Orthopaedic sequelae in neurologically recovered obstetrical brachial plexus injury: Case study and literature review

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Abstract

Purpose. Description of shoulder sequelae in obstetrical brachial plexus injury (OBPI) patients who had spontaneous functional recovery, in the context of historical and current conservative methods of treatment.

Method. Case study of a baby with serious complications, followed by a review of the literature from 1900 until 2001 about conservative treatment of OBPI with respect to the prevention of shoulder complications.

Results. Description of contractures and bony deformities did not show important discrepancies over time, other than more detailed images because of new technical possibilities. There is no agreement on the explanation of the development of these deformities. Secondary changes caused by muscular imbalance and longstanding contracture are recognised by all authors. A primary osteoarticular lesion was recognised as a possible cause in the beginning of the twentieth century, then forgotten for a long time and only in the 1980s had gained interest again.

The main change in treatment concerns the use of arm braces. This was strongly recommended in the first half of the twentieth century, then advised against and is at this moment not anymore mentioned.

Conclusions. There is no consensus on the cause of contractures and bony deformities in children with OBPI. Conservative methods of treatment have changed over the years, without research on the outcome of these treatment changes.
Introduction

Obstetrical brachial plexus injury (OBPI) is caused by traction on the brachial plexus during delivery. The incidence of OBPI as reported in the literature varies from 0.9 - 4.6 per 1000 live births\textsuperscript{11;14;28;33;34;39}. The natural recovery rate is usually reported as 80-95\text%\textsuperscript{12;14;18;33}. However, a recent study at the Academic Medical Center in Amsterdam showed a recovery rate of only 73\text%\textsuperscript{16}. One of the complications of OBPI is contracture of the joints, especially of the shoulder. A muscular imbalance may be followed by structural changes in the tissues surrounding a joint, causing a contracture. It has been emphasised that longstanding contractures can cause bony deformities, and that prevention of contractures should be an important treatment goal\textsuperscript{3;8-10;13;15;21;22;24;35;37;41}. Prevention of contractures is currently thought to be best managed by frequent and gentle exercises, which put all joints through a full range of motion. Moreover, in order to prevent the shoulder from dropping by gravity, all children should be seen by a physiotherapist to instruct the parents how to take care of the affected arm. After discharge of the hospital, physiotherapy should be continued with the aim of checking the range of motion and stimulating muscle function.

One of our patients, a baby girl, developed contractures and bony deformity of the shoulder, despite intensive physiotherapy and good compliance of the parents, and despite good neurological recovery. This discrepancy between the good neurological and the poor functional outcome triggered us to perform a literature review. This review was focussed on two points of interest:

1. The different types of shoulder complications in OBPI; and
2. The different forms of conservative treatment in OBPI, with particular reference to the prevention of shoulder sequelae

Case report

A full term baby was born by vacuum extraction after a normal pregnancy. Her birth-weight was 3200 grams. Directly after birth, no active motion in the left upper limb was seen apart from slight finger flexion. An obstetrical brachial plexus injury was diagnosed by physical examination. No further complications were present at birth. Physiotherapy with full range of motion exercises began one week after birth and the parents were taught how to place the baby’s arm in a well supported position in order to protect the shoulder joint. At this stage a full range of motion of the shoulder was present. Two months later her shoulder musculature, elbow flexors and supinator muscle (innervated from C5 and C6) were still completely paralysed.
but she did show improved function of the C7 musculature and had normal hand function. Again there was a full passive range of motion of all joints of the upper limb (Table 1). Physiotherapy was continued once a week to stimulate motor function and to prevent contractures. At the age of 4 months she could perform active motions against gravity in all directions with her arm, although not all muscle groups yet showed normal strength. Sensory assessment revealed no abnormalities. However, at that time a limited passive range of motion of the shoulder joint was noticed. External rotation, internal rotation and horizontal adduction were all 10 - 20° limited compared to the contralateral side.

Physiotherapy was intensified to restore full passive range of motion. The exercises were carried out daily by the parents and once a week by the physiotherapist. One month later (five months after birth), neurological recovery was complete, all muscle groups showed normal strength, but the passive

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**Table 1.** Passive range of motion (pROM) of the left shoulder in a girl with left-sided OBPI, as documented in the case records at varying ages

<table>
<thead>
<tr>
<th>pROM</th>
<th>Age 2 mth</th>
<th>4 mth</th>
<th>5 mth</th>
<th>9 mth</th>
<th>1.5 yr</th>
<th>2.5 yr</th>
<th>3.5 yr</th>
<th>4.5 yr</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total abduction</td>
<td>180°</td>
<td>180°</td>
<td>180°</td>
<td>170°</td>
<td>170°</td>
<td>170°</td>
<td>180°</td>
<td></td>
</tr>
<tr>
<td>Glenohum. abd.</td>
<td>90°</td>
<td>90°</td>
<td>90°</td>
<td>80°</td>
<td>80°</td>
<td>80°</td>
<td>90°</td>
<td>80°</td>
</tr>
<tr>
<td>External rotation</td>
<td>80°</td>
<td>60°</td>
<td>70°</td>
<td>80°</td>
<td>90°</td>
<td>80°</td>
<td>80°</td>
<td>70°</td>
</tr>
<tr>
<td>Internal rotation</td>
<td>60°</td>
<td>50°</td>
<td>50°</td>
<td>45°</td>
<td>50°</td>
<td>40°</td>
<td>35°</td>
<td>0°</td>
</tr>
<tr>
<td>Horiz. adduction*</td>
<td>normal</td>
<td>↓</td>
<td>↓</td>
<td>↓</td>
<td>↓</td>
<td>↓</td>
<td>↓</td>
<td>↓</td>
</tr>
</tbody>
</table>

* Horizontal adduction was not documented in grades but only in “normal”, “slightly diminished” (↓), “diminished” (↓↓), or “strongly diminished” (↓↓↓)

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Figure 1.
Girl at the age of 5 months with a left-sided OPBL. Notice the asymmetrical shoulder contour
horizontal adduction had worsened. The shoulder contour was asymmetrical in appearance (Figure 1). Roentgenogram of the shoulder showed a disappearance of the lower edge of the glenoid promontory with an inferior subluxation (Figure 2). CT scanning revealed no other abnormalities. The conservative treatment with physiotherapy was continued with gentle full range of motion exercises.

By the age of 4.5 years, all muscle groups still showed normal strength and she could use both arms symmetrically in grasping and playing. However, the contours remained asymmetrical.
and the passive range of motion was seriously restricted, especially in internal rotation and horizontal adduction (Table 1). At that time the glenoid promontory had disappeared (Figures 3 and 4). MRI showed that the deficit was replaced by cartilage.

**Literature review**

**Method**

A MEDLINE search between January 1966 and September 2001 and a Cochrane Library search were performed, and of all publications the reference lists were checked backwards up to 1900 for relevant articles and handbooks. The following keywords were used: obstetrical palsy, brachial palsy and brachial plexus were combined with shoulder joint as well as with conservative treatment. Articles and handbooks were included if they contained a clear description of conservative treatment or if there was special reference to complications of the shoulder joint. Exclusion criteria were articles focussed on operative treatment which only mentioned between the lines that postoperative physiotherapy had been performed. Finally, case studies describing less than five patients were excluded.
Results

No studies were found concerning our subject in the Cochrane Library. In our initial search through the other sources a total of 80 abstracts was found that seemed to meet our inclusion criteria. After reviewing the full text, it was learned that 32 articles focussed on operative treatment, in which conservative treatment was just mentioned by one line, for example “all children were sent for physiotherapy”. Twelve articles appeared to be case studies describing less than five patients, and in eight articles there was no clear description of conservative treatment or shoulder complications, although the title and abstract suggested this. These were not included in our review. Therefore, only 28 articles met our criteria for inclusion for the review (Table 2). During review only two papers had good methodological descriptions of all treatment modalities and of the neurological deficit of the patients. In order to compile all data of the 28 papers, only a descriptive literature review could be performed.

This historic review on conservative treatment for OBPI, shows important changes over the last 100 years. Until 1962 conservative treatment was a combination of massage, exercises and the use of braces (J.J. Thomas being a notable exception)\(^3,5,8,17,23,25-27,30,31,37,38\). The arguments for using a brace differ, from relaxation of paralysed muscles\(^8,23,37,38\) to prevention of contractures\(^3,6,17,25\), to protection of damaged nerves\(^3\) or a combination of these factors\(^5,26,30\). Directly after birth arm braces were used continuously for several months, and then used during the night for an additional 3 - 12 months. Frequently, a removable brace was used in which the arm was held in a right-angle at the shoulder and elbow, with the forearm supinated and sometimes with the hand and fingers included in extension. This is known as the “Statue of Liberty” brace\(^3,8,25-27,38\). Taylor preferred the use of a loop of gauze around the wrist on the damaged side, which pulled up the arm until the hand was near the occiput. Then the ends of the gauze were fastened on the opposite side\(^30\). In 1916 Sever was insistent on the use of braces, but in 1925 he discussed the complications during bracing, such as swelling around the wrist and elbow causing pain during treatment. As other drawbacks of bracing the restricted use of the arm and a probable slower convalescence of the OBPI were reported\(^27\). This warning against the use of braces had no effect on other authors. However, Wickstrom did recognise the risk for an abduction contracture and modified his braces. He changed the positioning of the arm in the brace in complete external rotation, 45° forward flexion and 70° abduction\(^37\). Only in 1967 Adler rejected the use of braces, as did Aston and Hardy in 1979 and 1981\(^1,2,14\). They noticed that their use could lead to external rotation and abduction contractures, especially in children who had no additional physiotherapy during the bracing period. Gilbert and Zancolli also warned that these contractures might happen, but they did
### Table 2. Literature review of conservative treatment and shoulder complications in OPBI

<table>
<thead>
<tr>
<th>First author</th>
<th>Year of publication</th>
<th>Design, number of cases</th>
<th>Contractures *</th>
<th>Bony deformities †</th>
<th>Conservative treatment ‡</th>
</tr>
</thead>
<tbody>
<tr>
<td>Clark⁵</td>
<td>1905</td>
<td>Clinical lecture</td>
<td>-</td>
<td>-</td>
<td>Massage, exercises, always brace</td>
</tr>
<tr>
<td>Fairbank⁸</td>
<td>1913</td>
<td>Clinical lecture</td>
<td>External rotation</td>
<td>Post. subluxation</td>
<td>Massage, exercises, always brace</td>
</tr>
<tr>
<td>Thomas¹¹</td>
<td>1914</td>
<td>Clinical lecture</td>
<td>-</td>
<td>-</td>
<td>Massage, exercises, always brace</td>
</tr>
<tr>
<td>Thomas¹²</td>
<td>1914</td>
<td>Clinical lecture</td>
<td>-</td>
<td>Post. subluxation</td>
<td>-</td>
</tr>
<tr>
<td>Platt¹³</td>
<td>1915</td>
<td>Clinical lecture</td>
<td>External rotation</td>
<td>Post. subluxation</td>
<td>Massage, exercises, always brace</td>
</tr>
<tr>
<td>Sever²⁶</td>
<td>1916</td>
<td>Report of 470 cases</td>
<td>External rotation</td>
<td>Post. subluxation, acromial hooking, elev./rotation scapula</td>
<td>Massage, exercises, sometimes brace</td>
</tr>
<tr>
<td>Taylor²⁰</td>
<td>1920</td>
<td>Clinical lecture</td>
<td>-</td>
<td>Acromial hooking</td>
<td>3 weeks rest, after that exercises, always brace</td>
</tr>
<tr>
<td>Sever²⁷</td>
<td>1925</td>
<td>Report of 1100 cases</td>
<td>External rotation</td>
<td>Post. subluxation, acromial hooking, flattening glenoid, elev./rotation scapula</td>
<td>Massage, exercises, sometimes brace</td>
</tr>
<tr>
<td>Jepson¹⁷</td>
<td>1930</td>
<td>Clinical lecture</td>
<td>External rotation</td>
<td>Ant./post. subluxation, curved clavicle, elevation scapula</td>
<td>Massage, exercises, always brace</td>
</tr>
<tr>
<td>Boorstein¹</td>
<td>1930</td>
<td>Report of 211 cases</td>
<td>External rotation, abduction</td>
<td>-</td>
<td>Rest for 5 days, after that massage, exercises, always brace</td>
</tr>
<tr>
<td>Scaglietti²⁵</td>
<td>1938</td>
<td>Report of 199 cases</td>
<td>External rotation, abduction, horizontal adduction</td>
<td>Flattening glenoid</td>
<td>Massage, exercises, warmth, always brace</td>
</tr>
<tr>
<td>Wickstrom³⁸</td>
<td>1955</td>
<td>Report of 54 cases</td>
<td>External rotation, abduction</td>
<td>Post. subluxation, acromial hooking, flattening humeral head, elev./rotation scapula</td>
<td>Exercises, always brace</td>
</tr>
<tr>
<td>Wickstrom³⁷</td>
<td>1962</td>
<td>Report of 87 cases</td>
<td>External rotation, abduction, adduction</td>
<td>Post. subluxation, acromial hooking, flattening humeral head, elev./rotation scapula</td>
<td>Exercises, always brace</td>
</tr>
<tr>
<td>Mumenthaler²⁹</td>
<td>1965-1998 Handbook</td>
<td>External rotation, abduction</td>
<td>Post. subluxation, flattening humeral head</td>
<td></td>
<td>Exercises, always brace</td>
</tr>
<tr>
<td>First author</td>
<td>Year of publication</td>
<td>Design, number of cases</td>
<td>Contractures *</td>
<td>Bony deformities †</td>
<td>Conservative treatment ‡</td>
</tr>
<tr>
<td>--------------</td>
<td>---------------------</td>
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<td>----------------</td>
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<td>-------------------------</td>
</tr>
<tr>
<td>Adler¹</td>
<td>1967</td>
<td>Report of 88 cases</td>
<td>External rotation, adduction</td>
<td>-</td>
<td>Rest for 5 days, after that exercises, no brace</td>
</tr>
<tr>
<td>Aston²</td>
<td>1979</td>
<td>Report of 80 cases</td>
<td>External rotation, abduction</td>
<td>Post. subluxation, flattening humeral head, retroversion glenoid</td>
<td>Rest for 2 weeks, after that exercises, no brace</td>
</tr>
<tr>
<td>Hardy¹⁴</td>
<td>1981</td>
<td>Report of 36 cases</td>
<td>Int./ext. rotation, abduction</td>
<td>-</td>
<td>Rest for 1 week, after that exercises, no brace</td>
</tr>
<tr>
<td>De Palma⁶</td>
<td>1983</td>
<td>Handbook</td>
<td>External rotation, abduction</td>
<td>Post. subluxation, acromial hooking, flattening glenoid</td>
<td>Rest for 1 week, after that exercises, always brace</td>
</tr>
<tr>
<td>Brown⁴</td>
<td>1984</td>
<td>Review</td>
<td>External rotation, abduction</td>
<td>Post. subluxation, acromial hooking, elev./rotation scapula</td>
<td>Review of above mentioned literature</td>
</tr>
<tr>
<td>Narakas²⁰</td>
<td>1987</td>
<td>Handbook</td>
<td>External rotation, abduction</td>
<td>-</td>
<td>Rest for 3 weeks, after that exercises</td>
</tr>
<tr>
<td>Gilbert⁹</td>
<td>1988</td>
<td>Clinical lecture</td>
<td>Int./ext. rotation, abduction</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Sjöberg⁹⁸</td>
<td>1988</td>
<td>Cohort of 48 cases</td>
<td>External rotation, abduction, flexion</td>
<td>-</td>
<td>Exercises</td>
</tr>
<tr>
<td>Zancolli⁶²</td>
<td>1993</td>
<td>Report of 368 cases</td>
<td>Int./ext. rotation, abduction, adduction</td>
<td>Post./ant. subluxation, humeral epiphyseolysis</td>
<td>-</td>
</tr>
<tr>
<td>Hoekema¹⁵</td>
<td>1998</td>
<td>Handbook</td>
<td>External rotation, abduction</td>
<td>-</td>
<td>Support shoulder when paralytic, after that exercises</td>
</tr>
<tr>
<td>Pearl¹²</td>
<td>1998</td>
<td>Report of 25 cases</td>
<td>External rotation</td>
<td>Flattening post. aspect glenoid, biconcave glenoid, pseudoglenoid</td>
<td>-</td>
</tr>
<tr>
<td>Waters¹⁶</td>
<td>1998</td>
<td>Report of 94 cases</td>
<td>Int./ext. rotation, abduction</td>
<td>Post. subluxation, retroversion glenoid</td>
<td>-</td>
</tr>
<tr>
<td>Hoekema¹⁶</td>
<td>2000</td>
<td>Cohort of 62 cases</td>
<td>Int./ext. rotation, abd., horiz. add.</td>
<td>-</td>
<td>Exercises</td>
</tr>
<tr>
<td>Sluijs vd²⁹</td>
<td>2001</td>
<td>Report of 16 cases</td>
<td>-</td>
<td>Convex/biconcave glenoid, post. subluxation, retroversion glenoid</td>
<td>-</td>
</tr>
</tbody>
</table>

* If contractures are mentioned, the direction of limitation is named. If not: only “-”; † Bony deformities: the type of bony deformities that are mentioned; if the existence of bony deformities was not mentioned: only “-”; ‡ Description of the main elements of conservative treatment; if conservative treatment is not described: only “-”
not take a clear position into whether to use or reject braces\cite{9,40}. Since then, braces are not mentioned anymore in articles. Only in two handbooks they were still advocated as a part of conservative treatment, De Palma in 1983 and Mumenthaler in the first edition in 1965, this was unchanged up to the seventh edition in 1998\cite{6,19}.

As for the moment to start physiotherapy, some authors advocated a period of rest from 5 days to 3 weeks because of the acute traumatic neuritis after OBPI\cite{1-3,6,20,30}. Until 1930 massage, of the gentle kneading form, was advocated, to stimulate growth. All authors recommend exercises. This is divided into two parts: (1) passive movements of all the joints in order to prevent contractures to develop; and (2) active exercises, to induce the child to use the arm voluntarily, which vary with the age of the child. J.J. Thomas made a clear prescription of these exercises in five categories of age until the age of three years\cite{31}.

The risk for shoulder contractures is mentioned by most authors\cite{1,2,4,6,8,9,14-17,19,20,22,23,25,28,37,38,40}. Limitation of passive external rotation and abduction is most frequently mentioned. The incidence of contracture within a non selected cohort who was followed since birth has only recently been described\cite{16}. In this study shoulder contracture occurred in at least one third of the children with delayed neurological recovery and in at least two third of the children with incomplete neurological recovery.

Bony deformities, seen on plain radiography, were already frequently mentioned in the first half of the twentieth century\cite{8,17,23,25,27,30,32}, and this was continued later on\cite{2,4,6,19,37,38,40,42}. There were no differences in their findings: posterior subluxation of the humeral head, acromial hooking, flattening of the glenoid fossa, flattening of the humeral head and elevation and rotation of the scapula. But they did not agree on the explanation of the development of these deformities. Throughout the last century many authors described the deformities as secondary to muscular imbalance and longstanding contracture\cite{2,4,17,30,37,38}. The possibility of a primary osteoarticular lesion, whether or not together with a plexus lesion, was recognised in the beginning of the twentieth century\cite{8,23,25}, then forgotten for a long period of time. It only regained interest in the 1980s\cite{6,40,42}. T.T. Thomas describes a direct injury to the shoulder joint as the primary cause of an obstetrical palsy, the plexus and its branches becoming involved in the adjacent axillary inflammation\cite{32}. Concerning bony deformity secondary to nerve injury, Dysart performed an experimental study in the rat about growth of the humerus after denervation\cite{7}. It appears that the form and growth of the humerus is influenced by muscle activity, especially the proximal portion of the humerus. Nothing is mentioned about the glenoid.

Studies concerning magnetic resonance imaging, computer tomography and arthrography of the shoulder reveal flattening of the posterior aspect of the glenoid, the formation of a
convex or biconcave glenoid or a pseudoglenoid, and posterior subluxation of the humeral head\textsuperscript{22,29,36}. Here, muscular imbalance and longstanding contracture were described as the only etiologic factors. Disuse atrophy of the humeral head and scapula was described in all articles that concerned bony deformities of the shoulder. The isolated disappearance of the glenoid promontory, as seen in this case, has never been described as such. Neither has the occurrence of shoulder deformity in neurologically recovered cases.

**Discussion**

Regarding our patient it can be said that the baby was treated according to the current standards. Despite this, but also despite continuing neurological recovery, she developed a serious contracture and bony deformity of the shoulder. Although many authors have emphasised that longstanding contractures can cause bony deformity, there is no evidence that this deformity is indeed a consequence of the contracture. Pearl found that not all glenoids deform in the presence of an internal rotation contracture\textsuperscript{22}. Therefore, other factors must also contribute to the abnormal development of the glenoid. Pearl hypothesised that a persistent, posteriorly directed force on the growing glenoid may either erode the posterior aspect of the glenoid or inhibit its development\textsuperscript{22}. This is in line with Waters et al.\textsuperscript{36}, who stated that the abnormality of the glenoid as seen on magnetic resonance images suggests an impairment of growth of the posterior aspect of the glenoid. When extrapolated to this case, there should have been a persistent caudally directed force on the glenoid promontory in the absence of an internal rotation contracture. This could be possible, since the parents were very well trained to prevent this contracture developing. Thereby they were preventing the tightening of anterior soft tissues, that could apply a posteriorly directed force. Therefore, it is possible that the combination of completely paralysed shoulder muscles for more then two months together with a normal passive range of motion is responsible for a constant caudally directed force on the promontory in this child by the latissimus dorsi muscle. In this way the glenoid promontory could be impaired to grow normally. The discrepancy of bony malformation of the glenoid and remodelling of the cartilage, as seen on the MRI, to fit the humeral head, should theoretically give the shoulder the same movement as in normally developed shoulders. It can be hypothesised however, that the malformation of the promontory, be it posteriorly or inferiorly, has a different origin than the muscle contracture moving the shoulder in subluxation. The only experimental study about growth after denervation of the shoulder musculature did not mention anything about the glenoid\textsuperscript{7}. 
On the other hand, an obstetrical trauma of the shoulder joint can not be rejected as a possible cause for the deformity either, as was described extensively by Zancolli. At birth the shaft of the humerus is nearly wholly ossified, but the two articular extremities of the humerus and the scapula are cartilaginous. Thus the junction side of the ossified humerus and the cartilaginous humeral head are prone for a (epi)physeal fracture. Furthermore, the scapula at birth is largely osseous too, with the exception of the glenoid fossa, the coracoid and the acromial process, and the posterior border and inferior angle, which are still cartilaginous. Due to the cartilaginous aspect of the shoulder components, it is very difficult to recognise a direct shoulder trauma just after birth.

With respect to these theories and the presence of shoulder deformities despite adequate physiotherapy, in neurologically recovered OBPI patients, one could reconsider the use of arm braces during the period of flaccid palsy of the shoulder muscles, as described in the first half of the 20th century. Although this seems to be an appropriate change in treatment, the authors feel that this could only be justified after a randomised clinical trial.

Conclusion

This case shows that contracture and bony deformity of the shoulder joint can occur as a complication of OBPI, even in the presence of good neurological recovery and conservative treatment according to the current standards. The literature review shows that there is no consensus on the cause of contractures and bony deformities in children with OBPI. Conservative methods of treatment have changed over the years, but without research on the outcome of these changes it will be difficult to find the right one. This might be due to the fact that large studies on non-selected patients are scarce and anatomic changes in the presence of neurological recovery, such as in this case, are not taken into consideration.
Reference List


Shoulder complications in OBPI: case study and literature review


