

# OBSTETRIC BRACHIAL PLEXUS PALSY

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**Background:** Obstetric brachial plexus palsy (OBPP) at birth is one important cause of neurological disability in children and adults; the incidence has increased substantially in Sweden over the past decade. Several risk factors for OBPP have been identified, e.g. high birth weight and shoulder dystocia, but it remains difficult to predict based on antenatally available information. Despite extensive research on OBPP, there is no generally accepted strategy for prevention. The overall objective of this thesis is to study predisposing risk factors for OBPP in vaginally delivered infants and to identify specific obstetric procedures that are strongly associated with OBPP. Based on this knowledge, our ultimate aim is to develop a prevention strategy with respect to manual assistance in the second stage of labour.

**Methods:** Paper I reports on a retrospective case-control study aimed at investigation of the incidence and risk factors for OBPP in a large population studied in 1987-1997. All deliveries recorded in the Swedish Medical Birth Register during the period were investigated. Cases of OBPP were compared with all cases without OBPP. Paper II describes a retrospective case-control study, the purpose of which was to identify risk factors for OBPP, specifically in women delivered by vacuum extraction. The groups with and without OBPP were compared with regard to possible risk factors, including those linked to the vacuum extraction procedure. The aim of the prospective population-based case-control study presented in Paper III was to evaluate the association between OBPP and obstetric manoeuvres during the second stage of delivery. Obstetric management in OBPP cases was compared to management in a randomly selected control group. Paper IV and Paper III are based on the same cases. The aims of the study presented in Paper IV were a) to describe neurological deficits in children with remaining OBPP at 18 months of age; b) to compare maternal, infant and obstetric data in infants with and without OBPP at 18 months of age and c) to evaluate if differences in force applied in downward traction of the head (ranked on a 100-point visual analogue scale) correlated to the number of affected nerve roots (C5-C6, C5-C7, C5-Th1).

**Results:** The incidence of OBPP in Sweden increased from 0.17% to 0.27% between 1987 and 1997. Infants with high birth weight, especially >4500 g, were at increased risk of shoulder dystocia and OBPP. However, high birth weight was not or only weakly over-represented among children with persistent OBPP, compared with those who recovered, suggesting that other factors are important. Hence, many factors related to prolonged second stage and difficulties in delivering the shoulders were overrepresented in the OBPP group, compared to controls. In particular, forceful downward traction applied to the head after the fetal third rotation represented an important risk factor for OBPP in cephalic-presentation vaginal deliveries. Downward traction of the head had been applied more often and with greater force in the group with persistent damage and there was a significant correlation between the force used and the number of affected nerve roots.

**Conclusion:** We have confirmed that well known factors such as birth weight and shoulder dystocia are indeed important risk factors for OBPP. Furthermore, obstetric procedures, especially forceful downward traction of the head, conferred additional risk of OBPP at birth, as well as at follow-up at age 18 months. Our data suggest that forceful downward traction of the head should be avoided and indicate that other measures should be taken to release the impacted shoulder, e.g. maximal flexion of the maternal hips, rotation of the shoulders and/ or extraction of the posterior arm.

Key words: obstetric brachial plexus palsy, shoulder dystocia, obstetrical manoeuvres

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## LIST OF ORIGINAL PUBLICATIONS

This thesis is based on the following original papers, which will be referred to in the next by their Roman numerals:

- I High birthweight and shoulder dystocia: the strongest risk factors for obstetrical brachial plexus palsy in a Swedish population-based study.**  
Mollberg M, Hagberg H, Bager B, Lilja H, Ladfors L.  
Acta Obstet Gynecol Scand 2005; 84:654-9.
- II Risk factors for obstetrical brachial plexus palsy among neonates delivered by vacuum extraction.**  
Mollberg M, Hagberg H, Bager B, Lilja H, Ladfors L  
Obstet Gynecol 2005; 106:913-18.
- III Obstetric brachial plexus palsy: a prospective study on risk factors related to the manual assistance during the second stage of labor.**  
Mollberg M, Wennergren M, Bager B, Ladfors L, Hagberg H.  
Acta Obstet Gynecol 2007; 86:198-204.
- IV Comparison of infants with transient and persistent obstetric brachial plexus palsy: differences in obstetric management**  
Mollberg M, Lagerkvist A-L, Johansson U, Bager B, Johansson A, Hagberg H, Uvebrant P.  
Manuscript.

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## **ABBREVIATIONS**

|             |                                    |
|-------------|------------------------------------|
| <b>AOR</b>  | adjusted odds ratios               |
| <b>BMI</b>  | body mass index                    |
| <b>CI</b>   | confidence interval                |
| <b>MBR</b>  | the Swedish Medical Birth Register |
| <b>NaCl</b> | Isotonic saline                    |
| <b>OBPP</b> | obstetric brachial plexus palsy    |
| <b>OR</b>   | odds ratio                         |
| <b>SD</b>   | standard deviation                 |
| <b>VAS</b>  | Visual Analogue Scale              |

# CONTENTS

|  |    |
|--|----|
| <b>INTRODUCTION</b>                                      | 1  |
| Anatomy and physiology                                   | 1  |
| Incidence  | 2  |
| A historical perspective and classification of OBPP      | 2  |
| Diagnosis of OBPP after birth and clinical presentations | 3  |
| Treatments   | 4  |
| Prognosis  | 5  |
| Consequences of OBPP                                     | 6  |
| Pathogenesis   | 6  |
| The force factor   | 7  |
| Uterine and maternal expulsive forces                    | 7  |
| Risk factors   | 8  |
| Delivery of the shoulders                                | 9  |
| Shoulder dystocia  | 9  |
| The mechanism of the force factor                        | 10 |
| <b>AIMS OF THE STUDY</b>                                 | 12 |
| <b>MATERIAL AND METHODS</b>                              | 13 |
| Paper 1  | 13 |
| Paper II   | 13 |
| Paper III  | 14 |
| Paper IV   | 15 |
| Statistical analysis                                     | 15 |
| Paper 1  | 15 |
| Paper II   | 16 |
| Paper III  | 16 |
| Power analysis   | 17 |
| Paper IV   | 17 |
| Methodological considerations                            | 18 |
| Ethical approval   | 18 |
| <b>SUMMARY OF RESULTS</b>                                | 19 |
| Paper 1  | 19 |
| Paper II   | 19 |
| Paper III  | 20 |
| Paper IV   | 22 |
| <b>DISCUSSION</b>  | 23 |
| <b>CLINICAL RELEVANCE OF THIS THESIS</b>                 | 29 |
| <b>CONCLUSIONS</b>                                       | 30 |
| <b>ACKNOWLEDGEMENTS</b>                                  | 31 |
| <b>REFERENCES</b>  | 33 |

## INTRODUCTION

Obstetric brachial plexus palsy (OBPP) at birth is one of the causes of permanent neurological disability. The delivery process is associated with several factors which, together, may result in OBPP in the newborn but it is difficult to predict OBPP based on antenatally available information. Despite extensive research on OBPP, there is no generally accepted strategy for prevention. The overall objective of this thesis is to study predisposing factors for OBPP in vaginally delivered infants and to develop a prevention strategy associated with manual assistance in the second stage of labour.

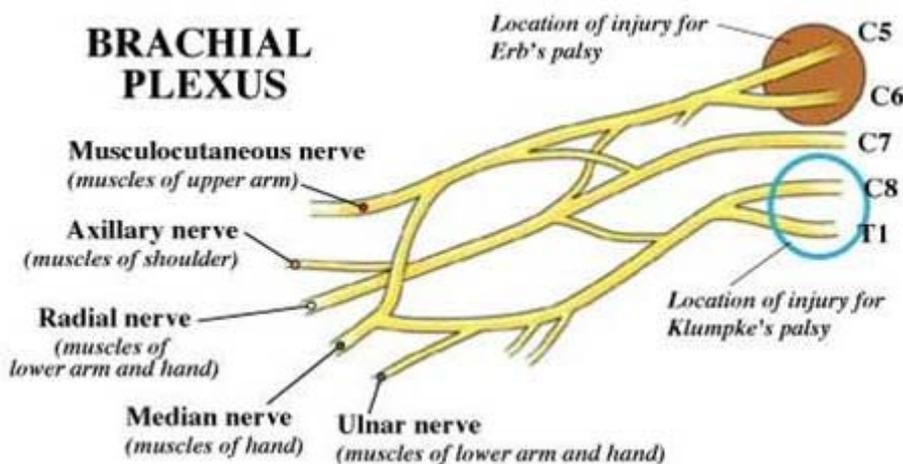


Figure 1 Anatomy of the brachial plexus

Knowledge of the regional anatomy of the brachial plexus is fundamental for understanding the manoeuvres that increase the risk of OBPP. The brachial plexus is composed of the five spinal nerve roots C5, C6, C7, C8 and Th1. These five nerve roots combine into 3 trunks formed above the clavicle, the upper at the C5-C6 level (location of damage underlying Erb's palsy), the middle at C7 and the lower at C8-Th1 (location of damage underlying

Klumpke's palsy). The cords terminate in 5 main peripheral nerves: the musculocutaneous, axillary, radial, median and ulnar nerves. The brachial plexus provides the nerves to muscles in the entire arm and shoulder and is essential to hand function (see Appendix I). OBPP is defined as neurological motor deficit resulting from damage to any of the C5-Th1 nerve roots. The preganglionic sympathetic nerve fibres from the Th1 nerve root are also important for nervous system function, providing a variety of autonomic functions including pupillary dilatation and eyelid movement (1-5).

### **Incidence**

The incidence of OBPP is between 0.5% and 5 % in countries with well developed obstetric services (6-12). Reduction in incidence from 1.56 per 1000 live births in 1938 to 0.28 per 1000 in 1962 was reported in New York. The authors ascribed the reduction to improved obstetric practice (13). Subsequent reports, including two population-based studies from Sweden (10, 11), have suggested an increase in the incidence, possibly due to an increase in mean birth weight (10, 11, 14-16). However, the incidence in the United Kingdom and Ireland has not changed in nearly 40 years (17). Among 776 618 live births surveyed in the United Kingdom and the Republic of Ireland, the reported incidence of OBPP was 0.42 per 1000 live births. The authors do admit that methodological factors may have led to underestimation of the incidence by up to 11% (17). They comment that the incidence is similar to that in 1962 despite an increase in caesarean section rates and a decline in the number of vaginal breech deliveries in recent years, one possible reason being the concurrent increase in birth weight.

### **A historical perspective and classification of OBPP**

OBPP was first described by the Scottish obstetrician William Smellie in an article for midwives in 1764 (18). However, it was not until a century later

(1872 and 1875) that a clinical description of the aetiology was presented by the French neurologist Guillaume Duchenne (19) and the German professor of neurology Wilhelm Erb (20). They described the injury involving the C5-6 (the upper trunk) and sometimes the C7 (the middle trunk) level. Erb and Duchenne, have since then been linked to this condition and OBPP is commonly called Erb-Duchenne palsy. Ten years later, in 1885, the French neurologist Augusta Klumpke (21) described the clinical picture of OBPP affecting the lower plexus trunk at C8-Th1, leading to paralysis of the muscles of the hand and ipsilateral pupillary dilatation, called Horner's sign (1, 3, 4). Therefore, OBPP affecting the lower trunk is often referred to as Klumpke's palsy. OBPP diagnosed at birth is defined into three groups in accordance with the International Classification of Diseases (22):

- P 14.0 Erb's palsy caused by injury at delivery
- P 14.1 Klumpke's palsy caused by injury at delivery
- P 14.3 Other injury of the brachial plexus at delivery

P 14.0 and P 14.1 are the most common diagnoses. P 14.3 is rarely used and seems to be applied in cases with a vague suspicion of OBPP at the time of birth. Alternatively, it is applied in cases when the OBPP has healed completely upon follow-up and more specific classification is thus difficult (personal communication, Dr Karin Sävman).

### **Diagnosis of OBPP after birth and clinical presentations**

OBPP is a clinical diagnosis made by the paediatrician, based on observations and manipulations of the infant. At the time of diagnosis the infant has a reduced Moro reflex on the affected side. Differential diagnoses are fractures which can camouflage OBPP during the first days of life and pseudoparesis and neurological lesions at other neuroanatomical levels (23). The paediatrician can

determine which of the spinal nerves (C5-Th1) are involved in the OBPP, but not how severely the nerve fibres are injured. There are different forms of OBPP classified according to the extent of damage (stretching, compression, rupture or avulsion of the nerve fibres) (1, 24). A. Narakas (25) introduced a classification in four groups, based on the clinical course during the first 8 weeks of life. The classification is not anatomic, but grades the overall severity of the OBPP according to the number of injured nerves and the severity of the nerve injury; Group I represents the mildest and Group IV the most severe form of disability.

Table 1 Narakas' classification of OBPP

|           |        |   |
|-----------|--------|---|
| Group I   | C5-6   | Paralysis of the shoulder and biceps                    |
| Group II  | C5-7   | Paralysis of the shoulder, biceps and forearm extensors |
| Group III | C5-Th1 | Complete paralysis of the limb                          |
| Group IV  | C5-Th1 | Complete paralysis of the limb with Horner's syndrome   |

### **Treatments**

There are two therapeutic approaches, conservative or surgical, the latter involving nerve reconstruction and shoulder surgery. Conservative treatment includes passive range of motion therapy aimed at avoiding contractures and stimulating the use of the hand while playing and during everyday activities. In Sweden, the parents are put in contact with a physiotherapist in the maternity ward; he/she gives instructions about passive range of motion therapy. Children with OBPP sequelae will subsequently be put in contact with the local rehabilitation team (26).

About 250 children contract OBPP every year in Sweden, some 25% of whom will have permanent sequelae (26). Surgical intervention to reconstruct injured brachial plexus nerves is performed when there is inadequate recovery of the

shoulder and elbow function during the first months of life (3, 27, 28). Absence of biceps function is regarded as the key indication for surgical exploration (28, 29). The decision to operate is based on a series of clinical examinations and OBPP cases recommended to undergo surgery are referred to the national brachial plexus clinic at the Astrid Lindgren Children's Hospital in Stockholm (26).

### **Prognosis**

The outcome of OBPP depends on the severity of the nerve injury (Table 1) and the number of roots involved (1, 30-32). Bilateral OBPP is reported in up to 23% of cases and occurs mostly in the setting of breech presentation, during which excessive traction on both shoulders may be applied to deliver the head (15, 33). The majority of infants with OBPP have a unilateral lesion affecting C5-C6 (C7) (34). Injuries at the C5-7 level usually lead to a mild form of OBPP and about 75% of the children recover within the first year of life. Newborns with complete OBPP, affecting C5- Th1 and with or without Horner's sign, never fully recover. According to several previous studies (15, 33), recovery begins early, by the age of a few weeks, and proceeds up to at least the end of the third month (1, 35-37).

In a review of more than 1000 studies of the natural history of OBPP (7), Pondaag et al. are critical to the often-quoted statement that the prognosis is good in up to 90% of cases; the authors point out methodological limitations in most studies. In their opinion (7), there are five methodological quality criteria that must be met when discussing prognosis: a prospective study design, follow-up of at least three years, pre-defined functional end stage, surgically treated children should be analyzed separately and only few cases (<10%) can be lost to follow up. Only two studies (10, 38) come close to this "ideal" study design; they showed that at least 20% to 30% of cases have residual deficits after OBPP.

## **Consequences of OBPP**

There is limited information about the long-term effects of OBPP in children and adults. The functional outcome in five-year-olds with C5-C6 involvement has been described; it is apparently more complex than is commonly believed. Hand function was affected due to the effect of limited shoulder movements on hand positioning and reduced grip strength. Children with total OBPP (C5-Th1) had diminished grip strength and tactile sensibility; performance of daily life activities was affected as were bimanual activities requiring use of the injured hand (26, 39). Differences in functional outcome have been described between 5 and 20 years of age. A decrease in elbow flexion and a commonly occurring restriction in external rotation of the shoulder were reported to occur with age (26). Other authors (40) reported increasing disability and exacerbation of symptoms, such as pain, joint problems and reduced function, with age.

## **Pathogenesis**

According to Smellie's (18) and Duchenne's (19) original descriptions, OBPP is caused by difficult delivery of the shoulders; this is supported by anatomic studies (41). This was the predominant theory regarding the aetiology of OBPP for many years (32) but additional pathogenetic models have recently been proposed (1, 2, 7, 42). The various aetiological models can be summarized as follows:

1. Intrauterine factors, i.e. abnormal intrauterine pressures arising from uterine anomalies, cause OBPP during pregnancy (43).
2. Damage of the brachial plexus is caused by traction of the head and neck in cases of shoulder dystocia or breech delivery (1, 2, 15, 44-48).
3. OBPP occurs as the result of a precipitous second stage. The rapidity of descent may prevent the shoulders from rotating adequately in the birth canal, which may cause nerve root trauma (45).

4. Endogenous maternal propulsive forces related to uterine contractions and active pushing cause OBPP (49-53).
5. In cases of caesarean section (9, 43, 54-59) or operative vaginal delivery (60), OBPP may result from forceful traction and manipulation by the obstetrician.

Most of the explanations offered by Stevens et al. (41) are hypothetical and, with one exception (60), based on reasoning rather than on evidence.

### **The force factor**

Traction of the head applied by the clinician in shoulder dystocia situations is likely to cause OBPP (61). The occurrence of OBPP depends on the degree, direction (axial or lateral) and rate of the force applied. Experimentally measured delivery forces showed that clinician-applied traction increases with difficulty of the delivery (62-65). Axially applied traction will not affect the brachial plexus, whereas even light lateral flexion of the neck will place significant tension on it (Figure 1).

### **Uterine and maternal expulsive forces**

Uterine forces may stretch the brachial plexus as the head and shoulders traverse the mid-pelvis and pelvic outlet in simulated deliveries (49). Two types of loading forces have been studied, endogenous (maternal pushing with uterine contraction) and exogenous (physician-applied traction of the head). The point of departure of each experiment was the assumption that the head had already been delivered. Stretching of the brachial plexus was seen with both endogenously and exogenously applied loads. Exogenous forces were applied to the head in a 45-degree downward direction. For clinician-applied forces, the head was either fixed, aligned with the axis of the infant's neck and torso or freely mobile to allow lateral flexion. The greatest amount of brachial plexus stretching was seen with endogenously applied loads in the lithotomy position or

with exogenous traction and lateral flexion of the head. But since uterine forces are axially transmitted, they are not likely to produce the lateral deviation of the head from the shoulders needed to stretch the brachial plexus beyond its elastic limit. Since the midwife and obstetrician can only control maternal pushing and uterine forces to a limited degree, the goal must be to avoid clinician-applied traction. OBPP occurring during the delivery process or in utero before delivery is caused by stretching of the brachial plexus (49).

### **Risk factors**

Most infants (99%) diagnosed with OBPP after birth are born in the cephalic presentation in spontaneous or vacuum-assisted vaginal deliveries (9, 66, 67). Numerous risk factors have been proposed for OBPP and the probability of its occurring is influenced by both maternal, delivery and infant characteristics. OBPP occasionally occurs in infants weighing <2500 g (68). The most significant combined maternal and infant risk factors referred to in the literature are neonatal macrosomia and shoulder dystocia (9, 12, 69-71). However, there are also reports contradicting an increase in OBPP with increasing birth weight (72). The definition of macrosomia is not universal; cut-off points vary between 4000 g, 4500 g and 5000 g. The risk of shoulder dystocia due to macrosomia must be related to the mother's size and the capacity of her pelvis; the risk of shoulder dystocia thus varies with maternal height (73). Several studies have shown that increasing birth weight is strongly associated with an increasing risk of shoulder dystocia (69, 70, 74). Many investigators focus on attempting to prevent shoulder dystocia rather than on the risks linked to obstetric manoeuvres to free the impacted shoulder (75). Much of the understanding and knowledge of shoulder dystocia is based on empiric observations. It is still unclear why some infants suffer from OBPP after delivery and how manual assistance has differed in cases of transient and persistent OBPP (61, 75, 76).

Shoulder dystocia is a serious clinical condition that may result in neonatal morbidity and sometimes even neonatal death. The incidence of perinatal death in deliveries complicated by shoulder dystocia varies between 0 % and 27% (77). The risk of clavicle and humerus fractures is estimated at 11% (78, 79) and 4% (58, 78, 79), respectively. It is usually the anteriorly located arm that is affected by OBPP and damage occurs during the third and fourth stages of delivery (1, 71, 80-82).

### **Delivery of the shoulders**

In cases of shoulder dystocia it is technically more difficult to deliver the shoulders and the delivery dynamics are different, compared to the normal situation. The head enters the maternal pelvis in the transverse diameter as the inlet is wider transversely than antero-posteriorly. During the second stage, the vertex rotates axially, as the outlet is wider in the sagittal plane, followed by a transverse positioning of the shoulders in the inlet. The shoulders subsequently rotate into the oblique, and finally into the sagittal plane, when delivered under normal circumstances. When the midwife actively assists delivery, one hand is placed on the head over the ears and a gentle downward traction is applied, allowing the anterior shoulder to slip beneath the pubic symphysis while the posterior shoulder remains in the vagina. When the anterior crease is seen, the head and trunk are guided in an upward curve to allow the posterior shoulder to escape out past the perineum (83). Gentle traction is defined as the traction required for delivery of the infant during uncomplicated conditions (82-84).

### **Shoulder dystocia**

Shoulder dystocia occurs when the shoulder(s) fail to pass through the pelvis spontaneously after delivery of the head, most likely due to a size discrepancy between the shoulders and the birth canal. It occurs either when the posterior shoulder enters the pelvis and the anterior shoulder remains hooked behind the

pubic symphysis, failing to rotate, or due to impaction of the posterior shoulder on the maternal sacral promontory. Less commonly, both shoulders remain at the level of the inlet (34, 85). Traction of the head at that point will further impact the anterior shoulder (83). Shoulder dystocia may present as the “turtle” sign but is not diagnosed until attempts to deliver the shoulders by downward traction of the head are unsuccessful (76). In cases of shoulder dystocia, completion of delivery depends on specific manual obstetric manoeuvres by the midwife and/or obstetrician (Appendix II), (87-95). There is no clear evidence that one particular action listed in the guidelines is more important than the others (95). There are three aims in the management of shoulder dystocia, i.e. to increase the functional size of the maternal bony pelvis, to decrease the shoulder diameter and to change the relationship of the shoulder diameter within the bony pelvis (Appendix II).

### **The mechanism of the force factor**

If OBPP occurs in utero before delivery, it is caused by stretching of the brachial plexus nerves. When there is no mechanical disruption to the nerve or the axons, the stretching results in temporary dysfunction of nerve conduction (23). Since midwives and obstetricians can only exert limited control over maternal pushing and uterine forces, the initial focus must be on how clinician-applied traction may be reduced (61).

Few studies have specifically attempted to correlate the number and type of manoeuvres used to manage shoulder dystocia with subsequent perinatal outcome. Some authors imply that OBPP is related to the force used in downward traction and the degree of difficulty of obstetric management (58, 95, 96). However, retrospective studies are unable to quantify the lateral traction exerted on the head and its correlation to OBPP (61, 95).

Others (46, 97, 98) go so far as to venture that “OBPP is avoidable by never applying head traction during any delivery and by using manoeuvres to deliver the shoulders that avoid any tension on the brachial plexus. Do not be tempted to use further traction until another corrective manoeuvre has been achieved. Never use excessive force and do not use fundal pressure because this pressure risks uterine and fetal damage and serves only to impact the anterior shoulder more firmly onto the pubic symphysis” (46, 97, 98).

There are no general recommendations to support elective caesarean delivery for prevention of shoulder dystocia in women at risk (99, 100), except in cases of extremely high estimated fetal weight, i.e.  $\geq 5000\text{g}$  (101, 102). Since historic risk factors are not clinically useful for predicting shoulder dystocia, interest has turned toward prophylactic manoeuvres during delivery. The absence of detailed documentation describing the manoeuvres used to resolve shoulder dystocia often results in an inconsistent and counterproductive representation of the facts (75, 103-112).

In summary, shoulder dystocia is an obstetric complication for which there is little evidence-based data, in the form of prospective and randomized controlled trials, to support specific methods of prediction, prevention and management. Much of the understanding and knowledge of shoulder dystocia is therefore based on empiric observations and studies of limited scientific quality (61, 75). In addition, there is also limited information on how the “theoretical” aetiology of OBPP applies to obstetric practice and clinical reality.

## **AIMS OF THE STUDY**

The overall objective of this thesis was to study predisposing factors for OBPP in infants delivered vaginally in the cephalic presentation and to develop a hypothetical prevention strategy associated with manual assistance during the second stage of labour.

The specific aims were to:

identify the incidence of OBPP in Sweden and its underlying risk factors.

study risk factors for OBPP in infants delivered vaginally, either spontaneously or by vacuum extraction.

prospectively evaluate which events and obstetrical manoeuvres during the second stage of labour were associated with subsequent OBPP, diagnosed after birth, and which of these factors correlated to severe OBPP with remaining motor disability at 18 months of age .

prospectively a) describe neurological deficits in children with remaining OBPP at follow-up, b) compare maternal, infant and obstetric factors in infants with and without OBPP at 18 months and c) evaluate if differences in force used in downward traction of the head correlated to the affected nerve roots (C5-C6, C5-C7, C5-Th1).

## **MATERIAL AND METHODS**

### **Paper I**

The study design was retrospective case-control. The aim was to study the incidence of OBPP and to analyze risk factors. All women recorded (n=1 213 987) in the Swedish Medical Birth Register (MBR) during the period from 1987 to 1997 were investigated. A total of 2 399 infants (singletons and twins) who were diagnosed with Erb's or Klumpke's paralysis in the maternal records in the register were analyzed. Antenatal, intrapartum and infant variables for all cases of OBPP were compared to those for all other deliveries in the database. Outcome if ultrasound had been used to detect fetuses with an estimated birth weight  $\geq 5000$  g at  $\geq 37$  weeks was analyzed. In addition, the effect on OBPP incidence of a scheduled caesarean section if ultrasound indicated a birth weight  $\geq 5000$  gram was evaluated. These calculations were based on information from the 1 135 404 deliveries at  $\geq 37$  weeks for which information about gestational age and birth weight was available.

### **Paper II**

The study plan describes a retrospective case-control study, the purpose of which was to identify risk factors for OBPP, specifically in women delivered by vacuum extraction. Data in the MBR consists of 120 variables. During 1995-1997, a national evaluation protocol with an additional 25 variables related to vaginal instrumental delivery was used. Data were collected in a national register for operative vaginal deliveries and linked to the MBR to supplement its data. Only vaginal deliveries with a completed instrumental delivery protocol were included; i.e. deliveries lacking a protocol or completed by forceps were excluded. According to the register, there were 281 575 deliveries during the period; 7.3% of the infants were born via vacuum extraction and 1.1% were diagnosed with OBPP in the register. The groups with and without OBPP were

compared with regard to possible antenatal and intrapartum risk factors, especially those linked to the vacuum extraction procedure. There are five indications for vacuum extraction in the protocol. In Paper II only two indications are presented because the variables “exhausted mother” and “prophylactic vacuum extraction” were combined with the variable “protracted second stage of labour” (see Table II). “Correction of fetal presentation” was excluded in the analysis due to too few deliveries on this indication. All variables in the analyses were obtained from the instrumental procedure protocol (Appendix III) except parity, diabetes, induction of labour and shoulder dystocia, which were extracted from the MBR.

### **Paper III**

Infants with OBPP diagnosed by the paediatrician in the maternity ward were included. Women delivered by caesarean section or vaginally with a breech presentation were included in the calculation of OBPP incidence in the total population, but otherwise excluded. OBPP cases were compared with a randomly selected control group with regard to obstetric management. OBPP and weakness in the arm related to delivery were inclusion criteria; neonates with fractures, luxation of the humeral head and hemiparesis of alternative aetiology were excluded.

A total of 127 infants with OBPP were diagnosed by a paediatrician at birth. Much effort, including active participation by midwives and obstetricians at the seven participating delivery units, was put into the preparation of the questions and parameters in the delivery protocol in order to ensure high quality of the acquired information. The original delivery protocol consisted of 35 questions, (Appendix IV) only 24 of which are presented in the paper, according to the publisher’s instructions. Recorded labour characteristics included duration of the active phase of the first (cervical dilatation  $\geq 4$  cm to 10 cm) and second stages

of labour (cervical dilatation 10 cm to complete delivery of the baby). Protracted delivery was defined as delivery not completed within 12 h (113). The second stage was classified as normal in the absence of documented difficulty in delivering the shoulders and if no ancillary manoeuvres were undertaken.

For evaluation of functional outcome related to OBPP, physiotherapists and paediatricians from the participating rehabilitation units also designed the follow-up examinations in detail. Two paediatricians and three physiotherapists were responsible for information to participating clinicians during the study period.

#### **Paper IV**

The study is based on the same cases as in Paper III. At discharge from hospital the infants diagnosed with OBPP were referred to a physiotherapist for evaluation of functional outcome related to the condition. The physiotherapists examined all infants regularly from the first and second weeks of life to 18 months of age. A protocol including muscle function and range of movement in the injured arm/hand was used at every examination. Anatomic location of the brachial plexus lesion and severity of the OBPP were evaluated during the first month of life. Range of movement was recorded as impaired or not impaired and sensibility in the injured arm was documented based on information from the parents.

#### **Statistical analysis**

##### **Paper I**

Univariate logistic regression was used to estimate the odds ratio (OR) with a 95% confidence interval (CI) for association (antenatal, delivery and fetal variables) between the respective factor and outcome. Multivariate logistic

regression analysis was performed and adjusted odds ratios (AOR) were calculated as the possible risk factors were adjusted for potential confounders.

## **Paper II**

Continuous data were tested for significance with Wilcoxon's rank sum test. Proportions were compared by Fisher's exact test and differences were considered significant at the  $p < 0.05$  level. The association between antenatal, delivery and infant variables and the occurrence of OBPP in neonates was tested with univariate logistic regression to estimate OR with 95% CI. Statistical significance was achieved if the 95% CI did not include 1. In the second analysis multivariable logistic regression analysis (including variables that turned out significant in the univariate analyses) was used to suggest predictor variables which consisted of apparently independent and significant predictors of OBPP. In the next step, logistic regression was combined with spline functions, resulting in a curve of probability of the risk of OBPP related to vacuum extraction duration.

## **Paper III**

The continuous data in Table I were tested for significance with Wilcoxon's rank sum test. The continuous data in Table III were tested with univariate analysis with Fisher's permutation test to estimate associations to OBPP. Statistical significance of differences in VAS scores is presented as mean  $\pm$  standard deviation. All p-values were two-sided and  $p < 0.05$  was considered to indicate statistical significance. Multivariable logistic regression to estimate OR with 95% CI was performed in order to evaluate the independent impact of risk factors on outcome. The independent variables analyzed were downward traction of the head after the third rotation, raising of the head with the intention to deliver the posterior shoulder before the anterior shoulder, attempting to deliver the shoulders by pulling the head straight out of the birth canal, rotation

of the shoulders into the oblique pelvic diameter, extraction by pulling the infant's armpit and delivery of the posterior arm by flexing the elbow and sweeping the forearm over the chest to deliver the hand. In the next step logistic regression was combined with spline functions, resulting in a curve of probability of the risk of OBPP related to VAS score when downward traction was applied. Attributable risk was calculated, i.e. the proportion of cases which could be prevented; OBPP incidence was set at 3.3‰ for this calculation.

### **Power analysis**

When planning the study we assumed that the occurrence of specialized obstetric manoeuvres (Table I, Paper III) was 5% in the control group (c.f. the actual occurrence in Table III, Paper III). Test of statistical significance was based on the hypothesis that there was a difference in the frequency of manual assistance and the use of specialized obstetric manoeuvres between cases and controls. With 70 cases of OBPP and 280 controls a minimum power of 80% and a significance of <0.05% is obtained if the true relative risk is at least 3.9.

### **Paper IV**

The main statistical analysis aimed at comparing maternal, infant and obstetric characteristics in the groups with and without residual OBPP at 18 months of age. The association between nerve root segment involvement and group was analyzed with Chi-square analysis. Continuous data were expressed as mean  $\pm$  standard deviation and analyzed with the Mann-Whitney U test. Categorical data were presented as numbers (%) and analyzed with Fisher's exact test. The Kruskal-Wallis test ( $p < 0.001$ ) with Dunn's Multiple Comparison Test (\* $p < 0.05$ ; \*\* $p < 0.01$ ) was used to evaluate the relationship between degrees of force used in downward traction of the head, ranked on a 100-point VAS, correlated to the number of affected nerve roots and to the severity of OBPP in children with

remaining motor disability, compared to those who had recovered from OBPP at age 18 months.

### **Methodological considerations**

The MBR has compiled antenatal, intrapartum, neonatal and infant data on all births in Sweden since 1973. It offers a unique possibility to obtain reliable prevalence figures regarding obstetric and neonatal outcome. The register has been evaluated three times, in 1976, 1988 and 2001. Experiences from the last quality control study indicate that there is a trend toward an increasing percentage of missing cases, perhaps related to use of computerized medical records. It is important to understand that errors occur and to know how frequent they are. One central source of errors concerns the validity of diagnoses. The most serious data loss is that related to infant diagnoses. Missing data will obviously affect all estimates of prevalence, but will usually have little impact on risk estimates if the lack of information is random (114, 115). A retrospective study on OBPP does not yield the same information as data from a prospective trial regarding uniformity and evidence of association to OBPP. The prospective study mode was preferable in studies III and IV as key factors related to manual assistance are often lacking in retrospective reports.

### **Ethical approval**

The studies in this thesis (Paper III, IV) were approved by the Ethics Committee at Göteborg and Malmö /Lund University Dnr: L 491-98.

## **SUMMARY OF RESULTS**

### **Paper I**

The incidence of OBPP in Sweden increased significantly during the study period from 0.17% in 1987 to 0.27% in 1997 but there were no regional differences in the incidence rate. Shoulder dystocia and infant birth weight of  $\geq 4500$ g are the strongest risk factors for OBPP. A higher frequency of infants with birth weight  $>4000$  g led to more cases of OBPP but the relationship with the increase in risk is weak. The mean birth weight was 3483 g (SD 586) in 1987 vs. 3525 g (SD 595) in 1997. This result was a significant rise but the mean birth weight increased only by 4.5 g annually in this period. The difference in the percentages of OBPP cases related to birth weight between 1987 and 1997 was calculated. Newborns with a birth weight below 4000 g had a risk for OBPP of 0.08% vs. 0.65% in newborns with a birthweight  $\geq 4000$  g. A regression analysis showed a covariation between a higher frequency of newborns with a birthweight over 4000 g and a raised frequency of OBPP. An annual rise in the frequency of newborns  $>4000$  gram by 1% would cause a rise in the incidence of OBPP by 0.03. In 1987 17.2% of infants had a birth weight  $>4000$  g vs. 19.2% of children in 1997, a rise by 2% (19.2 minus 17.2%). The corresponding rise in frequency of OBPP would be 0.06% (2% \* 0.03). Caesarean section was associated with a decreased risk of OBPP.

### **Paper II**

A total of 67.1 % of all neonates delivered by vacuum extraction during the study period were included; 11% contracted OBPP. The group of excluded mothers/infants (32.9%) was comparable to the OBPP group with regard to maternal and gestational age, sex, and birth weight. More nulliparous than parous women were delivered by vacuum extraction but the relative risk of OBPP was higher for parous women. Among the babies with OBPP delivered

by vacuum extraction, 74.5% weighed <4500 g. The strongest risk factors for OBPP were shoulder dystocia, the application of fundal pressure and long vacuum extraction duration.

### **Paper III**

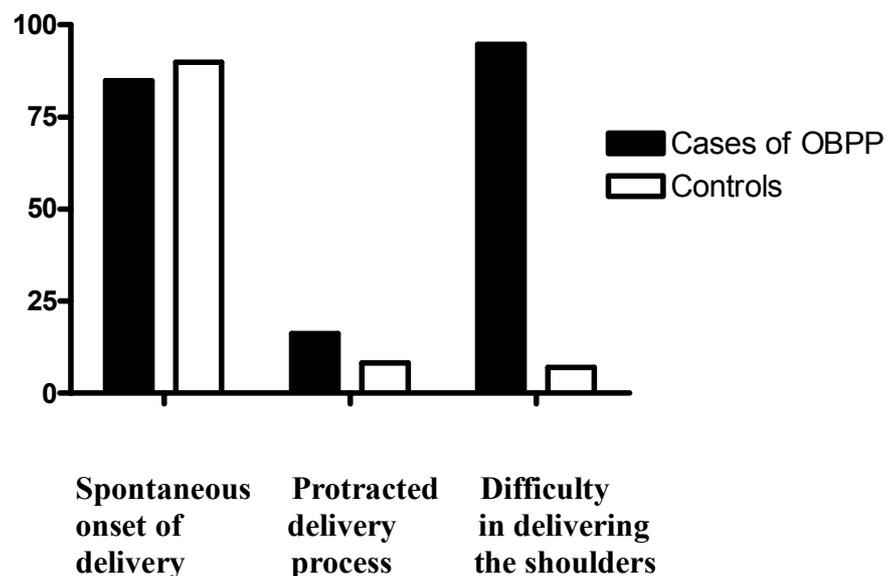
The incidence of OBPP in the population delivered vaginally in the cephalic presentation was 3.3‰. In the potential control group of women with recorded vaginal delivery in the cephalic presentation, 99.3% of the protocols were completed; only 223 of 31 828 protocols were lost to follow-up. The group of 15 infants excluded from the study did not differ with regard to intrapartum and neonatal characteristics, compared to the included 112 cases. None of the 15 excluded infants was registered in the rehabilitation register or clinics when an enquiry was made in 2004. We thus find it unlikely that any of the excluded children had persistent OBPP.

In the OBPP group, ancillary manoeuvres indicating difficulty in delivering the infant during the second stage of labour were more frequent than in controls. Downward traction of the head after the third rotation was strongly associated with OBPP and the risk increased significantly with the force applied. Most (60.7%) of the babies with OBPP had an actual birth weight <4500 g. The majority of infants with OBPP at birth exhibited complete recovery at 18 months of age. A residual functional deficit was documented in 18 infants, the delivery of all of whom had been forcefully assisted by downward traction. Since downward traction applied to the head was a very strong risk factor for OBPP, we calculated the attributable risk as a percentage of the incidence (3.3‰) of OBPP in the population of cephalic-presentation vaginal deliveries in western Sweden between 1999 and 2001. The attributable percentage decrease in the rate of OBPP associated with avoidance of downward traction of the head was 70.4%. Calculation of attributable risk does not require the existence of a cause-

effect relationship. When a contributory association exists (downward traction), attributable risk percentage represents the frequency of damage that may potentially be eliminated if the effect of that risk factor can be completely removed.

Most deliveries started with spontaneous contractions and/or spontaneous rupture of membranes in both study groups (Figure 2). The delivery processes in the OBPP group were somewhat more protracted and completed with great difficulty in nearly all cases (Figure 2).

**Figure 2** There was no difference in the percentage of deliveries that started spontaneously and the frequency of protracted delivery did not differ significantly between OBPP and controls. However, in most OBPP cases the second stage of labour was complicated with difficulties in delivering the shoulders, fundal pressure, numerous bearing-down contractions and forceful downward traction. Five or more manual manoeuvres had often been applied to resolve the shoulder dystocia. Values are given as %.



## **Paper IV**

There were 112 diagnosed cases of OBPP and follow-up to the age of 18 months was possible in 98 children. Among the 14 drop-outs, ten parents have been contacted, reporting that their children have improved. The majority of the included children recovered and functional deficits remained in 18 cases, corresponding to an incidence rate of 0.57‰ at the end of the follow-up period. Manual assistance in the deliveries resulting in permanent OBPP differed from management in cases that had recovered at 18 months of age. Births resulting in persistent motor impairment were all characterized by substantial difficulties in delivering the shoulders. Downward traction of the head had been applied more often, with greater force, in the group with persistent damage, as had external fundal pressure and “panic” actions aimed at completing delivery. The risk factors that have previously been shown to predict OBPP, such as shoulder girdle diameter, birth weight and large for date, were not statistically different in the two groups. Furthermore, parity and body mass index were only weakly associated with persistent OBPP. There was a correlation between the force used in downward traction and the number of affected nerve roots (Figure 3, Paper IV). Most infants that recovered during the 18 months had isolated lesions at the C5-C6 level, whereas the C6-C7 and/or C7-Th1 levels were significantly more often affected in cases with persistent deficits (Tables III and IV, Paper IV).

## **DISCUSSION**

We have confirmed that well known risk factors such as birth weight and shoulder dystocia are important risk factors for OBPP. Furthermore, obstetric procedures, especially forceful downward traction of the head, conferred additional risk of OBPP at birth as well as at follow-up at age 18 months. Our data suggest that forceful downward traction of the head should be avoided and indicate that other measures should be taken to liberate the impacted shoulder, e.g. maximal flexion of the maternal hips, rotation of the shoulders and/or extraction of the posterior arm.

Previous retrospective studies (74, 109, 111, 112) regarding manoeuvres used to resolve shoulder dystocia are discordant concerning the association between manual assistance and persistent OBPP and thus do not agree with our data (Paper III). In previous reports on OBPP (75, 107) it has been stated that direct manipulation manoeuvres used to resolve shoulder dystocia are not associated with an increased rate of OBPP but that more research is needed. McFarland et al (108) found that both type and number of manoeuvre increased the incidence of OBPP; Nocon et al (95), on the other hand, made no similar finding in their study. Since most studies are retrospective and lack detailed documentation of manual assistance in the second stage of labour, it is impossible to use them as a basis on which to evaluate critical second-stage events for avoiding OBPP.

During the last decades numerous data has been published on risk factors for OBPP. Nevertheless, the incidence has increased (16) rather than decreased in Sweden (10, 11, Paper 1, III). Researchers have attempted to identify risk factors (66, 70, 75, 76). The different populations and methods used to diagnose OBPP (Papers I - IV) affect the reported incidence. Some infants with OBPP might have been missed in reports to the MBR, thus creating discrepancies in

incidence in retrospective studies. Indeed, if the diagnosis is initially underreported (114, 115) and more cases are reported correctly over time, the incidence appears to increase despite the true occurrence remaining the same.

Prevention strategies often focus on attempting to prevent shoulder dystocia (75). Since large infants are at disproportionately high risk of this complication (9, 67), prevention strategies include antenatal detection of accelerated fetal growth. Should it be possible to identify women and infants at risk, there are two choices, i.e. planned caesarean section or trial of labour to deliver the baby before it becomes even bigger. Poor sensitivity and specificity in identifying the large fetus antenatally is one problem in this context. Since repeated analyses have shown that an excessive number of caesarean sections would be required to prevent just one case of permanent OBPP (100), the alternative is to plan for vaginal delivery with increased awareness that manual manoeuvre may be needed.

The present studies show that several risk factors related to OBPP are associated with manual assistance during birth. In the logistic regression analysis, shoulder dystocia, breech delivery, birth weight  $\geq 4000$  g, operative vaginal delivery and its management, diabetes mellitus, labour abnormalities, epidural anaesthesia, use of fundal pressure, downward traction of the head after the third rotation and rotation of the shoulders into the oblique pelvic diameter were risk factors. Most of these factors, except birth weight and maternal diabetes mellitus, can in fact be influenced by the midwife/obstetrician during delivery.

It is interesting to note that the factors traditionally held to constitute a major risk of OBPP, such as birth weight, did not differ statistically in the groups with persistent and transient OBPP. This might be interpreted as meaning that the

actual obstetric manoeuvres are more critical determinants of outcome than the size of the infant (Paper IV).

Many of the known risk factors interact in the last phase of delivery and shoulder dystocia is usually difficult to predict. The association between a prolonged second stage and shoulder dystocia has been reported in retrospective studies. However, in our prospective study (Figure 2, Paper III), spontaneous onset of delivery and protracted duration were not associated with OBPP. The risk did, however, increase in proportion to the duration of vacuum extraction (Figure 1, Paper II). It is relevant to investigate which factor in the vacuum application procedure is related to a significant risk of subsequent shoulder dystocia. The mechanisms involved have not been fully analysed but several explanations are possible (61, 104). The fetal head is normally in a position of flexion and the shoulders are adducted in the birth canal. As the instrument is placed on the vertex and traction begun, the vertex is pulled axially and the shoulders tend to abduct (104), increasing the likelihood of shoulder entrapment beneath the maternal pubic symphysis. On the other hand, prolonged extraction per se may not be the cause of OBPP. Alternatively, cephalopelvic disproportion or insufficient contractions cause prolonged labour, a higher risk of instrumental delivery, shoulder dystocia and OBPP. This explanation is supported by Beall et al. (110), who demonstrated that the duration of the second stage was associated with shoulder dystocia but that there was no association with operative vaginal delivery.

Complete OBPP (C5-Th1) involves the entire arm with impaired shoulder function; sometimes the arm is positioned in adduction with a flexed elbow or may hang loosely without motor control. The impact of a complete OBPP on sensory function and bimanual activity is severe. The severity of the condition depends not only on the number of affected nerve roots but also on whether the

spinal nerves have been disrupted or pulled out of the spinal cord. In cases with C5-Th1 lesions, spontaneous recovery cannot be expected (26, 39). We found substantial differences in obstetric management between the groups with and without persistent OBPP. All deliveries resulting in children with remaining OBPP at 18 months were characterized by major difficulties in delivering the shoulders and in the force applied in the downward traction of the head, correlated to the number of nerve roots affected in the brachial plexus (Figure 3, Paper IV).

At the end of the second stage of labour the brachial plexus is at a great risk of damage if downward traction is applied because this procedure will lead to overstretching of the C5- Th1 nerve roots. As presented in Figure 2, Paper III, a considerably increased risk was found in proportion to the VAS score. However, since gentle downward traction is routine and applied in most deliveries, it can be difficult to distinguish between gentle and forceful downward traction.

We have also shown that stretching of nerves in the brachial plexus is related to permanent OBPP and functional deficits at 18 months of age. Most infants with OBPP limited to the C5-C6 segments recovered, whereas those with C5-C7 or C5-Th1 injuries more often suffered from permanent disabilities. Presumably, the number of segments affected correlates with the severity of damage and with reduced potential for spontaneous healing.

There is a lack of documentation in current Swedish delivery records concerning which manoeuvres have been used to resolve shoulder dystocia. A protocol or register might be useful to improve understanding of the mechanisms behind OBPP, facilitate an objective definition of shoulder dystocia and provide insights into which ancillary manoeuvres are effective in releasing the impacted shoulders (34, 104, 105). The fact that only a minority of cases of shoulder

dystocia leading to OBPP in the present study were correctly classified as such indicates that the diagnostic routines are unsatisfactory.

Some investigators (109, 112) state that nothing can be done to improve management of shoulder dystocia. We noted that two studies (61, 75) agree with our finding in Paper III that manual assistance is important for the development of OBPP and that prospective studies are required in order to better understand which obstetric manoeuvres should be used or avoided.

The methodological strengths and weakness in Papers III and IV can be summed up as follows:

#### Strengths

1. Population-based, prospective multicentre study.
2. All relevant professions (midwives, obstetricians, paediatricians and physiotherapists) from the seven participating units took part in planning the study.
3. Use of a standardized delivery protocol, including detailed documentation related to manual assistance during the second stage of labour.
4. Pilot study in which midwives/obstetricians evaluate the delivery protocol before start of main study.
5. Low dropout rate for completing delivery protocols.
6. Standardized follow-up of children until 18 months of age.
7. A large percentage (86.2%, n=25/29) of infants lost to follow-up were contacted (parents) or investigated via rehabilitation clinics more than 3 years after termination of study. All 25 had recovered.
8. OBPP incidence in the population during the study period was checked against MBR data.

## Weaknesses

1. Non-randomized study, making it difficult to explore cause-effect relationships.
2. Scoring the degree of force using the VAS is a subjective method and is not easily translated into clinical practice.
3. Not all infants with OBPP in the population were included in the study and not all infants with OBPP were followed-up.
4. Although the midwives/obstetricians were unaware of which infants were diagnosed with OBPP, they might suspect that a particular infant would develop the condition, which might have led to a certain degree of bias when the delivery protocols were completed after delivery.
5. Only 18 cases had OBPP at 18 months of age, which means that the comparison between cases with and without recovery (Paper IV) suffered from limitations in power.

## **CLINICAL RELEVANCE OF THIS THESIS**

The most important finding, from a clinical point of view, is that all cases of OBPP resulted from complicated deliveries with substantial difficulties in delivering the shoulders, inferring that practical management is critical. The fact that development of both transient and permanent OBPP, as well as the severity of the condition, correlated to forceful downward traction of the head strongly suggests that this procedure should be avoided and be replaced by manoeuvres facilitating release of the shoulders.

The logical next step would be to test this hypothesis in a randomized controlled trial with and without forceful downward traction in situations of shoulder dystocia. However, because of the low occurrence of this more or less catastrophic situation we find such an undertaking unrealistic. Therefore, a reasonable strategy in future clinical practice is the drawing up of new guidelines discouraging the use of this manoeuvre and suggesting other means of assistance (see tentative guidelines in Appendix 1). We will follow up all cases of shoulder dystocia and OBPP at our department during the next five-year period with the aim of halting the increasing frequency of OBPP.

## CONCLUSIONS

Shoulder dystocia and a birth weight of 4500 g or more are the strongest risk factors for OBPP.

Shoulder dystocia in the setting of vacuum extraction is a prominent risk factor for OBPP. The risk increases with the duration of the vacuum extraction.

All infants suffering from OBPP were born in cephalic-presentation vaginal deliveries, more than 97% of which were complicated by shoulder dystocia.

Infants contracting OBPP are delivered with great difficulty; manual assistance including several manoeuvres was required before delivery could be completed.

Forceful downward traction of the head after the third rotation was the strongest risk factor for OBPP related to manual assistance.

Downward traction of the head had been applied more often and with greater force in the group with persistent damage and there was a significant correlation between the force used and the number of affected nerve roots.

There is no universal definition of shoulder dystocia.

Shoulder dystocia is underreported, according to the maternal records.

We propose a prevention strategy for OBPP in cephalic presentation and suggest drawing up of guidelines for shoulder dystocia in which avoidance of forceful downward traction, fundal pressure and long vacuum extraction duration are recommended.

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