are due to the surgeons of the Bristol area, and in particular to Mr A. L. Eyre-Brook under whose care most of the cases were, and to Mr A. H. C. Ratliff for much criticism and encouragement.

REFERENCES


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