CASE REPORT

Birth brachial plexus palsy: a race against time

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Accepted 20 June 2016

SUMMARY

A 5-year-old child presented to us with weakness of the left upper limb since birth. With the given history of obstetric trauma and limb examination, a diagnosis of birth brachial plexus palsy was made. Brachial plexus exploration along with microsurgery was performed at the same time which included extrinsic neurolysis of the roots and trunks and nerve transfer for better shoulder external rotation and elbow flexion. Both the movements were severely restricted previously due to co-contractions with the shoulder internal rotators and triceps. The problem of birth brachial plexus palsy is proving to be a global health burden both in developed countries and in developing countries such as India. The lack of awareness among the general public and primary healthcare providers and inadequate orthopaedic and neurosurgeons trained to treat the condition have worsened the prognosis. This case lays stress on the delayed complications in birth brachial palsy and its effective management.

CASE PRESENTATION

A 5-year-old firstborn male child was brought by his parents with symptoms of multiple deformities of the left upper limb since birth. The mother had an uneventful antenatal history. The child was born at full term by assisted vaginal delivery using forceps in view of shoulder dystocia during labour. The birth weight of the child was 3.9 kg and he did not cry immediately after birth, following which he was intubated and referred to a higher centre. He was admitted to a neonatal intensive care unit for 6 days and resuscitated, following which he was discharged. Parents noticed the inability of the child to move his left upper limb at around 1 month but did not seek any treatment for 1 year. They then took the child to a family physician who advised physiotherapy which was continued for 1 month and then stopped. He was later taken to a traditional complementary medicine practitioner who treated the child by immobilisation with splint and weights which did not result in improvement of his function. At around 2 years of age, they consulted a paediatrician who suggested seeking an opinion for surgical correction of the deformity for which the parents were reluctant due to financial constraints.

Social and personal history of the child

The child was doing well and was attending an Anganwadi school daily in spite of his disability. He had good social interaction with friends and played games and sport which involved minimal involvement of the affected hand. His general growth and development was appropriate for his age.

Examination findings

On examination, the child was cooperative and obeyed our commands with average intelligence. His left shoulder was in an attitude of 10° abduction, elbow flexed at 30°, wrist extended at 50°, and metacarpophalangeal joints flexed at 70° with thumb in palm deformity. The modified Mallet Scores were grade IV for shoulder abduction, grade III for external rotation, and grade II for hand to spine, hand to neck and internal rotation (figures 1 and 2, video 1). The Gilbert and Raimondi Scale for elbow function revealed a score of 2 for flexion, 1 for extension and -1 for extension deficit. The Raimondi scale for hand function score was 2. Except for finger extension, active movement could be demonstrated in all the joints. Except for the shoulder joint, the excursion of other joints was very poor. Perhaps the most disabling feature was that of a hallmark severe co-contraction between the muscles of the shoulder, elbow, forearm and hand. For every movement of the hand, there was simultaneous movement of the elbow and shoulder and vice versa. The classical trumpet sign was present. On giving instruction to take an object towards the mouth the child could neither complete flexion nor extension of the elbow in spite of presence of fairly good palpable contraction in the arm as both the biceps and triceps were firing simultaneously. In effect, there was no useful function in the left upper limb. The only use of the limb was in holding small objects with poor grasp. A diagnosis of birth brachial plexus palsy (BBPP) of the left upper limb was made and the parents were counselled regarding the need for microsurgery to improve the functional status of the child's left upper limb. The parents had recently come to know about the health insurance schemes of the state government and hence agreed for the surgery.

Operative procedure

Brachial plexus exploration and microsurgery was done at the same time. On exploration of the brachial plexus, extensive fibrosis was observed involving all the roots and trunks. Extrinsic neurolysis of the roots and trunks were done. It was decided to do a nerve transfer to get a better shoulder external rotation and elbow flexion which was severely restricted due to co-contractions of the shoulder internal rotators and triceps. The spinal accessory nerve was transferred to the suprascapular nerve; four intercostal nerves were harvested from the third to sixth intercostal space and transferred to the musculocutaneous nerve. Since there was no extension at the digits, it was decided to do tendon transfer as the nerve transfer option was not available for

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To cite: Patra S, Narayana Kurup JK, Acharya AM, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-215996

Figure 1 Note the child's inability to complete shoulder abduction and elbow extension.

finger extension. The extensor carpi radialis longus tendon was transferred to the extensor digitorum communis and extensor pollicis longus. The postoperative period was uneventful and the child was discharged on the fifth day and reviewed for hand

Video 1 Shows the incomplete movements of the affected shoulder, elbow and wrist due to the weakness and co-contractions.

therapy after a month. At 4 months postsurgery follow-up, the child was able to open up his hand for grip due to the tendon transfer which was done along with the nerve transfer.

GLOBAL HEALTH PROBLEM LIST

1. Lack of comprehensive global and regional data of the problem.

Figure 2 Note that the child is unable to externally rotate the shoulder and also internal rotate completely.

2. Poor prognosis and outcomes due to delay in presentation and seeking care.
3. Lack of awareness regarding diagnostic criteria for treatment options among the healthcare providers and the public.
4. Social impact and stress on the family due to problems of accessibility and affordability in seeking appropriate healthcare.

GLOBAL HEALTH PROBLEM ANALYSIS

Epidemiology: global and Indian

BBPP, in spite of significant advances in medical healthcare, continues to be a major burden reported worldwide with a frequency not very different from the records published since six decades.¹ Recent trends continue to show an incidence of just over 1 per 1000 births both in developed countries such as the USA and in developing countries such as India.^{2–3} There is sparse literature evidence on the incidence and prevalence of this injury in India. Bhat *et al*³ in their study among 32 637 deliveries over a period of 10 years noted an incidence of 1/1000 live births, which is comparable to other studies done worldwide. However, this may represent just the tip of the iceberg due to the lack of coordinated reporting of cases, lack of BBPP National registries and due to a substantial amount of non-hospital deliveries in rural areas. The incidence of BBPP is estimated to range from 0.4 to 4 per 1000 live births.^{4–6} The incidence in the USA and Canada is 1.5/1000 and 1.24/1000 live births, respectively. There are no global or Indian data available on the severity, long-term effects of disability and specific coordinated healthcare services available for brachial plexus palsy, even though its incidence is equivalent to that of autism and Down's syndrome.

Risk factors

Shoulder dystocia, prolonged or difficult labour, fetal macrosomia, instrumental deliveries, etc, are the most important perinatal risk factors for BBPP. Chauhan *et al*² suggested that concordance of the rates between various countries indicate that the reason for BBPP may not be due to a specific manoeuvre done by the clinicians and that the palsy can occur without difficulties in delivering an impacted shoulder.

Types of BBPP and prognosis

Narakas has classified the severity of BBPP from type I to type IV. Narakas's⁷ type I involves C5–C6 nerve root involvement and is the most common type of BBPP, carrying the most favourable prognosis. Type II is an injury to the C5, C6 and C7 nerve roots and carries a poorer prognosis than type I. Type III is a global palsy involving the C5, C6, C7, C8 and T1 nerve roots. Type IV is the most severe form with global palsy along with Horner syndrome. Neurapraxias recover fully in the first few months of life. Avulsion injuries (preganglionic) in which the nerve roots are completely disconnected from the spinal cord and characterised by Horner syndrome, winging of scapula, phrenic nerve palsy and cannot recover. Approximately 60% of neonatal injuries are mild and spontaneously resolve.⁴ Active Movement Scores grading system is available, which is reliable in infants and guides the treating doctor about surgical planning.^{8–9} Many of the caregivers are not aware of such a scoring system. Early microsurgical intervention is recommended for those with Narakas type IV BBPP. Return of elbow flexion strength is the key factor which determines the need for brachial plexus exploration and nerve reconstruction with most surgeons recommending this procedure when antigravity elbow flexion has not returned by 3–9 months of age.^{10–13}

Delay in presentation and seeking care

Nerve reconstruction procedures are best performed in infancy to allow the greatest potential for recovery of muscle strength. Coroneos *et al*¹⁴ assessed the time of referral of a case of BBPP to a multidisciplinary centre and found that the majority (55–60%) of patients with BBPP identified at birth were not referred for multidisciplinary care. They classified the referred patients on the basis of the month in which referrals were done into first month (good), third month (satisfactory) and beyond 3 months (poor) referral and found 'good' in 28%, 'satisfactory' in 66%, and 'poor' in 34% of the patient referrals. Primary care providers lack the expertise and can underestimate the injury and provide guardians with inaccurate information and education.^{15–17} This results in guardian distress and delayed specialist referral.¹⁵

Lack of regional data and usage of services

The fact that hardly half of the patients have one or more of modifiable risk factors suggests an unknown mechanism which needs further elaboration and research involving multiple centres.^{2–18} Systematic analysis on the natural history of BBPP has shown the fallacy and lack of scientific evidence in the often quoted excellent prognosis of over 90% in such birth injuries.¹⁹ These studies also mention the presence of permanent residual deficits which may be as high as 30% needing microsurgical intervention for prevention in the first year of life.^{5–18–20} Even in developed countries, studies have shown underutilisation of microsurgical services and 'limited access to care' for such conditions where only neonates with private insurance had access to necessary facilities.²¹ In India, the burden is even more in the presence of disparities due to socioeconomic status, rural/urban divide, rising hospital costs compounded by poor emergency services in primary health systems with high out-of-pocket expenditures met by more than three-fourth of the households.^{22–23} In India, three-fourth of healthcare facility and resources are available in urban areas where hardly 27% of the population lives.²⁴

Lack of awareness among the public and healthcare providers

Lack of awareness of healthcare facilities is another problem among the rural poor who do not seek treatment due to cultural, socioeconomic and geographic reasons added on by state's difficulty to integrate primary centres referrals with tertiary centres as seen with our patient. Our patient's parents were in the below poverty level group hailing from a rural background, residing 500 km away from our hospital. The parents were unaware of the government subsidy provided for surgeries related to neurological and congenital deficits for children. Unfortunately, these problems are further aggravated by a lack of information among the medical fraternity about the recent advances in the management of BBPP, particularly among paediatricians and orthopaedic surgeons who mainly deal with this condition in our country. An internal audit among paediatricians and obstetricians in our institution showed that one-third were unaware of the current guidelines for management for BBPP with respect to microsurgical reconstruction (unpublished data). Those who were aware suggested that the reasons for the delay were: reluctance of the parents and lack of knowledge in recent advancement among surgeons who send these children for hand therapy. We regularly come across older children and adults like the one mentioned in our case who might have benefited from early surgery in infancy but were denied the opportunity due to

various reasons including lack of timely referral and inadequate training and skill of the medical personnel.

Impact on family

Having a child with BBPP can affect a family through social, financial and emotional factors. Most often, the impact of the child's disability is underestimated. Focus of care often gets concentrated on treating the child's injury, and attention is diverted from the impact on the family. Louden *et al* studied the impact of BBPP on the family using the Impact on Family Scale (IFS). The IFS is a self-administered four point (strongly agree to strongly disagree) scale consisting of 27 questions which measures a parent's perception of their child's health condition on family life,^{25–27} and has four dimensions: financial burden; familial/social challenges; personal strain of the primary caretaker's and the family's ability to cope with the stress through mutual support.^{26–27} A total of 102 caregivers were included in the study. According to them, the areas which were most affected were strain for the family members, social interactions and economic burden. The income and level of education correlated negatively with the impact on the family. The distance travelled for BBPP treatment services was a major concern for the caregivers and caused a huge economic burden, especially for those who travelled nationally compared with those travelling locally. Mothers of children with BBPP were found to have a lower quality of life than healthy controls and psychological stressors were more for them.^{28–29} Akel *et al*³⁰ reported poorer quality of life scores in children with BBPP compared to their healthy peers. Parental distress and dissatisfaction with communication with the treating doctor on details of diagnosis and treatment information has been reported in BBPP. This situation is even worse among illiterate parents and in rural areas of India.

Healthcare and insurance system in India

In India, healthcare is mainly the responsibility of state governments with assistance from the central government. The total expenditure on public health expenditure as a percentage of gross domestic product in India as per the World Bank data is 4%, but it is far below the global median of 8.7%.³¹ The lack of adequate funding for public health forces people to purchase services from the private sector. The private medical sector is the mainstay of healthcare in India providing service to ~70% of the population in urban and rural areas.³² Most of the expenses for healthcare are paid out of pocket by the patients. As per the World Bank data, only about 25% of the Indian population had some form of health insurance in 2010.³³ However, a study by the Indian government done in 2014 found that the World Bank data was an overestimate and only about 17% of the population had health insurance coverage.³⁴ A universal healthcare system is still not in place in India. This has led to a detrimental effect on the basic standard of living of people, especially in rural areas.

This calls for a multidisciplinary approach to address the lacunae in treatment, education and creating awareness, and providing resources and financial support for these families. Creating proper awareness through clinician-led camps, family programmes and other public teaching modalities may prove to be an effective first step in helping families. Squitieri *et al*,²¹ in their study on adolescents who experienced BBPP, have stressed that such understanding was essential for care of the affected child. Counselling can be provided to the parents to cope with the stressors and anxiety and to explain the child's status to them. Formation of support groups will provide an opportunity

for the children and their caregivers to meet similar people and share their experience and provide mutual support.

Patient's perspective

We belong to a lower middle-class family. This disease has brought an upheaval on me and my family because of its prolonged course. I feel sad that my son could not move his hand like other children. I did not realise it earlier but noticed when he failed to hold objects and play with his hand like his peers. I do not know clearly the reason for my child's disability. Doctors mentioned that it had something to do with the difficult delivery I had. I have taken my son to many hospitals before coming here. Initially, nobody advised surgery for my child and we wasted a lot of precious time. The doctors here have diagnosed and operated on my son. The treating doctor told us that if surgery had been done very early in his life, our son would have got very good results with the movement of his left upper limb. There was no one to guide us properly regarding the seriousness of his problem. We feel guilty now that at the age of 2 years, when another doctor advised surgery for him, we were reluctant due to financial constraints and did not go ahead with the surgery. We had difficulty covering the expenses, but the insurance policy offered by the state government provided some help. I understand that he has to undergo regular physiotherapy exercises and follow-up, which will be challenging because of the lack of local physiotherapy centres and I will have to carry him all the way here for follow-up.

Learning points

- Birth brachial plexus palsy continues to prove a burden for developed and developing countries due to the involvement of multiple risk factors.
- Detailed and clear guidelines regarding diagnostic criteria for surgical intervention versus conservative approach should be made aware to all healthcare providers.
- Measures should be taken to spread awareness among the general public regarding the presentation and outcome of the disease.
- Affordable and easily accessible healthcare should be provided to treat these children to avoid late complications and help them achieve full recovery potential.

Contributors All authors contributed to conception and design of the paper, SP and JKN has done the acquisition of data, drafting the article AA and AKB has done corrections in the final manuscript, and was involved in the final approval of the version to be published.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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