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Atypical Erb's Palsy Presentation in a Premature Newborn: A Case Report

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Abstract

Objectives: The aim of this study is two-fold: to present an atypical Erb's palsy presentation in a premature newborn and to report his recovery through conservative care after three months without spontaneous resolution.

Case presentation: A 3-month-old male presented at the clinic with right side Erb's palsy following a cesarean delivery at 33 weeks of gestation from a mother with a bicornuate uterus. The mother and the baby had no other risk factors commonly associated with this condition.

Intervention and outcome: Treatment consisting of light mobilization and soft tissue techniques was performed twice a week for 9 weeks for a total of 18 visits along with postural advice. After 18 visits, there was no mobility restriction of the neck, and bilateral abduction and flexion of arms was possible. Biceps and grabbing reflexes were improved. The baby was able to lie in the prone position with the head upright and arms at 90° flexion.

Conclusion: We are reporting a rare case of Erb's palsy in a premature baby, which is not documented in the scientific literature as a common risk factor. Before surgery is considered, chiropractic care should be considered as a conservative and non-invasive approach for clinical management of Erb's palsy in cases without spontaneous recovery.

Keywords: Obstetrical brachial plexus palsy; Erb's palsy; Chiropractic; Conservative management; Uterine malformation; Premature baby

Introduction

Obstetrical Brachial Plexus Palsy (OBPP) is an injury affecting the brachial plexus and most often occurs during childbirth. The incidence ranges from 0.4 to 5.1 cases per 1000 live births [1-7]. OBPP is frequently reported in babies with macrosomia (>4000 g birth weight) and shoulder dystocia [8]. Those risk factors are observed mostly in women with gestational diabetes [9]. Other risk factors for OBPP include breech presentation [5,6], instrument use (forceps, vacuum) [1-6], intrauterine force during labour [10], impaction of the posterior shoulder on the sacral promontory [9,10], or dysfunctional labour [6,11]. Cesarean section tends to decrease the incidence of this injury [5-12], though it does not exclude the possibility of OBPP [6]. The incidence of this injury in children born by c-section is approximately 1% [13]. Additionally, there is some evidence that maternal uterine malformation may be a risk factor for OBPP [13], but more studies are needed to determine the exact prevalence of this risk factor. It is difficult to know the natural history of this condition because of the lack of long-term follow-up. Many studies overestimate the complete recovery rate, as demonstrated by Pondaag et al. in their systematic review [14].

The most commonly affected branches of the plexus are C5-C6 [15-20], also known as Erb's palsy, which has a better prognosis than total palsy (C5-T1) [6-21]. Upper lesions are more frequent because of the mechanism of injury, which is caused by tensioning of the upper roots due to forceful lowering of the shoulder [19-22]. Recovery time depends on the severity of the injury. Neuropraxia and axonotmesis result in complete recovery, whereas neurotmesis and root avulsion result in permanent loss of arm function. Surgical intervention is performed depending on injury severity, the resulting muscle palsy, and the possibility of spontaneous recovery during the first three months of life [4,20]. According to Nelson and Armenta in their review article [15], there is a three-criteria algorithm to undergo primary reconstruction: First, the patient has total paralysis of the arm without improvement at 3 months of age; second, the patient has no biceps function at 3-6 months of age; and third, the patient has no active movement or less than antigravity movement of the arm at 3-6 months of age. Indicators for surgical intervention are the following: the absence of contraction of the biceps at 3 months of age, the age of recovery, the prognosis of the condition, and the type of intervention needed [20-24]. The Mallet score is commonly used to determine the muscle function of the arm, the evolution, and the improvement of the movement over the time [25,26]. It has been determined that only 19% of infants with OBPP either fully recover or clearly require surgery; the other 81% remain in a gray zone as to which intervention should be done [27].

There are also many conservative alternatives in the treatment of OBPP. Philandrianos et al. [24] applied intensive adapted re-education treatment, including passive and active mobilization and splints on the affected arm for the treatment of infants with upper (C5-C6-C7) obstetrical palsy without the recovery of biceps muscle contraction at 3 months. Their results demonstrated long-term improvement of the shoulder function equivalent to children who had recovered earlier. Vaz et al. [1] had reasonably good results with a constraint-induced movement therapy (CIMT) in a 2-year-old child with Erb's palsy who did not have surgical intervention. Additionally, Ferrero et al. reported a case of typical Erb's palsy that resolved following chiropractic care in a 5-month-old female with severely reduced active shoulder ROM (range of motion), upper limb deep tendon reflexes within normal limits, and no response of the affected arm during Moro and ATNR (asymmetrical tonic neck reflex) testing [28]. However, as far as we know, there is not much evidence supporting chiropractic care for the management of OBPP. The best evidence based in conservative care concerning

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chiropractic care available to the clinician is not well documented.

The aim of this study is two-fold:

1) To present an atypical Erb's palsy presentation in a premature newborn

2) To report his recovery through conservative care after three months without spontaneous resolution. This case report is uncommon because of the absence of major risk factors and because the delivery was performed by cesarean section.

Case Presentation

The primiparous mother went into sudden labour at 33 weeks of pregnancy, with placental abruption. A bicornuate uterus was then noted. The baby was born by cesarean section and exhibited signs of Erb's palsy on the right side at the initial examination, for which a "watch and see" approach was proposed along with monthly physiotherapy rehabilitation treatment. The birth weight was 1814 g, and the mother was not suffering from gestational diabetes or any other health condition except bicornuate uterus.

The baby presented at the outpatient chiropractic clinic at the University du Québec à Trois-Rivières for a second opinion at 3 months of age. Upon examination, the arm was held in adduction, internal rotation and elbow extension, along with the head fixed in full extension and contralateral rotation (Figures 1A and 1B). The neurological exam revealed right biceps and supinator deep-tendon reflexes at 1+. Grip strength was present but decreased compared to the unaffected arm. No active shoulder abduction or elbow supination and flexion movements were noted on the right side. No abnormalities were found in the right clavicle or humerus. The evaluation at Ste-Justine hospital by the physiotherapist showed no muscle contraction in the active range of motion for shoulder external rotation at 0 and 90 degrees of abduction and elbow supination. Shoulder flexion and abduction and wrist extension were less than 50% of the normal active range of motion, and elbow flexion was more than 50% of the normal active range of motion. The other arm active ROM was complete, as was the passive ROM. The Toronto test score for the affected arm was 6.9/10. A 3-month follow-up and a splint were prescribed. The diagnosis of Erb's palsy is appropriate for this case considering all the subjective and objective signs that the baby showed. It is not possible to objectively determine the gravity of the injury, but we assume that there is no complete avulsion of the nerve based on the presence of bicipital reflex. More severe conditions, such as cerebral palsy, brain tumor, or perinatal stroke should be ruled out as they can exhibit signs similar to Erb's palsy. Other palsy, humerus or clavicle fracture and underlying conditions such as Horner's syndrome must be considered because these conditions will influence the prognosis and management of the condition.

The presence of a bicipital reflex on the affected arm, the localization of the nerve injury, and the severity of the injury are favourable for the prognosis in this case. On the other hand, the age of the baby suggests that a spontaneous or complete resolution is less predictable. The presence of a secondary torticollis is also a complication factor for this case.

Results, Intervention and outcome

Treatment consisted of light mobilization and soft tissue techniques for the right arm and neck and was performed twice a week for 9 weeks (for a total of 18 visits). Postural advice for sleep and daytime activities was given, such as avoiding head posture in left rotation while he is playing and attracting his attention to the right side with his toys. The aim was to encourage the utilization of his right arm. There were no red flags for chiropractic management of this case, and the spinal manipulations were executed following the best practices recommendations for chiropractic care in infants [29]. Meningeal stretch was also performed, consisting of gentle traction at the occiput and the sacrum in the opposite direction in order to mobilize the spine and to stretch the meninges. Minor adverse events were observed; the baby was more irritable at the beginning of the treatments but became more tolerant over time.

After the first treatment, an improvement of the active cervical ROM was already noted, and by the 10th appointment, there was no residual restriction of the cervical ROM. At the 5th treatment, the shoulder was moving in flexion, and an improvement in wrist movement was noted. By the 7th treatment, the mother reported that both arms moved in abduction (Figure 2). At the 9th treatment, an improvement in grip reflex and bicipital reflex was measured. At the 15th treatment, the mother reported that the baby was grab-bing toys and rubbing his eyes with his right arm (Figure 3). An improvement was observed in shoulder abduction and horizontal abduction of the shoulder. The patient was able to support his weight on his forearm (elbow flexed at 90 degrees) and maintain his head straight in prone position. By the 18th appointment, the baby was able to flex the elbow and shoulder to reach for a toy that the mother presented to him in front of his face while sitting (Figure 4). The baby was receiving physiotherapy monthly (rehabilitation, mobilization, and electrostimulation) concurrently with chiropractic care. The evaluation of the physiotherapist at 6 months of age showed improvement of all ROM for the affected arm but persistent limitation (less than 50% of normal ROM) for shoulder flexion, abduction and external rotation, and elbow flexion. MRI and the cookie test at 9 months were recommended prior to the consideration of surgical intervention. The patient's visits are summarized in Table 1.

Discussion

The case presented here is uncommon due to the absence of usual risk factors for this injury and because of the improvement observed after three months of age under chiropractic care, which is not often reported in the scientific literature. This case suggests that uterine malformation can be a risk factor for OBPP and corroborates the findings of Alphonso et al. [13].

1A 1B

Figures 1A and 1B: Patient at first appointment (November 26th, 2015).

Figure 2: Patient at his 8th appointment (December 15th, 2015).

Several studies have examined the need for surgical intervention for the brachial plexus and the long-term results. According to Philandrianos et al., a spontaneous recovery with adapted rehabilitation between 3 and 6 months can improve shoulder function with a greater global Mallet score than an early surgical intervention [24]. Guermazi et al. [25] in a retrospective study of 129 OBPP cases concluded that with exclusive rehabilitation, 75% of patients obtained a score greater than or equal to 3 in the Mallet scale (with 2 considered to be a satisfactory recovery). The authors also concluded that reconstructive surgery results in the absence of deltoid contraction at 3 months and that a delay in surgical intervention with total palsy (C5-T1) gives poor results. On the other hand, muscle surgery showed improvement on the functional prognosis of the shoulder when performed on the sequelae (loss of active external rotation of the shoulder, joint deformity, soft tissue and joint contractures). Hale et al. [6] also supported the importance of early intervention for patients with total palsy.

The long-term consequences of this injury can be critical for the development of the children. The condition might affect their activities such as playing outdoors and going up stairs. It can affect their writing skills and result in anxiety and limited self-confidence. It was also reported that 1/3 of children with OBPP will need extra help in class [4]. In a long-term follow-up study including 70 participants with OBPP [30-34], difficulties with daily activities (getting dressed, cutting food, washing and doing hair, shaving the armpits) were revealed.

The main criteria for undergoing surgery are well-established, but it appears that some patients may benefit from this intervention even if they succeed in every step of the examination. Bade et al. [7] performed primary nerve operation on fourteen patients at more than 9 months of age who had passed the tests but had poor recovery of shoulder function, and the authors noticed an improvement in the majority of the cases.

Conservative management of patients should be initiated as early as day one of life to maintain the flexibility of soft tissue and joint, as noted by Yang in her study [20]. Conservative treatment should aim to maintain range of motion to avoid contracture formation, encourage muscle strengthening, prevent compensatory movement patterns, and promote childhood development. All these components can be achieved with chiropractic care.

The pediatric population represents 5% to 20% of chiropractic patients [30-32]. The safety of treatments in infants and children has been questioned many times, but studies have concluded that adverse events due to chiropractic care are exceedingly rare [31]. It is important to adapt the techniques considering patient size, structural development, the flexibility of the joints, and patient preferences [32].

Figure 3: Patient at his 12th appointment (January 21st, 2016).

Treatment	Subjective	Observation	Application and prescription
1	St-Justine diagnosis of Erb's palsy at birth. The baby is in physiotherapy for rehabilitation with no improvement of the condition.	Head posture in left rotation and extension. Arm is held in adduction, pronation, and wrist flexion.	Scapulothoracic mobilization and distraction Meningeal stretch
		Right pectoral, SCM, cervical and thoracic erector spinae hypertonic	TM: Right pectoralis and SCM
	The mother reports hypomobility of the right arm in relation to the left.		AJX: T3 distraction
		PID, T2-3 hypomobility	P: Avoid prolonged posture in right rotation. Can be done with a rolled towel by the neck (left side), especially in car seat
			Note: Specific mobilization of the cervical spine was impossible due to muscle spasm.
2	Improvement in active head mobility	_____	SAB
3	Prescription of a splint at night to maintain wrist in extension and forearm in supination	Passive right rotation of the head (+)	Cervical mobilization and distraction (left lateral flexion and traction)
		Active cervical ROM (+)	Scapulothoracic and thoracic mobilization and distraction Meningeal stretch
		Turns his head to the right when mom calls his name	TM: Right pectoralis and SCM
		Prescription of a splint at night to maintain wrist in extension and forearm in supination	AJX: T3 distraction
			P: Avoid prolonged posture in right rotation
4	Improvement in active right arm mobility	Active flexion of the shoulder (+)	SAB
5	Stable	Active cervical ROM (++) Improvement in the shoulder and wrist movement (flexion and	SAB
		rotation) (++) Movement of the right arm more symmetric in relation to the unaffected arm	
6	Mother observes improvement in general motion and tone of the head. Improvement in arm mobility	_____	SAB
			AJX: C1 left posterior, T3 distraction
7	Stretches his two arms over his head	_____	SAB
			P: Stimulate right side with toys and avoid prolonged head posture in right rotation
8	Stable	Improvement in strength of the arm (+)	SAB
9	Stable	Right bicipital reflex present 1+	SAB
		Improvement in grip strength (+)	TM: Right triceps, deltoid post, pectoral
		Improvement in arm muscle tone (against gravity and in resistance to the movement of the chiropractor)	P: Ventral 2 min 5x AM, 3x PM
10	Improvement in arm abduction. Has some difficulties with ventral position, cries and does not have the reflex to lift his two arms under his chest by himself to support his upper body	Complete active and passive cervical ROM (+++)	Meningeal stretch
			TM: Right biceps and triceps
			AJX: C1 left posterior, T3 distraction, sacroiliac right posterior/inferior
			P: SAB
11	Improvement in arm movement	Improvement in arm strength (++)	SAB
12	Maintains head in extension in ventral position. Baby tends to look more to the right	Improvement in shoulder flexion and elbow flexion	Meningeal stretch
			AJX: C1 left post, T3 distraction, T9 extension, sacroiliac right post-inf, right radial head (flexion, supination)
		General movement increased	TM: Right triceps, biceps and deltoid post
			P: Ventral position
13	Baby slept with his right arm in complete abduction Can stay in ventral position for 2 min before he cries	_____	SAB
14	Stable	Right shoulder ROM: Horizontal abduction	SAB
15	Plays with toys with his two hands Rubs his eyes with right hand	Complete cervical ROM (+++)	SAB
		Active shoulder abduction: 90°	
		Passive shoulder abduction: complete	
		Bicipital reflex: 2+	
		Right hand grip strength equals left hand	

16	Stable	Maintain elbow flexion against gravity	SAB
17	Holds his fist in front of his mouth	_____	SAB TM: Right subscapular and biceps
18	Stable	_____	SAB
19	Medical referral to a physical therapist for rehabilitation	_____	SAB

Table 1: Summary of care provided to the patient.

Figure 4: Patient at his 19th appointment (March 15th, 2016).

Spinal manipulation should be adapted to the patient age following the model of care proposed in the literature [33]. Chiropractic care given to infants or children should always follow the best practices recommendations [29-34].

Limitations

Considering the low level of scientific evidence from a case report, this report provides an interesting introduction for further research to inform the health care population of a different approach for the management of this condition. It is difficult to discriminate the effect size of the chiropractic intervention because of the co-management with physical therapy. It should be noted that improvements began once chiropractic care started.

Conclusion

We report a rare case of Erb's palsy in a premature baby, which has not been reported in the scientific literature as a common risk factor. This case suggests that uterine malformation may be a predisposing factor for OBPP for which the clinician should be aware. Chiropractic care is to be considered as a conservative and noninvasive approach for clinical management of Erb's palsy in cases without spontaneous recovery before surgery is considered. Since the proportion of spontaneous recovery tends to be less than what the literature has shown in the past, early detection and long-term follow-up must be carried out by a qualified healthcare provider who is aware of the complications and the long-lasting impacts of this condition on the neurological and motor development of these children. Chiropractic care could be an interesting option for the management of patients with OBPP. Early detection and appropriate clinical management could significantly reduce the direct and indirect costs of this condition related to the immediate need of care. This type of approach may improve patient outcomes regarding the normal developmental consequences that can be observed. There is a lack of evidence supporting the treatment of children with OBPP with chiropractic care, and we are aware of the necessity to investigate the efficiency of that kind of management in a clinical trial with a larger cohort.

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