Chapter 5

The natural history of OBPL

A systematic review

Willem Pondaag
Martijn J.A. Malessy
J. Gert van Dijk
Ralph T.W.M. Thomeer

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Abstract The natural course of obstetric brachial plexus lesions (OBPLs) is generally considered to be very good, with complete or almost complete recovery stated to occur in over 90% of patients. The validity of this presumption is important if one is to ensure that patients are treated appropriately, that parents of OBPL infants receive accurate information on prognosis and that a standard is set for epidemiological comparison of different treatment modalities.

In order to obtain a scientifically-based outcome figure we performed a systematic literature review of the natural history of OBPL using the MEDLINE database. The following pre-defined criteria were used to assess the quality of the study: prospective design, population-based series, 90% follow-up for at least three years, and an accurate assessment of end stage without surgery.

A total of 1020 papers were screened using title, keywords and abstract, resulting in 76 papers for evaluation. Thirty-four articles failed to meet any inclusion criterion. Thirty-five papers met one inclusion criterion, seven met two inclusion criteria. None of the reports met more than two points inclusion criteria. The studies that most closely matched our inclusion criteria reported an incomplete recovery in 20 to 30% of patients.

From this systematic literature review, it can be concluded that the natural history of OBPL is, in fact, not known. The rate of spontaneous recovery in OBPL may be worse than is generally believed.

Obstetric Brachial Plexus Lesion (OBPL) is caused by traction to the brachial plexus during labour.[1,2] In the majority of cases delivery of the upper shoulder is blocked by the mother's pubic symphysis (shoulder dystocia). If additional traction is applied to the child's head, the angle between the neck and the shoulder is forcefully widened, overstretching the ipsilateral brachial plexus. The resulting traction injury may vary from neurapraxia or axonotmesis to neurotmesis and avulsion of rootlets from the spinal cord. Recently, the exact origin of OBPL was again a matter of debate.[3] It was suggested that intrauterine maladaptation, not nerve traction, causes the plexopathy. However, medicolegal aspects may play a role this discussion.

The incidence of OBPL varies from 1.6 to 2.9 per 1000 births in prospective studies.[4,5] The upper brachial plexus is most commonly affected, resulting in paresis of the shoulder and biceps muscles, as first described by Erb and Duchenne.[6] Hand function is additionally impaired in approximately 15 % of patients,[4,7,8] isolated injury to the lower plexus (Déjèrine-Klumpke's type) is rare.[9]

The extent of neural damage can only be assessed evaluating of recovery in the course of time, because nerve lesions of different severity initially present with the same clinical features. Neurapraxia and axonotmesis eventually result in complete recovery. Neurotmesis and root-avulsion, on the other hand, result in permanent loss of arm function, which may lead in time to the development of skeletal malformations, cosmetic deformities, behavioural problems and socio-economic limitations.[10-14]

At present most authors advise surgical exploration of the brachial plexus if spontaneous recovery is considered insufficient at a preset age.[15-17] Absence of biceps function at 3 months of age is regarded as the key indicator for surgical exploration by some authors.[15,18] Others use a combined score of different movements to decide whether nerve surgery should be performed at 9 months.[16] Ancillary testing, in particular electromyography, is not considered reliable enough for prognostication.[19,20]

Methods of repair include nerve grafting after resection of the neuroma and nerve transfer in the case of root-avulsion.[15,21-25] Results achieved by these surgical approaches are claimed to be superior to the outcome in conservatively treated patients with equally severe lesions.[15,26-28] However, this comparison relies on historical controls;[29] no randomized study has been performed.[6,30]

In this context, the true percentage of infants who do not recover from OBPL becomes important as these children might benefit from reconstructive surgery. Reliable data on the frequency and severity of functional deficits in the natural course of OBPL are a prerequisite for developing adequate treatment strategies. Furthermore, such data would provide the parents of newborn infants with OBPL with realistic information on prognosis.

The prognosis of OPBL is generally considered to be very good, with complete or almost complete spontaneous recovery in over 90% of patients.[25,31-35] However, this view is based on a limited number of studies[36,37], which are cited indiscriminately without considering the methodology used.

In the present review we performed a systematic literature search[38] to clarify the natural course of untreated OBPL. Ideally, a study on the natural course of OBPL should be a prospective analysis of a demographic population with sufficient follow-up and a clear end-stage assessment. We applied four pre-defined criteria to assess the methodological quality of the available studies: study-design, population, duration of follow-up, and assessment of endstage.[39]

Method

Search strategy

The Medline database 1966-2001[40] was searched using keywords as well as text words (Table 1). Additionally, a similar search was performed in the Science Citation Index (Expanded)[41] from 1988 onward. All articles were initially screened by title, abstract and keywords. If any reference was made to incidence, natural history or outcome of OBPL, the article was selected for further reading. Reviews were also selected for reading, as well as articles whose content was not sufficiently clear from title, abstract or keywords. No restriction for language was applied: articles in languages other than English, French, German or Dutch were translated with the help of medically-trained speakers.

Reference lists of all selected articles were studied, and articles published in medical periodicals not identified in our initial Medline query were added if they had appeared after 1965. Studies that only presented results of surgically treated patients (either nerve reconstructive surgery or tendon transfers) were excluded from analysis.

Table 1: **Medline search strategy**

	Query	Results
1	"Paralysis, Obstetric"[MESH]	290
2	((Plexus[TITLE] AND Brachial[TITLE]) OR "Brachial Plexus"[MESH:NOEXP] OR "Brachial Plexus Neuropathies"[MESH:NOEXP]) AND ("Birth Iinjuries"[MESH] OR "Child"[MESH:NOEXP] OR "Infant"[MESH:NOEXP] OR "Infant, Newborn"[MESH:NOEXP])	830
3	(Erb[TEXT] OR Erbs[TEXT]) AND (Palsy[TEXT] OR Palsies[TEXT] OR Paralysis[TEXT])	135
4	#1 OR #2 OR #3	1022
5	#4, Limits: Publication Date to 2002-01-01	1020

Inclusion criteria

Four pre-defined inclusion criteria applied to judge the articles were (Table 2): 1) The study design should be prospective to avoid the risk of an unknown inclusion bias. 2) The study population should be constituted on a demographic basis. Results based on referrals to specialized centres or on hospital records may be biased by selection towards more severe cases. 3) Follow-up should be sufficiently long and complete. Three years was considered as the minimum for obtaining a reliable assessment of a functional end stage in OBPL for the following reasons. Firstly, the end stage of recovery might take up to three years, and secondly, the neurological investigation of younger children is difficult to quantify. Thirdly, functional disabilities in OBPL may only become apparent when the complexity of daily tasks increases with age. The percentage of patients that might be lost to follow-up was arbitrarily set at 10%. 4) Assessment of the endstage of recovery should be accurate and reproducible, preferably using a specialized pre-defined assessment protocol. If any of the patients in the series had been surgically treated, thereby influencing the outcome of the end-stage, this criterion was not met. As the decision to operate is usually based on an early assessment of severity, and not on the end stage, such studies cannot be used for an analysis of the natural course.

The validity of these criteria will be debated in more detail in the Discussion. Each of these four items was allocated one point; the points were added to produce a sum score. Articles receiving three or four points were to be read by a second reviewer.

Results

Using the aforementioned Medline search strategy 1020 articles were addressed. Screening title, keywords and abstracts resulted in 103 articles for further analysis. Of these, 35 were categorized as reviews without original patient series and thus excluded. Screening reference lists resulted in seven additional studies.[42-48] Another report[49], which was not identified in the initial Medline search, was added because a commentary letter[50] was addressed. Thus, 76 articles were evaluated: three of these were abstracts from meetings.[42,43,47] A similar search in the Science Citation Index did not result in any new papers.

A few authors published the outcome (partially) of the same patient series in several papers.[51-53/54,55/47,56/57,58/59,60/61,62/63,64] These were judged individually.

Table 2: **Inclusion criteria**

	Description
Study design	prospective design, no retrospective design
Population	demographic population only
Follow-up	follow-up minimally 3 years and loss to follow-up < 10%
Endstage	endstage assessment well-defined using a reproducible scoring system and no operation performed

Thirty-four publications did not meet any of the present inclusion criteria.[11,43,44,46,49,51-55,57,58,60,62,63,65-83] Of the remaining 42 articles, 35 met one criterion, and seven articles met two criteria (Tables 3 and 4). As none of the articles met three or four criteria, the protocol was changed in that a second reviewer evaluated the papers meeting two inclusion criteria. These results were in agreement with those of the first reviewer.

Study design

Eleven studies had a prospective design.[5,27,36,37,42,47,56,84-87] Four of these described the prospective follow-up of patients referred to a tertiary center.[27,42,47,56] In the remaining seven, the infants were screened prospectively on the presence of OBPL. Three of these seven focused on the incidence of OBPL or associated risk factors, and follow-up was not performed.[85-87] Three of the eleven prospective studies met a second criterion: one on population[36] and two on proper end stage assessment[37,84]

Study population

Six studies described a demographic population.[4,7,8,36,88,89] Follow-up was not performed in two.[88,89] Two of the remaining four studies had sufficient assessment of endstage, thereby meeting a second criterion.[8,36]

A second population under study were hospital-born patients, comprising approximately 1300 patients in 36 reports.[5,37,44-46,49,54,55,57,58,63,64,66-68,70-72,76,77,79,81-87,90-97] The third type of population under study were patient series referred to tertiary centres. Thirty-four such studies were identified, reporting approximately 2000 patien ts.[10-12,27,42,43,47,48,51-53,56,59-62,65,69,73-75,78,80,98-108]

Follow-up

The follow-up period was at least three years with less than 10% of patients lost in five studies.[12,59,101,106,107] All five presented the outcome of patients referred to a tertiary centre. Three of these provided sufficient end stage assessment without including surgical results.[12,106,107]

Assessment of endstage

Twenty-seven studies used a well-defined and reproducible scoring system without surgical procedures influencing the outcome.[8,10,12,37,45,48,61,64,84,90-100,102-108] Six of these met a second inclusion criterion for design[37,84], population[8], or follow-up[12,106,107].

Discussion

The main result of this systematic literature study was that no publication on the natural history of OBPL met more than two of our four inclusion criteria. These results imply that the common perception of spontaneous recovery from OBPL is, in fact, inaccurate.

The validity and importance of each criterion will be discussed in more detail.

Table 3: **Studies which met at least one inclusion criterion**

Publication		Study characteristics			Inclusion criteria			
Citation	Year	Location	Period	Patients	Design	Population	Follow-up	Endstage
Studies meeting 2 criteria								
36	1973	Collaborative Perinatal Study, USA	1973-1973	60	x	x		
84	1988	San Fransisco, USA	1983-1986	21	x			x
37	1993	Oulu, Finland	1981-1983	10	x			x
8	1988	Malmo, Sweden	1973-1983	48		x		x
12	1966	Copenhagen, Denmark	1920-1930	103			x	x
106	1982	Milan, Italy	1965-1975	34			x	x
107	1984	Kagawa, Japan	N/M	15			x	x
Studies meeting 1 criterion								
87	1991	Kuala Lumpur, Malaysia	1986-1987	42	x			
86	1993	Abha, Saudi-Arabia	1986-1987	10	x			
42	1996	Denver, USA	1984-1992	155	x			
47	1996	St Louis, USA	N/M	67	x			
85	1996	Haifa, Israel	1994-1995	11	x			
5	1997	Al Ain, United Arab Emirates	1993-1995	27	x			
27	1999	Boston, USA	1989-1995	66	x			
56	2001	St Louis, USA	1991-1997	94	x			
7	1971	Islands Lolland and Falster, Denmark	1960-1970	12		x		
88	1986	Washington State, USA	1980-1982	106		x		
4	1997	Skanaborg, Sweden	1980-1989	52		x		
89	1999	California, USA	1994-1995	164		x		
101	1991	Stanford, USA	1976-1982	25			x	
59	1998	Stockholm, Sweden	1987-1993	105			x	
48	1965	Rostock, Germany	1947-1962	25				x
10	1967	New York, USA	1939-1962	123				x
61	1971	Washington, USA	1967-1969	25				x
95	1973	Singapore	1969-1971	57				x
64	1974	Lille, France	1989-1995	20				x
90	1976	London, England	1960-1974	21				x
96	1976	Amsterdam, Holland	1959-1973	40				x
99	1977	Glanzing, Austria	1961-1971	26				x
92	1981	Auckland, New Zealand	1969-1978	36				x
104	1984	Nancy, France	N/M	31				x
91	1984	San Fransisco, USA	1972-1982	61				x

Table 3 – continued

Publication		Study characteristics			Inclusion criteria			
Citation	Year	Location	Period	Patients	Design	Population	Follow-up	Endstage
103	1986	Lagos, Nigeria	1980-1982	26				x
93	1991	Yaroslavl, Russia	1984-1987	122				x
45	1993	Indiana, USA	1986-1990	33				x
102	1996	Kuopio, Finland	1975-1990	46				x
105	1996	Oslo, Norway	1990-1995	39				x
100	1996	Washington, USA	1981-1993	162				x
94	1997	Messina, Italy	1990-1994	28				x
97	1998	Lille, France	1989-1995	20				x
108	1999	Istanbul, Turkey	N/M	13				x
98	2000	Shamiya, Kuwait	N/M	52				x

N/M, not mentioned; x indicates criterion present.

Study design

Preferably, studies describing the prognosis of OBPL should have a prospective design because retrospective studies may have underestimated the number of patients as a result of the shortcomings of coding systems and registrations. This difference can be illustrated by two large retrospective population-based studies of birth records[88,89] in which an important lower incidence of OBPL was found compared with a prospective study[36] in the same country, namely 0.15 and 0.50 versus 1.89 per 1000 births. A possible explanation for the difference may be an inclusion bias in the retrospective studies. Patients with good spontaneous recovery may have been excluded.

Similarly, the severity and prognosis may appear worse in retrospective compared with prospective studies. This is demonstrated in Table 4 where the three prospective series show rates of failing recovery ranging from 5 to 19% and the four retrospective studies from 27 to 93%.

Study population

An ideal series would consist of a population which is not selected in any way, and in which inflow and outflow are restricted to a minimum. One of the best examples of such a secluded demographic population are the inhabitants of an island. We found one paper describing a 10-year period on the islands of Lolland and Falster (in Denmark). Twelve cases of OBPL were reported among 18 640 births, eight of whom recovered fully.[7] Other demographic studies investigated the natural history of OBPL in a city, district or state.[4,8,88] Two studies pooled data from several hospitals to create a 'demographic' population.[36,89]

The second type of population under study consisted of hospital-born children. The major disadvantage of these studies is a referral bias. Midwives may perform low-risk deliveries at home, which might result in a tendency to overestimate the incidence and severity of OBPL in hospital-based series.[54]

The third type of population dealt with OBPL patients referred to tertiary centres. These studies are likely to have underestimated the rate of spontaneous recovery because, usually, only severe cases are referred.[29]

Follow-up

The preferred minimum follow-up period for OBPL is three years. By that time regenerating axons should have reached their target organ, in view of the relatively short distance and speed of axonal outgrowth. The validity of this somewhat arbitrary limit is borne out by reports describing a steady functional state after two to three years.[4,10,74,90] Only two authors reported ongoing recovery after this period, in sporadic cases.[102,107]

Additionally, neurological examinations become more accurate as children get older because of their increased ability to cooperate.[8] End stage assessment should preferably take place in adult patients as subtle remnants such as "increased fatigability" can only be documented by that time.[106]

Length of follow-up varied from a few days (after discharge from the hospital)[49] to 64 years[12]. Only five publications met the present inclusion criteria for duration of follow-up (> 3 years) and proportion of patients lost to follow-up (<10%).[12,59,101,106,107] All five studies dealt with patients referred to specialized centres, and, therefore, contribute only in a limited way to the survey of the entire spectrum of OBPL.

Assessment of end stage

Ideally, the end stage of OBPL should be assessed using a well-defined and reproducible protocol. A few such scoring systems have been introduced which could also serve as useful tools to compare and pool patient series. Examples are the Mallet score[15] and the Toronto Scoring System.[16,78] Other protocols assessed the functional use of the affected limb.[59,106] A few studies investigated cognitive or psychological features in addition to a neurological examination.[11,36]

In this review 27 studies used a well-defined scoring system. However, a wide range of different scoring-systems was encountered. Most authors used their own definitions to assess outcome as being good, moderate or poor. These assessments are often based on volitional muscle power. While these articles provided a reasonably useful survey of the outcome in their respective populations, data documentation was not sufficiently clear to compare them with other series. Therefore, we could not pool the results for a meta-analysis.

Today nerve reconstructive surgery is widely applied to improve the function of patients with incomplete spontaneous recovery.[16,17,22-27] The decision for surgery is generally taken in a time window ranging from 3 to 9 months, i.e. long before an end stage of spontaneous recovery is reached. Therefore, the outcome from series in which some of the patients were selected for surgery, cannot be used to describe the natural course of this condition.

Studies meeting two criteria

The seven studies that met two inclusion criteria will be discussed in more detail. (Table 4)

The Collaborative Perinatal Study[36] was prospective and population-based, and showed the most favourable outcome encountered in this review: only 5% of patients

Table 4: **Analysis of studies meeting two inclusion criteria**

Publication	Inclusion criteria				Outcome			
Citation; Year	**Study design**	**Population**	**Follow-up**	**Assessment of endstage**	**Patients/Births Incidence**	**Not recovered**	**Recovered**	**Lost to follow-up**
Gordon et al. [36]; 1973	**prosp**	**pooled demographic population**	min 2 mo	poor	60 / 31700 1.89	5%	88%	7% *
Walle et al. [37]; 1993	**prosp**	hospital-born series	'until recovery' min not stated	**good**	10 / 5082 1.97	10%	90%	-
Jackson et al. [84]; 1988	**prosp**	hospital-born series	min 12 mo max 4 yr not recovered min 26 mo	**good**	21 / 8258 2.54	19%	71%	9%
Sjöberg et al. [8]; 1988	retrosp	**demographic population**	'until pronounced to be fit' min 1 mo 15 patients at 3-12 yr	**good**	48 / 25736 1.87	27%	73%	-
Tada et al. [107]; 1984	retrosp	tertiary center	**min 5 yr**	**good**	15#	93%	7%	-
Rossi et al. [106]; 1982	retrosp	tertiary center	**min 3 yr**	**good**	34#	88%	12%	-
Gjørup [12]; 1966	retrosp	tertiary center	**min 3 yr** **max 64 yr**	**good**	103#	29%	64%	7%

Criteria in **bold** met inclusion criteria; min: minimal; max: maximal; mo: months; yr: years; incidence: per 1000 births; prosp: prospective; retrosp: retrospective

incidence could not be calculated from studies describing referred patients

* one patient was excluded by the author from follow-up because of twin pregnancy

did not recover. However, the presence of mild deficits may have been underestimated, as good recovery was diagnosed as early as one year of age in one-third of patients.

Two studies concern prospective identification and follow-up of patients born in a university hospital, along with a good end stage assessment.[37,84] The first comprised only 10 patients, one of whom did not recover.[37] Four of the 21 patients in the second series had residual deficits.[84] Both series lacked sufficient follow-up.

A population based study with good end stage assessment found residual paresis in approximately one quarter of their patients.[8] This study is of particular interest in terms of follow-up. Four patients diagnosed as 'almost well' at the age of 3 to 17 months were thoroughly re-examined at ages ranging from 4 to 12 years. The authors found atrophy and restricted joint movements in these four patients, although no limitations were recorded in daily life. The authors stated that the functional demand for the arm may increase with age, so these patients may encounter more difficulties when they get older. The rate of incomplete recoveries in this series increases to 35%, when these four cases are added to the number of persistent palsies.

The remaining three studies had a sufficiently long follow-up and adequate end-stage assessment. However, they concerned a retrospective analysis of patients referred to a specialized centre.[12,106,107] Because of this selection their outcome does not represent the natural course of OBPL.

These seven studies differ greatly in study design, patient selection, follow-up and assessment. As a consequence the conclusion which can be drawn from these studies varies greatly. It could be argued that the inclusion criteria should not be weighed evenly. However, each separate criterion is mandatory for an unbiased evaluation of the natural history of OBPL. It should be clear that given the absence of papers meeting all our criteria, no definite conclusion can be drawn.

However, the two studies which come closest to the "ideal" study show a tendency towards some 20-30% residual deficits[8,84], in contrast to the optimistic view of over 90% complete or almost complete recovery which is often encountered.

Conclusions

Although the natural history of OBPL has been described in a large number of studies, no study had the width of scope necessary to answer the question posed. Therefore, the often-cited excellent prognosis for this type of birth injury cannot be considered to be based on scientifically sound evidence.

The few studies that met two of the four inclusion criteria suggest that spontaneous recovery is notably worse than is generally suggested in most reviews. Therefore, physicians should exercise caution in predicting excellent recovery shortly after birth, and seek an active treatment attitude to avoid life-long limitations for the individual patient.

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