

Neonatal Brachial Plexus Palsy: Risk Factors and Its Prognostic Value

Sofia Ataíde¹, Filipe Bettencourt², Ana Cadete¹, Leonor Prates¹

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Abstract

Introduction: Neonatal brachial plexus palsy affects 0.7 to 5.8 per 1,000 newborns and is characterised by upper limb paresis detected in the immediate neonatal period. Shoulder dystocia, instrumental delivery and foetal macrosomia are well-known risk factors. Most neonatal brachial plexus palsy evolve favourably, while 3%-27% of newborns have sequelae.

Methods: A retrospective cross-sectional study was conducted to characterise neonatal brachial plexus palsy in the newborn population of a hospital with differentiated perinatal support and to assess the relationship between the risk factors and lesion prognosis. The authors reviewed the newborn medical records referred to the physical medicine and rehabilitation clinic between January 2006 and December 2016.

Results: During the study period, 137 cases of neonatal brachial plexus palsy were identified in 36,833 births, which translate into an incidence of 3.7/1,000 live births. Foetal macrosomia was found in 41% and shoulder dystocia in 40%. According to the Narakas classification, 58% were included in group I, 30% in group II, 9% in group III and 3% in group IV. The majority of patients were discharged without sequelae. Newborns with group II, III and IV lesions as well as macrosomic newborns were more likely to develop sequelae ($p < 0.05$). Shoulder dystocia and operative delivery did not present a statistically significant relationship with the prognosis of the lesion.

Discussion: The incidence of neonatal brachial plexus palsy in this population was similar to is described in other series. The relationship between macrosomia and neonatal brachial plexus palsy with sequelae found may be of importance in the attempt to prevent this lesion.

Keywords: Birth Injuries; Brachial Plexus/injuries; Brachial Plexus Neuropathies/aetiology; Infant, Newborn; Portugal; Prognosis; Risk Factors

Introduction

Neonatal brachial plexus palsy (NBPP) is characterised by a decrease or absence of spontaneous movements of the affected upper limb, detected in the first hours after birth.¹ It is a clinical diagnosis characterised by a decrease in the active range of motion, and the preservation of the passive range of motion.² Since at this age it is often difficult to evaluate the active range of motion, the Moro reflex can be tested, which will be asymmetric in these cases.³

The aetiology of NBPP remains controversial and is still the subject of investigation. Classically, it is argued that the injury is caused by the stretching of the C5 to T1 nerve roots, which constitute the brachial plexus, due to the traction exerted between the cervical spine and the shoulder during delivery. It is also known that caesarean section has a protective role, but does not exclude NBPP.^{4,5} Consequently, other pathophysiological mechanisms have been described, such as poor intrauterine adaptation or uterine contractions exerted on the posterior shoulder against the maternal sacral promontory during the first stage of labour.^{1,6} It is, therefore, an injury that is impossible to anticipate, although several risk factors and their relationship with the injury have already been established.^{5,7}

The identified risk factors may be related to ⁵⁻⁹:

- Delivery, namely shoulder dystocia (SD), operative vaginal delivery, prolonged second stage of labour;
- Maternal factors, such as gestational diabetes, older age, obesity, short stature;
- The foetus, with macrosomia and malposition being more frequent.

Some newborns with NBPP have one or more risk factors while, in others, no risk factor is identified.

NBPP has an incidence ranging from 0.7 to 5.8 per 1,000 live births.^{1,4,5,9-14} It affects homogeneously both sexes and the right upper limb is the most often affected.³

There are several classifications of NBPP, but the most commonly used classification is the one proposed by Narakas.¹⁶ This classification categorises NBPP into four

1. Physical and Rehabilitation Department, Hospital Prof. Doutor Fernando da Fonseca, Amadora, Portugal

2. Physical and Rehabilitation Department, CUF Alvalade Clínica, Lisbon, Portugal

Corresponding Author

Sofia Ataíde

sofiaataidemfr@gmail.com

Hospital Prof. Doutor Fernando Fonseca EPE

IC19, 2720-276 Amadora, Portugal

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groups according to the clinical presentation and is also indicative of prognosis (Table 1).

The severity of the injury and its prognosis depend on the affected nerve roots and the degree of stretching to which they were subjected. In preganglionic lesions (i.e. proximal to the dorsal root ganglion) the prognosis is poor, since these lesions cannot be surgically corrected. In postganglionic lesions (i.e. distal to the dorsal root ganglion), three types of lesions may occur according to the Seddon/Sunderland classification.^{3,17} In cases of neuropraxia (nerve-stretching myelin injury with intact axons) and axonotmesis (axonal rupture), re-innervation and spontaneous clinical recovery may be expected, whereas in neurotmesis (complete nerve section), spontaneous clinical recovery is impossible; however, surgical repair is possible.^{3,17} The final prognosis is directly correlated to the type of neurological injury, which is clinically impossible to assess.^{3,13} It is known, however, that early clinical recovery is related to a favourable outcome.² Fortunately, most NBPP cases do not have any sequelae, with reported rates ranging from 73% to 97%.¹⁵

Our study aims to characterise NBPP and its prognosis in the newborn population of a hospital with differentiated perinatal support in the last 11 years and to compare risk factors, namely macrosomia, operative vaginal delivery and shoulder dystocia, with the severity of the injury.

Methods

A retrospective casuistic analysis of the population of newborns diagnosed with NBPP and referred for Physical and Rehabilitation Medicine (PRM) evaluation, born between 1 January 2006 and 31 December 2016, was conducted. The clinical records of the newborns were analysed and the following variables were studied: sex, birth weight, route of delivery, existence of SD, classification of the injury and existence of sequelae. The inclusion criteria included the diagnosis of NBPP performed by a PRM consultant in the first month of life

and a minimum follow-up of one year.

The diagnosis of macrosomia was established with a birth weight higher than 4,000 g. Shoulder dystocia was defined as the foetal shoulders becoming impacted during delivery with the need for specific manoeuvres to enable delivery. The Narakas classification was used to characterise the lesions based on the evaluation made at the first PRM visit.

The clinical evolution of the lesions was only studied between 2009 and 2016 as there was no record of the evolution of lesions in previous years. Thus, the statistical analysis was performed in a sample of the study population. In this sample of newborns, shoulder, elbow and hand range of motion deficits, muscle strength in the territories of the affected roots, muscle and tendon retractions and glenohumeral dysplasia/luxation, and winged scapula were considered as sequelae.

Statistical analysis including descriptive statistics (absolute and relative frequencies, mean and standard deviations) and inferential statistics was performed using Statistical Package for Social Sciences (SPSS) version 24.0. To identify the risk factors related to the prognosis of the injury, a multivariate stepwise logistic regression analysis was used. The significance threshold was set at $p \leq 0.05$.

Results

In the 11-year period, there were a total of 137 NBPP cases among 36,833 live births, which translates into an incidence of 3.7 per 1,000 newborns.

The distribution by gender was similar and the right upper limb was most frequently affected (68%). The vast majority of newborns (76%) had a birth weight over 3.5 kg and 41% had macrosomia.

The SD occurred in 40% of the cases. 32% had an operative vaginal delivery, and there was an NBPP case in a caesarean delivery.

According to the Narakas classification, 80 (58%) were classified in group I, 41 (30%) in group II, 12 (9%) in group III and 4 (3%) in group IV.

Table 1. Narakas classification of neonatal brachial plexus palsy

Group	Roots injured	Clinical features	Prognosis
I	C5, C6	Shoulder adduction and internal rotation, extended elbow	Spontaneous recovery without sequelae in over 80% of cases
II	C5, C6, C7	Shoulder adduction and internal rotation, extended elbow, flexed wrist and fingers	Spontaneous recovery without sequelae in about 60% of cases
III	C5, C6, C7, C8, T1	Flaccid upper limb with complete paralysis	About 30% of cases recover without sequelae
IV	C5, C6, C7, C8, T1 and stellate ganglion of the sympathetic nervous system	Flaccid upper limb and ipsilateral Claude-Bernard-Horner syndrome (pupil miosis and palpebral ptosis)	Poor prognosis; severe sequelae

Table 2. Injury distribution according to their outcome

Narakas classification	Without sequelae	With sequelae
Group I (n = 55)	54 (98.2%)	1 (1.8%)
Group II (n = 19)	10 (52.6%)	9 (47.4%)
Group III (n = 9)	1 (11.1%)	8 (88.9%)
Group IV (n = 3)	-	3 (100%)
TOTAL (n = 86)	65 (75.6%)	21 (24.4%)

Regarding the evolution of the lesion, in the study period (between 2009 and 2016), there were 89 NBPP cases, of which three cases were excluded as they did not fulfil the minimum follow-up time. Of the 86 lesions evaluated, 34.9% (n = 30) occurred in macrosomic newborns and 40.7% (n = 35) had SD. According to severity, 55 (64%) were classified in group I, 19 (22%) in group II, 9 (10.5%) in group III and 3 (3.5%) in group IV.

As shown in Table 2, the vast majority of group I lesions evolved without sequelae (98.2%), while the rest had a poorer outcome. Overall, 75.6% of the NBPP cases evolved without sequelae and 24.4% with sequelae.

Based on the statistical analysis, operative vaginal delivery and SD were not associated to the prognosis of the injury ($p > 0.05$). On the other hand, macrosomia and group II, III and IV lesions are significantly associated with sequelae (Table 3).

Discussion

The incidence of NBPP in our study is within the values reported in the literature, although slightly higher than most studies. The variation in incidence in the literature can be attributed to the geographical differences in obstetric care and birth weight,¹⁰ to studies being conducted on selected populations rather than on national registries⁵ as well as inadequate diagnosis of the injury or to its early recovery which may lead to an underestimation. Our cohort is based in a maternity hospital in which the PRM evaluation is performed within the first 24 to 48 hours of life leading to a complete register of all newborns with NBPP, including mild cases. Moreover, it is a hospital that receives a relevant percentage of immigrants, which often does not have any obstetric care,

with a probable increase in the prevalence of risk factors such as gestational diabetes and macrosomia, which may explain the relatively higher number of NBPP cases in this centre. In Portugal, there is no national registry of NBPP, and only two case series of this condition have been published, reporting an incidence varying between 0.77 and 0.98 per 1,000 newborns.^{11,12}

As described in other studies, this study found no gender predominance and that the right upper limb was most often affected.

This study also found a similar rate of sequelae (24.4%) to what is described in the literature. Many studies do not report sequelae, but rather whether the lesion was temporary (lasting less than 12 months in most studies) or permanent.^{1,4,9}

Complete brachial plexus palsy and the presence of Claude-Bernard-Horner syndrome are the main indicators of poor prognosis. Regarding the lesion of the phrenic and long thoracic nerves, whether or not they are signs of unfavourable prognosis has been recently a matter of discussion.³ The main indicator of the speed of recovery of the lesion is the muscular strength of the biceps brachii after the first six months of life.^{3,10,18} Children recovering elbow and shoulder muscle strength against gravity by 3 months are expected to have a full recovery.

In children in who early neurological recovery does not occur, glenohumeral dysplasia is observed with a progressive instability of the shoulder joint and frequent progression with posterior displacement of the humeral head.¹⁰ Muscular denervation leads to muscle contractures, usually with an internal rotation of the shoulder and flexion of the elbow.¹³ All of these sequelae contribute to an important functional limitation of the upper limb.

Regarding risk factors, most studies identify macrosomia, SD and operative delivery with higher risk of NBPP,^{4,5,12} but do not associate them with the prognosis of the injury.^{1,4,9,11} Our study could not associate them with higher risk of injury (due to an absence of control group, i.e. without NBPP). However, when the risk factors present in the neonates of our sample were studied

Table 3. Association between several studied factors and neonatal brachial plexus palsy with sequelae

Factor	Odds ratio	95% confidence interval		p
		Lower endpoint	Upper endpoint	
Operative delivery	-	-	-	0.820
Shoulder dystocia	-	-	-	0.712
Macrosomia	14.402	1.406	147.563	0.025
Group II injury	130.495	8.352	2038.897	0.001
Group III and IV injury	2268.05	69.211	74232.996	0.001

as potential exposures for the prognosis of the injury, macrosomia and lesion group are associated with NBPP with sequelae in this sample. This indicates that macrosomic newborns and those classified at birth as group II, III and IV lesions are more likely to have sequelae.

The association of SD with the occurrence of NBPP is also well established,^{9,14,15} but unrelated to the evolution of the disease with or without sequelae,^{1,4,9,11} which is similar to our findings. Nevertheless, it should be noted that the percentage of deliveries with SD in our sample is considerable, comparable to another study,⁴ with 54% of SD in a sample that is similar to ours, but other authors report an SD rate of 17%⁵ in a sample of 17,334 NBPP. It is important to note that the definition of shoulder dystocia varies in the literature, with some authors considering it to be the impaction of the shoulders after the first contraction following the delivery of the foetal head, while others consider the impaction of the shoulders, even after the employment of extraction maneuvers.¹ Furthermore, since it is a retrospective study, one of its limitations is related to the fact that the information about this risk factor collected from the medical records of the newborns, which may have underestimated its incidence, due to underreporting. However, as the majority of newborns with NBPP do not present SD, the thesis that other intrauterine phenomena contribute to this injury is, therefore, supported. Furthermore, NBPP still occurs in newborns delivered by caesarean section,⁷ as found in one case of our sample. Operative delivery is not associated with the prognosis of the lesion in our sample, as reported in the literature.^{5,7,11}

Knowing that NBPP is impossible to anticipate, its prevention is based in controlling major risk factors, namely gestational diabetes and macrosomia. Compared to other studies,^{1,4,5,12} we found that our sample has a high number of macrosomic newborns, which may explain the positive association between the macrosomia and progression of the lesion with sequelae. On the other hand, this association may also be a factor to be considered in centres with a population without regular pregnancy care, for any reason and, therefore, at a greater risk of macrosomia.

The initial classification of the injury is also important to establish the prognosis. Our work showed that the vast majority of group I lesions are self-limiting lesions, unlike the rest, including those of group II, which are more likely to progress with sequelae.

The early identification of NBPP, referral and provision of rehabilitation programmes, are important to minimise these sequelae. The main aims of the rehabilitation treatment are to maintain ranges of motion in order

to avoid contractures, stimulate the contraction of the paretic muscles, prevent the onset of compensatory movements and, more importantly, promote adequate cortical motor patterns in order to integrate the upper limb into the corporal scheme.^{2,13,18} Whenever NBPP is suspected, the child should have an early evaluation by a PRM physician, with the aim of the timely introduction of a rehabilitation programme, of which teaching parents the proper handling and positioning of the limb is a fundamental part.^{3,18}

Surgical treatment is a therapy option for cases with no neurological recovery. The great majority of the authors consider as surgical criteria those children with a total lesion with or without Claude-Bernard-Horner syndrome and the absence of force against gravity in the biceps brachii by 3 months.^{10,13} The ideal timing for surgery is not consensual, varying between 3 and 9 months.³

The knowledge of the natural history of the disease and its sequelae allowed for the establishment of therapy and follow-up criteria. The timely identification of the lesion and its follow-up by a multidisciplinary team are the best strategies for the NBPP patient.

WHAT THIS STUDY ADDS

- The results of this paper are similar to those published in the literature, except for the statistically significant association between macrosomia and NBPP with sequelae.
- This may warrant special clinical attention in the evaluation, follow-up and therapy of newborns with NBPP and birth weight above 4,000 g.
- PRM has a primary role in maximising the functionality and consequent autonomy of these children.

Conflicts of Interest

The authors declare that there were no conflicts of interest in conducting this work.

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Protection of human and animal subjects

The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of data

The authors declare that they have followed the protocols of their work centre on the publication of patient data.

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Lesão Neonatal do Plexo Braquial: Fatores de Risco e Seu Valor Prognóstico

Resumo:

Introdução: A lesão neonatal do plexo braquial afeta entre 0,7 a 5,8 em cada 1000 recém-nascidos e caracteriza-se por paresia do membro superior detetada no período neonatal imediato. A distócia de ombros, o parto instrumentado e a macrosomia fetal são fatores de risco conhecidos. A maioria evolui favoravelmente, enquanto que 3%-27% dos recém-nascidos fica com sequelas.

Métodos: Estudo transversal retrospectivo para caracterizar a lesão neonatal do plexo braquial na população de recém-nascidos de um hospital com maternidade de apoio perinatal diferenciado e relacionar os fatores de risco com o prognóstico da lesão. Foram revistos os processos clínicos dos recém-nascidos orientados para a consulta de medicina física e de reabilitação entre janeiro de 2006 e dezembro de 2016.

Resultados: Identificaram-se 137 casos de lesão neonatal do plexo braquial em 36 833 nascimentos, o que se traduz numa incidência de 3,7/1000 nados-vivos. Verificou-se macrosso-

mia fetal em 41% e distócia de ombros em 40%. De acordo com a classificação de Narakas, 58% estavam incluídos no grupo I, 30% no grupo II, 9% no grupo III e 3% no grupo IV. A maioria teve alta da consulta sem sequelas. Conclui-se que os recém-nascidos com lesão do tipo II, III e IV tinham maior probabilidade de evoluir com sequelas, bem como os recém-nascidos macrosômicos ($p < 0,05$). A distócia de ombros e o parto instrumentado não apresentaram relação estatisticamente significativa com o prognóstico da lesão.

Discussão: A incidência da lesão neonatal do plexo braquial nesta população foi semelhante à descrita noutras séries. A relação entre macrosomia e lesão neonatal do plexo braquial com sequelas pode ser um dado importante na tentativa de prevenção da lesão.

Palavras-Chave: Fatores de Risco; Neuropatias do Plexo Braquial/etiologia; Plexo Braquial/lesões; Portugal; Prognóstico; Recém-Nascido; Traumatismos do Nascimento