Chapter 7

General discussion and recommendations for treatment and research on OBPI

The studies described in the previous chapters focus on three aspects of obstetrical brachial plexus injury (OBPI): incidence, neurological recovery and shoulder complications. These aspects will be discussed separately and also in terms of their interdependent association. As a result of this, recommendations will be made for treatment and future research on OBPI.
General discussion

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      3.3.1  Unavoidable changes with no functional consequences
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Chapter 7

1 Incidence of OBPI

Our first study focused on the incidence and risk factors of OBPI in a cohort of all children born alive from 1988 through 1996 in the Obstetric Department of the Academic Medical Center (AMC) in Amsterdam (Chapter 2). This was possible because the AMC has had a combined obstetrical and neonatological database since 1988. In a total of 13,366 children, 62 were born with an OBPI, an incidence of 0.46%. When looking at the data per year from 1988 until 2002, the incidence was fluctuating, but no tendency to increase or decrease could be detected during this period. The obstetrical risk factors in our study were identical to those described in other publications: maternal age ≥ 35 years, multiparity, ethnic descent, diabetes, second stage of delivery >60 minutes, breech presentation, operative vaginal delivery in multiparae, and birth weight. Birth weight appeared to be the most important variable in predicting OBPI. Unfortunately, antepartum estimation of fetal weight either by clinical examination or by ultrasound assessment is unreliable, especially in macrosomia. We could calculate a predictive model by multivariate logistic regression analysis, but the predictive value of this model was too low to be effective in clinical management, as was also concluded by other authors. Several investigators specifically stated that a policy of elective caesarean delivery in cases of suspected fetal macrosomia will have no significant effect on the prevalence of brachial plexus injury. Rouse et al. studied the potential effectiveness of a policy of elective caesarean delivery for fetal macrosomia diagnosed by ultrasound; to prevent one case of permanent brachial plexus injury, 3,695 caesarean sections in cases with an estimated birth weight exceeding 4,500 g would be needed. Furthermore, it is recognised that OBPI can also occur during caesarean section. There has been much discussion concerning a policy of planned caesarean section in breech presentation, especially since the publication of the “Term Breech Trial”. The results of this trial were in favour for planned caesarean section in the case of term breech presentation. This has not completely been confirmed by other studies, e.g. two recent Dutch studies. In the Netherlands this has lead to revised guidelines being issued by the Dutch Society of Obstetrics and Gynaecology, concerning breech presentation. This guidelines still include planned vaginal delivery, after counselling. In our study, analysis was also performed to identify possible risk factors in the sub-group of children with a non-recovered OBPI, because they might have suffered a more severe birth trauma. However, we could not detect any difference in risk factors between children with and without complete neurological recovery. This is in line with the findings of Ouzounian et al., who also studied a sub-group of children with a non-recovered OBPI. Therefore, even with regard to the non-
recovered OBPI cases, a predictive model appeared to be of limited value.

The incidence was found to be higher than usually reported. When reading articles and textbooks, it is noticeable that many authors refer to each other, with no reference to one or more of the original studies on incidence. Twelve other studies investigating the incidence of OBPI in western and in non-western countries could be identified, that were distinct in their methods of collecting data without selection of the study population. It is difficult to compare data from studies in the various countries, because of differences in obstetric population characteristics and practices. Therefore, Table 1 presents the data arranged according to geographical regions. In all western studies an increase in incidence is observed from the 1970s until the late 1990s. This may partly be explained by a trend of increasing birth weight. Bager et al. presented an ongoing investigation into the national incidence of OBPI from 1980 until 1994 in Sweden. There was a steady and significant increase from 0.14% in 1980 to 0.23% in 1994, but no explanation for this increase could be given. The findings of Sjöberg (Sweden) and Walle (Finland) are in line with this development. Our study demonstrated a remarkable increase in comparison to an earlier study in the same

**Table 1. Studies on the incidence of OBPI (%), arranged in different geographical regions**

<table>
<thead>
<tr>
<th>Region</th>
<th>Study</th>
<th>Incidence</th>
<th>Year</th>
<th>'70 – '80</th>
<th>'80 – ’90</th>
<th>&gt; ’90</th>
</tr>
</thead>
<tbody>
<tr>
<td>Scandinavia</td>
<td>Bager</td>
<td>0.14</td>
<td>'80 – '94</td>
<td></td>
<td></td>
<td>0.23</td>
</tr>
<tr>
<td></td>
<td>Sjöberg</td>
<td>0.19</td>
<td>'73 – '82</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Walle</td>
<td>0.20</td>
<td>'81 – '83</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>U.S.A.</td>
<td>Gordon</td>
<td>0.19</td>
<td>&lt; '73</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Greenwald</td>
<td>0.20</td>
<td>'72 – '82</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Jackson</td>
<td>0.25</td>
<td>'83 – '86</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>The Netherlands</td>
<td>Treffers</td>
<td>0.13</td>
<td>'59 – '73</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Wolf</td>
<td></td>
<td>'88 – '96</td>
<td></td>
<td></td>
<td>0.46</td>
</tr>
<tr>
<td>Middle East countries</td>
<td>Soni</td>
<td>0.36</td>
<td>'83</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Benjamin</td>
<td>0.45</td>
<td>'86 – '87</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Peleg</td>
<td>0.44</td>
<td>'92 – '95</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Dawodu</td>
<td>0.29</td>
<td>'93 – '95</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Malaysia</td>
<td>Boo</td>
<td>0.16</td>
<td>'86 – '87</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* In 4 studies, a differentiation into ethnicity has been made, with significant variations of incidence in the various ethnical groups (see Tables 2a and 2b)
hospital, from 0.13% in the period 1959-1973 to 0.46% between 1988 and 1996\textsuperscript{88,95}. In 1984 the hospital moved to another region of Amsterdam that is inhabited by a large number of immigrants, mainly from Surinam, West Africa and the Indian sub-continent. This ethничal variance in the population is quite different from that in the former location. Non-Caucasian women showed a much higher risk for OBPI after correction for birth weight. The incidence in the Caucasian group in our study was found to be 0.22%, which still means an increase in comparison with the findings of Treffers et al.\textsuperscript{88}, and is comparable with the findings of the Scandinavian authors\textsuperscript{5,78,89}. The three American studies also report findings that are in line with an increase over a decade\textsuperscript{36,37,48}. In New York, a remarkable and significant race difference

### Tables 2a and 2b. Characteristics of the differentiation in ethnicity in 4 studies on the incidence of OBPI

<table>
<thead>
<tr>
<th>Gordon\textsuperscript{16}</th>
<th>Brachial Plexus group</th>
<th>Control group</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>White</td>
<td>14</td>
<td>45</td>
<td></td>
</tr>
<tr>
<td>Negro</td>
<td>40</td>
<td>47</td>
<td>0.01</td>
</tr>
<tr>
<td>Other</td>
<td>5</td>
<td>8</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Dawodu\textsuperscript{25}</th>
<th>Brachial Plexus group</th>
<th>Control group</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>UAE</td>
<td>1</td>
<td>19.0</td>
<td>n.s.</td>
</tr>
<tr>
<td>Asians</td>
<td>6</td>
<td>40.5</td>
<td>- (reference group)</td>
</tr>
<tr>
<td>Other Arabs</td>
<td>197</td>
<td>30.7</td>
<td>0.003</td>
</tr>
<tr>
<td>Others</td>
<td>1</td>
<td>9.8</td>
<td>n.s.</td>
</tr>
</tbody>
</table>

### Table 2a. Ethnicity in the brachial plexus group and the control group

<table>
<thead>
<tr>
<th>Boo\textsuperscript{12}</th>
<th>Live births</th>
<th>OBPI cases</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N</td>
<td>N (perc.)</td>
</tr>
<tr>
<td>Malay</td>
<td>14870</td>
<td>27 (0.18)</td>
</tr>
<tr>
<td>Chinese</td>
<td>6095</td>
<td>13 (0.21)</td>
</tr>
<tr>
<td>Indian</td>
<td>3877</td>
<td>2 (0.05)</td>
</tr>
<tr>
<td>Others</td>
<td>1334</td>
<td>0 (0.0)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Wolf\textsuperscript{95}</th>
<th>Live births</th>
<th>OBPI cases</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N</td>
<td>N (perc.)</td>
</tr>
<tr>
<td>Caucasian</td>
<td>4985</td>
<td>11 (0.22)</td>
</tr>
<tr>
<td>African</td>
<td>2489</td>
<td>26 (1.04)</td>
</tr>
<tr>
<td>Indian</td>
<td>867</td>
<td>7 (0.80)</td>
</tr>
<tr>
<td>Others</td>
<td>1571</td>
<td>12 (0.76)</td>
</tr>
</tbody>
</table>

### Table 2b. Incidence of OBPI in the various ethnic groups
was found, with a much higher proportion of OBPI in the children of Negro gravidas than in the total group (Table 2a)\textsuperscript{36}. The data from the Middle East present yet another picture. The studies in Saudi Arabia, Israel and Libya show particularly high incidences, especially when compared to western studies in the same period of time (Table 1)\textsuperscript{9,65,82}. A study in the United Arab Emirates shows percentages that are more comparable with those found in western countries in the same period of time, but they show remarkable ethnical differences (Table 2a)\textsuperscript{25}. This is also the case with the Malaysian study (Table 2b)\textsuperscript{12}. In short, it is clear that the incidence of OBPI can differ between ethnical groups, although it was not possible to detect any explanation for this. However, the increasing migration of different races all over the world might be responsible for an increase in incidence in the western countries.

To summarise, although risk factors can be identified, the preventive value is too low for effective prevention of OBPI. Therefore, we should realise that this obstetric complication will continue to be an important problem.

2 Neurological recovery of OBPI

OBPI has been extensively described for more than a century (Chapter 6). At the beginning of the twentieth century the publications mainly consisted of clinical lectures or reports on large numbers of patients, with extensive descriptions of conservative treatment methods. In recent decades, however, surgery has become an important treatment option. This concerns intraplexal reconstructive as well as functional surgery, i.e. tendon and bone corrections. Since these techniques have become popular in the past decade, the literature mainly focuses on these surgical techniques. However, this tendency gives rise to much confusion; when a child does not need operative treatment, this is easily translated as “good recovery”. And, as was already mentioned in the discussion concerning the incidence, many authors refer to each other, with no reference to original outcome studies. Therefore, many care-givers still expect that good recovery can be achieved in 80 - 90% of cases. In our opinion, in general, these descriptions of the incidence as well as neurological recovery from OBPI were too optimistic. Therefore, our research on these aspects consists of only cohort studies, in order to obtain insight into the magnitude of the problem.

Our first study, described in Chapters 2 and 3, mainly focused on incidence and the obstetrical risk factors related to non-recovered OBPI. We did, however, also investigate the clinical course by using case notes from the outpatient clinical records. In this way it was possible to find out whether a child had achieved an early complete neurological recovery (within 3
weeks after birth), a delayed complete neurological recovery (after > 3 weeks) or incomplete neurological recovery. The findings suggested a less favourable neurological recovery than the reports that were usually referred to. This stimulated us to complete the historical cohort study that is described in Chapter 4. This cohort consisted of all OBPI children born in the AMC between 1991 and 1998.

A drawback of this study, however, is the duration of the follow-up period, that ranged from 1 to 7 years. A final physical examination of all children at the same age would have been preferable. However, analysis of the different age-groups separately did not change the findings on neurological outcome. Furthermore, because of the differences in age, it was not possible to obtain accurate insight with regard to possible functional limitations as a consequence of OBPI. According to our data, functional outcome was quite good. However, it is likely that the functional problems will increase with age, because of the growing demands for independence in the activities of daily living. Therefore, all children should be re-examined in fixed age-groups during a follow-up over a much longer period of time.

2.1 Neurological findings

Our findings confirmed the results of our first study: with distinct definitions of “neurological recovery” and “persistent paresis”, only 66% of the cases showed complete neurological recovery (Chapter 4). In half of these children there was a delay of 6.5 ± 4.2 months, during which time shoulder complications could occur, as described in Chapters 5 and 6. Although in the current literature these children are easily forgotten, it concerns a large group of children who require extensive treatment in order to prevent irreversible complications.

One aspect that was not discussed in the former chapters, is the fact that full muscle strength together with normal sensibility, does not automatically mean normal muscle performance. This has been the focus of two recent publications17,27. Brown et al. speak of apraxia in children with an OBPI, as the clumsiness of the arm that can be the result of an inability to recruit the motor units that are available. They suggest a failure of the motor areas in the brain or spinal cord to construct programmes for the affected arm, and that this, in turn, is due to the presence of paralysis at a critical time in infant development17. Van Dijk et al. refer to disruption of normal maturation of the central nerve system (CNS) motor programmes, leading to motor unit potentials (MUPs) that are not embedded in functional movement programmes27. This could be an explanation for the findings of Gjörup et al. and Strömbeck et al., who observed limited hand function in children with upper plexus injury33,84. It could also explain the occurrence of late complications of the shoulder in children with a
complete return of muscle strength in all muscle groups, as is described in Chapter 5.

Another phenomenon that can negatively influence motor performance after full muscle strength is regained, is the occurrence of involuntary postparalytic antagonistic movements. This can be explained by misled axons after spontaneous or postoperative nerve regeneration. It can occasionally cause severe cocontractions that hamper coordinated movements\textsuperscript{72}.

In conclusion, it is likely, in the first place, that our definitions of recovery provide a more realistic view of the magnitude of the problem of persistent paresis in OBPI, and secondly, that our conclusions on this subject might even be too optimistic, when focused on normal arm and hand function. Furthermore, when the phenomena that might cause abnormal motor performance are taken into account, this could mean that we have to reconsider our treatment programmes, for instance by including botulinum toxin therapy or electrical stimulation.

2.2 Follow-up assessment

There is a great disparity between two categories of outcome measures which are used in OBPI studies. Firstly, the assessments that combine various movements of the arm in order to create a simple quantitative item for determining the effect of surgical treatment\textsuperscript{7,32,53}, and secondly, a more detailed assessment per joint and per muscle, as Adler already advocated in 1967\textsuperscript{1}. In recent decades it is recognised that, in addition to the overall categories such as the Mallet score, motor function of all different movements should be described for a precise prognostication of the outcome as well as for the prediction of the need for surgical intervention\textsuperscript{3,10,11,22,24}. The sheets in our medical records only partly meet this requirement (Figure 1). Nonetheless, the data could be used, because in all children the various active movements were documented not only in grades of strength, but also in grades of range of motion. However, for further clinical and research purposes, the lower part of the sheet will be adapted (Figure 2). We prefer to keep the upper part of the sheet unaltered, because this is very helpful in providing the parents with insight into the progress made in the neurological recovery of their child (Figures 3a and 3b).

2.3 Predicting neurological outcome

In Chapter 4 we have described three major problems in studying the prediction of neurological outcome: firstly, the various definitions of recovery; secondly, the confusion between “recovery” and “need for surgical intervention”; and thirdly, the study populations that are easily biased by selection. Unfortunately, after performing thorough research into the incidence of OBPI,
Chapter 7

Figure 1. This sheet was used for the periodic documentation of the neurological and orthopaedic condition of the children. The squares for muscles are shaded according to the observed muscle function, using the Narakas (or Gilbert) motor scale (Table 1 in chapter 4).
Name: _____________________________   OBPI: _____________________________   right/left
Date of birth: _____________________________   Horner’s sign: _____________________________   yes/no
Date physical examination: _____________________________   Diaphragm function: _____________________________   asymmetrical/normal

<table>
<thead>
<tr>
<th>Muscle / Movement</th>
<th>Right</th>
<th>Left</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Passive / Active</td>
<td>Passive / Active</td>
</tr>
<tr>
<td>Shoulder:</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total abduction</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Glenohumeral abduction</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Lateral rotation in add.</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Lateral rotation in 90° abd.</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Medial rotation</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Horizontal adduction</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Elbow:</td>
<td>Flexion / extension</td>
<td></td>
</tr>
<tr>
<td>Forearm:</td>
<td>Pro / supination</td>
<td></td>
</tr>
<tr>
<td>Wrist:</td>
<td>Flexion / extension</td>
<td></td>
</tr>
<tr>
<td>Hand:</td>
<td>Finger flexion</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Finger extension</td>
<td></td>
</tr>
</tbody>
</table>

**Figure 2.** This sheet is recommended for the documentation of the neurological and orthopaedic condition of children with OBPI. The squares for muscles are shaded according to the observed muscle function, using the Narakas (or Gilbert) motor scale (Table 1 in chapter 4).
Figure 3a at 6 weeks of age:

<table>
<thead>
<tr>
<th>m.serratus anterior</th>
<th>m.deltoides</th>
<th>m.biceps</th>
<th>mm. pronatores</th>
<th>mm.flexores digitorum</th>
<th>thenar</th>
</tr>
</thead>
<tbody>
<tr>
<td>m.bracialis</td>
<td>m.brachialis</td>
<td>m.fl.carpi rad.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>m.supraspinatus</td>
<td>m.brachioradialis</td>
<td>m.triceps</td>
<td>m.flexor poll.l.</td>
<td>hypotenar</td>
<td></td>
</tr>
<tr>
<td>m.infraspinatus</td>
<td>m.supinator</td>
<td>mm.extens.dig.</td>
<td>mm.ext.pollis</td>
<td>mm.interossei et lumbricales</td>
<td></td>
</tr>
<tr>
<td>m.teres major</td>
<td>m.latissimus dorsi</td>
<td>m.flexor carpi ulnaris</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

m.pectoralis major

| M0 | M1 | M2 | M3 |

Figure 3b at 3 months of age:

<table>
<thead>
<tr>
<th>m.serratus anterior</th>
<th>m.deltoides</th>
<th>m.biceps</th>
<th>mm. pronatores</th>
<th>mm.flexores digitorum</th>
<th>thenar</th>
</tr>
</thead>
<tbody>
<tr>
<td>m.bracialis</td>
<td>m.brachialis</td>
<td>m.fl.carpi rad.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>m.supraspinatus</td>
<td>m.brachioradialis</td>
<td>m.triceps</td>
<td>m.flexor poll.l.</td>
<td>hypotenar</td>
<td></td>
</tr>
<tr>
<td>m.infraspinatus</td>
<td>m.supinator</td>
<td>mm.extens.dig.</td>
<td>mm.ext.pollis</td>
<td>mm.interossei et lumbricales</td>
<td></td>
</tr>
<tr>
<td>m.teres major</td>
<td>m.latissimus dorsi</td>
<td>m.flexor carpi ulnaris</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

m.pectoralis major

| M0 | M1 | M2 | M3 |

Figure 3 a / b. The upper part of the sheet in the medical records of one child at two different follow-up moments. This will provide the parents a clear insight into the progress of the neurological recovery of their child.
some authors have continued to describe the outcome of their cohort in terms such as “almost good strength”, “moderate to severe residual disability”, or “satisfactory spontaneous recovery”\textsuperscript{2,5,25,36,82}. Other authors describe the outcome in a selected population only in terms of whether or not there is a need for surgical treatment\textsuperscript{56,57,85,90}.

We have tried to overcome these problems by performing a cohort study to investigate the incidence of OBPI, together with a follow-up measurement of the strength of all different muscle groups at fixed time-intervals. Furthermore, we looked for prognostic signs with regard to complete neurological recovery, defined as normal strength in all muscle groups together with normal sensibility. Finally, we sub-divided the group of children with a non-recovered OBPI into those with and without neurosurgical intervention.

Our findings concerning the neurological prognosis of OBPI are as follows:

1° We agree with Clark, who, already one century ago, stated that “the degree and extent of the lesion cannot be determined immediately after its occurrence”\textsuperscript{21}.

2° Biceps function at three months of age can be considered to be the best indicator for neurosurgical intervention.

3° Active external rotation and supination at the age of three months are superior in predicting eventual neurological recovery.

\textbf{Figure 4.} A boy, 6 years old, with a left-sided OBPI. According to the Narakas classification, his condition would be described as “good recovery”\textsuperscript{57}. 
We emphasise that the parents should be informed about the prognosis of the neurological recovery of their child, and about whether or not it needs neurosurgical intervention, as two separate subjects. This can help them to accept a situation in which their child may not achieve complete neurological recovery, even though its condition is too good to benefit from surgical treatment. Just describing this future condition as “good”, can eventually lead to great disappointment for the parents. Figures 4 and 5a/b are examples of such situations.

Figures 5a/b. A boy, 9 years old, with a left-sided OBPI. He has accomplished almost complete active elevation (Figure 5a) and elbow flexion, and, therefore, his condition would be classified as “good recovery” according to the Narakas classification\(^ {57} \). However, there is an absence of any active external rotation, and this prevents him from reaching his left shoulder (Figure 5b) or bringing his hand to the mouth.

3 Shoulder complications

Shoulder problems in OBPI were already described in the literature at the beginning of the last century (Chapter 6). This included contractures as well as bony deformities, seen on plain radiography. Since 1988 computer tomography (CT) has been utilised for this purpose, and since 1995 also magnetic resonance imaging (MRI). To our knowledge, our study was the first to determine the prevalence of shoulder complications in an unbiased, complete cohort of children with an OBPI.
3.1 Pathogenesis

There is difference of opinion in the literature about whether or not contractures and osseous deformity are solely due to persistent muscular imbalance, or that they can also be a consequence of direct birth trauma of bones, joints and muscles. This latter opinion was first advocated at the beginning of the twentieth century, renewed proposed by Scaglietti in 1938, and more recently many authors have expressed it again since the 1980s. Zancolli extensively described various osteoarticular and muscular injuries as primary obstetrical lesions, in addition to any secondary lesions as a consequence of OBPI. Other authors consider shoulder deformities to be only a result of muscular imbalance caused by OBPI.

We have found a significant association of clavicular fracture with the development of osseous deformity of the glenoid at a later stage, but not with shoulder contracture. This underscores the opinion that not all shoulder sequelae can be explained as a consequence of longstanding muscular imbalance. Furthermore, our findings of contractures and osseous deformity in children with an early complete neurological recovery seem to support this theory.

Therefore, early assessment of possible associated lesions should be considered in all newborns with an OBPI. Since the cartilaginous component of the epiphysis in babies is radiographically invisible, as are any muscular lesions, ultrasound could be a good alternative as an initial method of assessment. It has been used and found to be reliable in studies concerning epiphyseolysis in newborns, fractures of humerus and clavicles in children, and shoulder congruity in children with a chronic OBPI. Ultrasound is non-invasive, carries no known radiation risk, and there is no need for general anaesthesia, as is required for CT and MRI. It usually causes no distress to the children, who can be held by their parents. Therefore, early ultrasound assessment of the shoulder could be performed in all new-borns with an OBPI. In this way, the theory of osteoarticular and muscular injuries as possible primary obstetrical lesions can be evaluated and, if found true, treatment or preventive measures must be reconsidered in order to prevent subsequent complications.

3.2 Contractures

In our cohort, the frequency of shoulder contractures of >10°, >20°, and >30° was 56%, 31%, and 17%, respectively (Chapter 5). The various opinions concerning shoulder contractures throughout one century are summarised in Table 2 of Chapter 6. All authors since 1913 describe limitations of external rotation and, since 1930, also of abduction. In the 1960s, Wickstrom and Adler observed diminished adduction, as a consequence of too rigorous use of the ‘Statue of Liberty’ brace. Since the 1980s Zancolli also described decreased adduction,
as a consequence of a direct joint lesion during birth\textsuperscript{96,97}, and decreased passive internal rotation has also been reported\textsuperscript{32,42,45,83,91,96}. Finally, a kind of diminished range of motion of the shoulder has been extensively described by Birch as a posterior glenohumeral contracture\textsuperscript{10}. The clinical feature is winging of the scapula in anteflexion of the arm, and it is caused by capsular tightness (Figures 6 and 7a-d). This symptom has also been described by Scaglietti in 1938\textsuperscript{77}. We refer to this phenomenon as a reduction of horizontal adduction. In our study, horizontal adduction was the most frequently affected direction (Chapter 5). Unlike Birch, we felt unsure about the reliability of estimation of the degree of horizontal adduction, and classified this only as “reduced” or “normal”, compared to that of the contralateral side. This was later supported by research carried out by De Winter et al., who found that examination of the degree of restriction (i.e. the difference between the affected side and the contralateral side) has acceptable reproducibility for all directions in the shoulder joint, except for horizontal adduction\textsuperscript{94}. We agree with Birch that this is an important phenomenon. In our experience, this is the first symptom that occurs in the development of a shoulder contracture. The treatment consists of daily exercises, in which the parents place the child’s afflicted hand on its opposite shoulder, with the axis of the humerus parallel to the ground. In this position the winging scapula has to be firmly depressed onto the chest wall (Figures 7a/b)\textsuperscript{10}. Although this may prevent further deterioration, in our opinion return to normal range of motion is not possible. A mild restriction of horizontal adduction will not reduce hand and arm function extensively, but it can be responsible for an abnormal appearance of the shoulder girdle that can bother both the child and the parents (Figure 6). It can also be a warning sign for the development of more severe contractures that can, indeed, have serious consequences. Therefore, we strongly advise that passive horizontal adduction should be an obligatory part of every physical examination of a child with an OBPI.

\textbf{Figure 6.} A girl, 7 months old, with a left-sided OBPI. She has a reduced horizontal adduction causing winging of the scapula during active flexion of the arm.
Figures 7a/b. A girl, 6 years old, with a right-sided OBPI. She has a reduced horizontal adduction. This can be assessed by placing the afflicted arm on the opposite shoulder, with the axis of the humerus parallel to the ground. In this position the angle between the scapula and humerus can be compared to that of the opposite side.

Figures 7c/d. A caudally directed view of the angle of the horizontal adduction of the same girl. The horizontal adduction is strongly reduced at the right side.
Contractures can occur rapidly, also in cases with transient paresis. Therefore, physiotherapy should start in the first week after birth, by instructing the parents how to handle the baby. After one week, daily exercises, moving all joints through a full range of motion, are recommended. In addition to physiotherapy, regularly consultation of a specialist in rehabilitation medicine is also advisable. In this way a differentiation is made between weekly treatment sessions given by the physiotherapist, and once every six to eight weeks documentation of the active and passive ranges of motion and functional assessment by the specialist in rehabilitation medicine. Especially during the period of initial neurological recovery, all attention is usually directed towards this improvement, but at the same time the first contractures can develop. It is our experience that a differentiation between treatment and assessment will facilitate the early identification of this complication.

When a steady state has been achieved in active and passive range of motion, physiotherapy can be stopped. However, during the final examination of our cohort of children with an OBPI, we noticed that some children had developed a more severe contracture after several years. Therefore, regular check-ups with documentation of active and passive range of motion in all directions are needed for a long period of time after physiotherapy has ceased.

In addition to physiotherapy, the use of braces might be reconsidered as a measure to prevent the development of early internal rotation contracture. As described in Chapter 6, this method of treatment has been abandoned, but no research has been carried out to evaluate the consequences of this change of approach. Complications resulting from the use of a brace, i.e. an abduction contracture, can easily be prevented by combining this with daily range of motion exercises. However, this should only be reconsidered as a part of a randomised clinical trial, in order to study the effect of this change in treatment.

### 3.3 Osseous deformities

We have tried to differentiate between those changes that are a direct consequence of denervation on the maturation and growth of the glenohumeral joint, and have no negative influence on arm function, and the potentially avoidable changes that could cause functional limitations.

#### 3.3.1 Unavoidable changes with no functional consequences

In Chapter 5 we have described hypoplasia of the humeral head and scapula, and elevation of the scapula, as unavoidable changes after denervation. Furthermore, we also observed delayed ossification of the humeral head and glenohumeral subluxation. Altogether, such changes in the glenohumeral joint were found in 13 out of 56 children (23%). In the literature
we could find only two studies on the effect of denervation on growth, but both focused on
the humerus and not on the glenoid. Dysart et al. performed an experimental study to
investigate the growth of the humerus after denervation in rats, and proved that muscle
activity contributed to the shape and growth in length of a long bone\(^2\). McDaid et al. studied
upper extremity limb-length discrepancy in OBPI\(^5\). In a group of patients with persistent
paresis, he found that children with an upper brachial plexus lesion had significantly less
forearm and total limb shortening than those with a global brachial plexus lesion.

3.3.2 Potentially avoidable changes that might cause functional limitations

The radiographic findings were classified as abnormal if a non-spherical humeral head, a
short abnormally formed clavicle, or an abnormal glenoid fossa was evident on an oblique
anteroposterior radiograph of the shoulder. Glenoid malformation was defined as hypoplasia
of the inferior aspect of the glenoid promontory. Radiographically, this is represented by a
line between the superior and inferior aspect of the glenoid rim, which is normally perpendicular
to the scapular spine. Based on this definition, the prevalence in our cohort was 33% (16 out
of 48 patients with complete radiographic follow-up). In these cases, an abnormal glenoid
fossa was observed in 80%, and a non-spherical humeral head in 32%. No abnormalities of
the clavicles were found, not even in the children with an initial clavicular fracture.

It is difficult to determine the clinical relevance of these findings. As discussed in Chapter 5,
a single oblique anteroposterior radiograph is not sufficient to determine the glenoscapular
angle and the degree of congruity of the glenohumeral joint. These symptoms have been
described by several authors, and are especially important with regard to the implications for
surgical treatment of the shoulder joint\(^4;10;39;43;46;63;68;79;86;91;97\). However, it was not our intention
to describe these deformities in detail, but merely to give an impression of the magnitude of
the problem of any osseous shoulder deformation in children with an OBPI.

Hypoplasia of the glenoid promontory could be a direct effect of denervation on the glenoid
development. To our knowledge, no studies have been published on this subject. In an
additional study, we have compared plain radiography with CT and MRI in 9 children with an
OBPI (mean age 8 years, range 4 to 13), all with severe limitations of the passive range of
motion of the shoulder. Unfortunately, this study could not be finished because of the sudden
death of our radiologist, and the study ended too early for us to draw definite conclusions.
However, one of the findings was that in three of the four children showing a hypoplastic
glenoid promontory as the only deformity on plain radiography, extra cartilage formation as
a filling of the space was seen on MRI. This was also found in the case we described in
Chapter 6 (see Figures 2 - 4 of Chapter 6). These findings are comparable with those in congenital glenoid dysplasia (CGD), described by Currarino et al. They suggested that the increased space in the inferior part of the glenohumeral joint seen in CGD on radiographs, was produced by unossified cartilage. However, they report that a well-developed inferior glenoid promontory is normally present in children of all ages, including newborn and premature infants. Furthermore, in some of the children in our study hypoplasia of the promontory only developed at a later stage. Therefore, this phenomenon cannot be solely explained as delayed ossification caused by denervation. However, regardless of the aetiology, it is still a frequent occurrence in our cohort, and it can be a cause of subsequent problems and symptoms of the shoulder. This has been described by Smith et al., who reviewed 12 patients with CGD who has variable problems such as pain, often with some associated limitation of movement, and symptoms of instability. The description of these late problems are similar to those reported by several OBPI patients who complain of shoulder pain after the age of 20 to 30 years. Therefore, although we do not understand the aetiology of the frequently found hypoplasia of the glenoid promontory, it is a phenomenon that has to be studied carefully in order to understand and prevent further shoulder problems.

As already stated, our data only concern the prevalence of any osseous abnormality, seen on plain radiography in an unselected cohort of children with an OBPI. We believe that it is important to realise how often these abnormalities can occur. Further studies on aetiology and clinical relevance are needed and these should be prospective cohort studies that start to observe babies immediately after birth.

4 Recommendations for treatment and future research
As a consequence of our findings and the above mentioned considerations, some recommendations can be made regarding (1) the diagnosis and treatment of children with an OBPI, (2) further research into these aspects, and (3) the organisation of the care for children with an OBPI in the Netherlands.

4.1 Recommendations regarding the diagnosis and treatment of children with an OBPI
1) For a description of the neurological recovery of OBPI, a dichotomy into complete or incomplete neurological recovery is inadequate. In children with a delayed complete neurological recovery, many problems can occur: e.g. contractures, osseous deformities,
abnormal motor performance. These children are imperceptible in the above mentioned dichotomy, and therefore it is insufficient for the indication of treatment. This problem can be overcome by a sub-division into three groups. In the first place the children with early complete recovery and those with delayed or incomplete recovery are divided, because these last groups are in need for longstanding treatment, preventive measures and follow-up assessments. At the end of the treatment period, a further division can be made into complete and incomplete neurological recovery, as an important outcome measure. In this way a division into three groups can be made: early complete recovery (neurological recovery within 3 weeks), delayed complete recovery, and incomplete neurological recovery. Although this final sub-division can only be made in retrospect, the distinction between

<table>
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<tr>
<th>Neurological recovery</th>
<th>Treatment goals</th>
<th>Possible treatment measures</th>
</tr>
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<tr>
<td>Early complete</td>
<td>Optimal daily care of the baby</td>
<td>Informing the parents</td>
</tr>
<tr>
<td></td>
<td>Stimulating muscle function</td>
<td>Conservative treatment, Exercises</td>
</tr>
<tr>
<td>Delayed complete</td>
<td>Prevention and treatment of contractures</td>
<td>Conservative treatment, Exercises, braces</td>
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<tr>
<td></td>
<td>Stimulating muscle function</td>
<td>Operative treatment, Release, tendon transfer</td>
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<tr>
<td></td>
<td>Prevention and treatment of contractures</td>
<td>Conservative treatment, Exercises, braces</td>
</tr>
<tr>
<td>Incomplete</td>
<td>Prevention and treatment of functional problems</td>
<td>Operative treatment, Release, tendon transfer</td>
</tr>
<tr>
<td></td>
<td>Prevention and treatment of contractures</td>
<td>Conservative treatment, Exercises, braces, adaptations</td>
</tr>
<tr>
<td></td>
<td>Prevention and treatment of functional problems</td>
<td>Operative treatment, Tendon transfer, derotation osteotomy, arthrodesis</td>
</tr>
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**Figure 8.** Treatment goals and possible treatment measures in the 3 sub-groups of OBPI
children without and with need for intensive treatment can now be made in the proper time. The treatment goals and possible treatment measures in the 3 sub-groups of OBPI are shown in Figure 8.

2) Regular assessments of a child with an OBPI during the first year of its life should focus on comparing each joint and each muscle group of the affected arm with those on the contralateral side. Concerning passive range of motion, special attention should be paid to horizontal adduction (see section 3.2). The findings can be recorded on a sheet, as shown in Figure 2. With the help of these sheets, the parents can easily be informed about (1) the progress of the neurological recovery, (2) the prognosis of the neurological outcome, (3) the indication for neurosurgical treatment, and (4) the development of any contracture and the necessity of appropriate treatment. Furthermore, the sheets can be used as a database for future research into these aspects.

3) At the age of three months, biceps function is considered to be the best indicator for neurosurgical treatment, whereas active external rotation and supination at the same age are better in predicting eventual neurological recovery. At a later stage, functional limitations can be an indication for tendon and bone corrections. The parents should be informed about the prognosis of the neurological recovery of their child, and about whether or not it needs neurosurgical intervention, as two separate subjects. The overall information on conservative and surgical treatment can provide a more realistic insight and expectations for the parents and could prevent them from disappointment at a later date.

4) Physiotherapy should start immediately after birth, by informing the parents how to handle their child. After the first week, full range of motion exercises should be performed daily by the parents. The physiotherapist should teach the parents how to perform these exercises and how to stimulate muscular strength and motor development. Regular check-ups, recording the active and passive range of motion of all movements should be performed by another care-giver who, in the Netherlands, would preferably be a specialist in rehabilitation medicine. There should be direct contact between these two care-givers in the case of any change in the situation, in order to adjust the therapy. When a steady state has been achieved, physiotherapy can be stopped, even if there is residual paresis or contracture. In that case, check-ups by the specialist in rehabilitation medicine have to be continued once or twice a year until the child reaches the age of 7 to 8 years. If any problems occur at a later stage, therapy can be considered for a limited period of time.

5) Early ultrasound assessment of possible associated lesions is advised in all newborns with an OBPI (section 3.1). This can indicate whether a direct birth trauma of the shoulder joint
can be related to future shoulder complications. Our study did not provide definite conclusions about the significance of plain radiography in children with an OBPI. With regard to surgery of the shoulder joint, most authors consider MRI or CT to be satisfactory imaging techniques. In our experience, a three-dimensional reconstruction of a spiral-CT provides useful additional information (see Figure 4 in Chapter 6).

4.2 Recommendations regarding future research on children with an OBPI

6) Studies investigating neurological recovery and complications of OBPI should be performed as cohort studies with unselected populations. The children in our cohort need to be followed for a much longer period of time, especially for the determination of functional limitations. Because of growing demands with regard to occupation, housekeeping and the care for offspring with increasing age, the follow-up period should preferably be until adulthood.

7) Braces might be reconsidered as a measure to prevent the early development of internal rotation contracture (see section 3.2). For the positioning of the arm in this brace, attention should be paid to the advice of experienced authors, especially Wickstrom in 1962. Figures 9a-c show an example of such a brace. The use of braces should be imbedded in a prospective study to evaluate the possible positive and negative consequences.

8) As other possible future treatment modalities, two therapies can be taken into consideration:

- Botulinum toxin therapy could be an effective treatment for involuntary postparalytic antagonistic movements, as Rollnik reported in 2000.
- Electrical stimulation could have a positive influence on the development and maturation of motor programmes in the CNS, as is suggested by some authors.

Both therapeutic measures might have a supplementary value in the treatment of children with an OBPI, but their application should only be considered after thorough investigation in a properly designed study to evaluate the consequences of changes of treatment.

4.3 Recommendations regarding the organisation of the care for children with an OBPI in the Netherlands

9) In the Netherlands many children with an OBPI are treated, or have been treated, by a physiotherapist in a free-standing practice, and some by a physiotherapist or occupational therapist in a rehabilitation centre or an outpatient rehabilitation clinic of a hospital, under the medical supervision of a specialist in rehabilitation medicine. Some of these children are seen by one of the three specialised brachial plexus teams (BP-teams). In
these BP-teams, children are observed by a neurosurgeon, an orthopaedic surgeon, a plastic surgeon, a specialist in rehabilitation medicine, and sometimes also a physiotherapist. However, there is no complete overview of the organisation of the care for children with an OBPI. This can lead to a situation in which some children do not receive the appropriate therapy because the care-givers lack the necessary experience. On the other hand, a child can be seen by many care-givers who all focus on the same problems and treatment, with the risk of contradictory advice.

Optimal care in the first years of a child’s life is time-consuming. Apart from the intensive assessments and facilitation of the active and passive range of motion, this usually consists
of frequent contacts between the care-givers and the parents. These are focused on their understanding of the injury and mentally coping with the events of the traumatised delivery, and at a later stage will also involve the care for any functional impairments\(^{44}\). It is not possible that the three BP-teams can provide all this care. (When our data on the incidence of OBPI could be generalised for the whole country, this would indicate that in the Netherlands each year at least 900 children would be born with an OBPI).

In the Netherlands the care for children with an OBPI should be organised in the same way as the care for children with a spina bifida, i.e. a specialised multidisciplinary team, in addition to a local non-surgical treatment team. For children with an OBPI this local treatment team will consist of a specialist in rehabilitation medicine, together with a physiotherapist, an occupational therapist and, if necessary, a social worker. This local treatment team is responsible for the regular assessments and the non-operative treatment, and the BP-team focuses primarily on operative treatment. Furthermore, the BP-team can advise the local treatment team whenever changes in therapy are indicated. In such an organisation, all children with an OBPI will be seen by a physiotherapist. If complete neurological recovery is achieved within 3 weeks, physiotherapy can be stopped and no further check-ups are necessary. In all other cases, the child should be seen by a specialist in rehabilitation medicine, starting at 6 weeks of age. From that time onwards, regular assessments of active and passive range of motion should be performed and documented. If serious paresis of one or more muscles is still present at the age of 3 months, the child should be observed in a BP-team, in order to evaluate the indication for surgical treatment. The non-operative treatment will remain in the hands of the local treatment team. Physiotherapy should be continued until a steady state of active and passive range of motion has been achieved, together with an adequate overall motor development. In a non-operated child with a residual paresis, this is usually at the age of 3 to 4 years, and in an operated child it will be 1 or 2 years later. When physiotherapy is stopped, regular check-ups by the specialist in rehabilitation medicine should still be continued, with careful documentation of active and passive range of motion. Check-ups in the BP-teams are necessary in order to obtain proper feedback on possible neurosurgical treatment and, later on, to focus on functional surgical treatment, if necessary.

The organisation of the care for children with an OBPI according to this model would ensure that all children received maximal treatment without an overkill of check-ups and no confusing contradictory advice from the various care-givers involved.
Reference List


General discussion


