

SURABAYA MEDICAL JOURNAL (SMJ IDI SURABAYA)

Volume 2, Issue 1, May 2024, p. 71-79

e-ISSN: 2986-7584 p-ISSN: 2986-2469

Dewi & Gunawan (2024)

∂ OPEN ACCESS

CASE REPORT

Peripheral congenital facial palsy: a rare case report

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Article Info	ABSTRACT
<i>Article history:</i> Received: 21-05-2024 Revised: 25-05-2024 Accepted: 28-05-2024 Published: 31-05-2024	Background: Congenital facial nerve palsy is a rare condition that is typically associated with a traumatic birth event or manifests as a characteristic of a syndrome that necessitates a rehabilitation program. Objective: This report aims to present a rare case and its subsequent outcome after undergoing a rehabilitation program.
<i>Keywords:</i> Congenital; facial palsy; neuropathy; rehabilitation	Case: An eight-month-old infant presented with complaints of an asymmetrical mouth when crying and smiling since birth. The left eye was unable to fully close. No evidence of a syndrome was found. The results of the electromyography of the left facial nerve revealed the presence of partial axonal loss, axonal malfunction,
ORCID ID: Donny Gunawan https://orcid.org/0009-0004- 2127-7719	or neuropathy. The patient was subjected to a comprehensive treatment plan, involving rehabilitation programs such as neuromuscular electrical stimulation (NMES) and laser therapy over a period of 18 months. Discussion: It is important to determine if congenital facial palsy is a standalone condition or part of a broader syndrome. Rehabilitation programs, such as NMES and laser therapy, can stimulate the neurons and improve muscle weakness. The House-Brackmann scale demonstrated a clinical improvement, moving from grade V to grade II. With consistent physical therapy, the patient demonstrated improvements in facial expressions and eye closure. Conclusion: NMES and laser therapy, as well as facial massage and biofeedback, are potential treatments for congenital facial palsy due to their efficacy and favorable outcomes.



Citation:

Dewi, MB. and Gunawan, D. (2024). Peripheral congenital facial palsy: a rare case report. Surabaya Medical Journal, 2(1), p. 71-79, doi: <u>10.59747/smjidisurabaya.v2i1.52</u>

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Highlights

- 1. It is important to determine if congenital facial palsy is a standalone condition or part of a broader syndrome through a comprehensive history and physical examination
- 2. NMES, laser therapy, daily facial massage, and biofeedback have been shown to improve the clinical signs.



BACKGROUND

The incidence of facial nerve palsy is rare in the pediatric population, which has led to considerable distress and anxiety among parents (Baelen et al., 2023). This is due to the rapid onset of symptoms, which can have a significant impact on facial expression, vision, eating, drinking, psychological wellbeing, self-esteem, and quality of life (QoL) (Renault and Quijano-Roy, 2015). The onset is acute and presents with unilateral weakness or paralysis of the facial muscles (upper and lower) due to dysfunction of the peripheral facial nerve on one side of the face (Bilge et al., 2022; Iacono et al., 2022). The prevalence of this disease in the pediatric population is estimated to range from 8% to 25%. The incidence of congenital facial nerve palsies is less common, at approximately two cases per 1,000 individuals (Renault and Quijano-Roy, 2015). Birth traumas or intrauterine developmental abnormalities can result in the development of congenital facial paralysis including prematurity, forceps use, cesarean delivery, birth weight greater than 3,500 g, and firstborn (Ciorba et al., 2015). This results in weakness of the facial muscles, which affects facial expression, oral competence, verbal communication, social interaction, taste and vision in children (Shargorodsky et al., 2010).

Congenital facial palsy can manifest as a standalone condition or part of a broader syndrome, such as Moebius, Poland, and Goldenhar syndromes (Baelen et al., 2023; Buhaibeh et al., 2019). Moebius syndrome is characterized by the dysfunction of the sixth cranial nerve (Souni et al., 2023). Poland syndrome is characterized by the absence of both the major and minor pectoral muscles (Yuan, 2018). Goldenhar syndrome is characterized by an insufficient growth of the first and second branchial arches, resulting in an inadequate unilateral development of the ear, nose, soft palate, lip, and jawbone (Ivănescu et al., 2015). However, Moebius syndrome is frequently observed in cases of congenital facial palsy in the form of bilateral, severe cases (Souni et al., 2023). A number of case studies have reported that congenital facial palsy is accompanied by microtia (Mahale et al., 2016; Sundari and Srinithi, 2022). Therefore, it is essential that a careful and accurate physical examination is conducted to determine if the condition is isolated or part of a syndrome. In addition, the peripheral type of this condition is typically the result of idiopathic causes, including infection, trauma, neoplasm, and immune disorder (Bilge et al., 2022). Consequently, it is commonly referred to as Bell's palsy, which is the most common neurological disorder in children (Khair and Ibrahim, 2018).

It is the responsibility of clinicians to ascertain the etiology, therapeutic options, and potential outcomes for both young patients and their parents, taking into account the functional and aesthetic results that may be expected in the future. Facial palsy can cause significant psychological and social problems due to facial disfigurement (Mat Lazim et al., 2023) and asymmetry. The disfigurement can range from oral incontinence, which can make it difficult to eat and drink, to speech impairments, including difficulties interpreting nonverbal facial cues and an inability to blink, which can result in visual issues (Morea and Jessel, 2020).

This case report presents a congenital isolated facial palsy that demonstrated improvement functional outcomes following a comprehensive treatment with rehabilitation programs incorporating neuromuscular electrical stimulation (NMES) and low laser therapy for 18 months. The therapeutic progress was evaluated using the House-Brackmann scale with photographic documentation.

OBJECTIVE

This report aims to present a rare case of isolated congenital facial palsy and the outcomes of a rehabilitation program.

CASE

An eight-month-old female infant was evaluated by a pediatrician for a suspected congenital facial palsy. The infant exhibited an asymmetrical mouth when crying and smiling since birth. Her left eye was unable to fully close. Her mother reported that the child exhibited no difficulty with feeding or drooling saliva. Furthermore, this was the mother's first pregnancy. During the pregnancy, she was diagnosed with COVID-19 at fifth months gestation and received routine prenatal care from the health services. The mother reported that she had never consumed alcohol or taken certain drugs. She reported



Surabaya Medical Journal (SMJ IDI Surabaya)

e-ISSN: 2986-7584; p-ISSN: 2986-2469

taking prenatal vitamins to support her pregnancy as prescribed by her midwife. In addition, she had no history of hypertension or diabetes.

Two days prior to the anticipated date of delivery, premature rupture of membranes (PROM) occurred and the mother was taken to a hospital and underwent a labor induction. The induction was continued for 24 hours, but no complete opening of the cervix was achieved. Consequently, the decision was made to proceed with a cesarean section. The baby was full-term, with a weight of 2,750 g and length of 49 cm. However, the baby was unable to cry spontaneously. In addition, she exhibited cyanosis, prompting observation in the neonatal intensive care unit (NICU) for one day. The developmental history of the infant is as follows: able to roll over at three months of age, able to sit at six months of age, and able to babble and respond well to calls at eight months of age.

Upon physical examination, the following observations were made: the body weight was 7 kg, the body length was 70 cm, the heart rate was 115 beats per minute, the respiratory rate was 32 breaths per minute, and the oxygen saturation was 98% in ambient air. The examination of the head and neck area revealed a facial asymmetry with no microtia. The examination of the thorax revealed no retraction of intercostal or subcostal muscles nor enlargement of the cardiac border. The examination of the heart revealed that the first (S1) and second (S2) heart sound was normal without the presence of murmurs or gallops. Pulmonary examination revealed vesicular sound without wheezes or rhonchi. Abdominal examination revealed a normal peristaltic sound with no palpable hepatomegaly or splenomegaly. The extremities were observed to be normothermic without any evidence of swelling. The musculoskeletal status was as follows: the range of motion of the neck, trunk, as well as upper and lower extremities was full. In addition, the functional muscle strength of the left facial nerves (frontalis, corrugator supercilii, orbicularis oculi, nasalis, orbicularis oris, zygomaticus mayor, and buccinator) was weak. The examination of the special sensory and parasympathetic left facial nerve was challenging to evaluate using the Schirmer's test, acoustic reflex test, and sensory test. The neurological status was as follows: The impressions of the cranial nerves I, II, III, IV, V, VI, VIII, IX, X, XI, XII were found to be normal. The cranial nerve VII presented with peripheral paresis of the left side. The physiological reflexes, namely BPR, TPR, KPR, and APR, were observed to be positive. The patient exhibited no pathological reflexes in response to the Hoffman, Tromner sign, Babinski reflex, and Chaddock reflex tests, which were all negative. The local status of the left face was unremarkable, with no redness nor swelling and a gap in the left eve. Palpation revealed no warmth and reduced flexibility in the orbicularis oris muscle. The functional state was as follows: the patient exhibited adequate balance when sitting and standing, as well as the ability to roll over. Transferring from a lying to sitting position and from a sitting to standing position was also possible. Hand to eye, hand to hand, and hand to mouth coordination was found to be satisfactory. The physical examination revealed a facial asymmetry, with the angle of mouth deviating to the left side. There were no forehead furrow lines and the left eye did not fully close. The patient exhibited a severe dysfunction, with a grade V on the House-Brackmann (HB) scale.



Figure 1. Newborn of grade V (HB scale)





Figure 2. Eight-month infant of grade V (HB scale)

An electromyography (EMG) was conducted on the infant, but a needle EMG was not performed. The results of the nerve conduction study (NCS) indicated that the evaluation of the left zygomatic and marginal mandibular branch demonstrated normal distal latency, decreased amplitude, and normal velocity. It was concluded that the motor conduction was normal and that low amplitudes indicated partial axonal loss or axonal dysfunction or neuropathy of the left facial nerve.

Site NI	R Onset (ms)	Norm Ons	et (ms)	O-P	Amp (mV)	Nor	m O-P An	p Dist (cn	n) Vel (m/s)	Norm Vel (m/s
Left Facial -	Oculi Motor (Orbic Oculi)	1.200	-							26
Mastoid	3,0	<3,1		0,2			>0,8	7	23,3	<30	
Left Facial -	Oris Motor (C)rbic Oris)	1000	1	1		1.57				26
Mastoid	3,2	<5,4	1	1-121×	0,1	1000	111	>0,8	6	20	<30
CV [Left Facial -	Oculi Motor]		NCV [Left Facial - Oris Motor]								
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Figure 3. NCS results of electromyography indicating a partial axonal loss or axonal dysfunction or neuropathy of the left facial nerve

The patient has underwent NMES and laser therapy for 18 months. Her mother indicated that her development has been considered significant to date. A visible forehead furrow line has emerged. From the outset, the eyes did not fully close. However, this is no longer the case. In addition, she exhibits normal speech and chewing abilities, with no difficulty when drinking with a straw without leakage although the lips are asymmetrical. Her asymmetrical lips are particularly evident when she smiles. As a result, the House-Brackmann scale has been upgraded from a severe disfunction (grade V as illustrated in Figures 1 and 2) to a mild disfunction (grade II as illustrated in Figure 4). The parents expressed a desire for their daughter to have lips that were as symmetrical as those of other children to facilitate her psychosocial development.



Surabaya Medical Journal (SMJ IDI Surabaya) e-ISSN: 2986-7584; p-ISSN: 2986-2469



Figure 4. Two-year child of grade II (HB scale)

DISCUSSION

Congenital facial palsy can result from embryonic or acquired causes. In most cases, it presents at birth in children. Its anatomical placement makes it more susceptible to harm from inflammatory responses. Facial palsy in newborns is uncommon, affecting between 0.23% and 1.8% of live births, and 78% to 90% of the cases are related to birth trauma (Saini et al., 2013). However, in this report, the infection of COVID-19 during pregnancy appears to be the cause of the disease (Bilge et al., 2022), as viral infections can induce facial palsy, such as those observed in cases of Varicella zoster (Zhang et al., 2020). A case report documented facial nerve palsy resulting from COVID-19 infection in a five-year-old boy (Iacono et al., 2022). In addition, a case series involving six patients with face palsy at birth reported that the mothers had tested positive for COVID-19 infection and had experienced vacuum extraction (one case) or postpartum complications (two cases). However, no birth trauma or a history of facial palsy was reported in all the cases (Decraene et al., 2020). Furthermore, systematic review of adult patients with COVID-19 revealed that 30.14% of cases had the Guillain-Barre syndrome, with 20.97% presenting with bilateral facial paralysis and 77.03% with ipsilateral paralysis (Khurshid et al., 2022). It was previously believed that coronaviruses have a neurotropic effect and a transcribial route to the brain, although the exact mechanism remains unclear. Once they enter the central nervous system (CNS) through the olfactory nerve and bulb, they cause direct damage to the brain by binding to angiotensinconverting enzyme 2 receptors, which are prevalent in glial cells and neurons (Ferreira et al., 2022).

The presence of facial asymmetry when a baby is crying is an important indicator in differentiating between facial paralysis due to nerve damage and that due to congenital absence of the depressor anguli oris muscle (DAOM) (Ferreira et al., 2022). The condition is referred to as neonatal asymmetric crying facies (NACF), which is a specific phenotype of asymmetry of the mouth and lips during grimace or smiles, marked by the presence of asymmetrical lower lips depression (Arya et al., 2017). Furthermore, the ability of a patient to smile and close both eyes is the important indicator in distinguishing between peripheral and central facial palsy. In the partial type, the unaffected side will move normally (Garro and Nigrovic, 2018). Consequently, a comprehensive clinical examination is essential for infants with facial nerve palsy. In addition, parents must be interviewed in detail regarding their pregnancy and delivery. It is important and highly recommended to identify the causes (Bilge et al., 2022). Drooling of saliva indicates that the infant is unable to regulate oral secretion due to a dysfunction in the coordination of orofacial and palatolingual musculatures, which results in impaired swallowing (Meningaud et al., 2006). Therefore, comprehensive medical records should include details regarding perinatal history, chronological progression of the palsy, psychomotor development, and visual impairment (Decraene et al., 2020).

COVID-19 infection during pregnancy is associated with an increased risk of premature birth by 3.34 times and caesarean section by 3.63 times. However, the infant in this report was confirmed to be negative for severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) and have no abnormal computed tomography (CT) results (Wen, 2022). The majority of premature births are attributed to iatrogenic causes and are not associated with preterm premature rupture of membranes (PPROM), which occurs four times more frequently in pregnant women who tested positive for SARS-CoV-2



(Martinez-Perez et al., 2021). PROM may also result from a viral infection which spreads to the uterus and amniotic fluid. Viral infection may also result in a proinflammatory response that increases the number of leucocyte cells. Therefore, COVID-19 infection has been associated with increased levels of leucocytes in pregnant women and the occurrence of PROM (Margono et al., 2021). PROM is also associated with morbidity and mortality in both mothers and infants due to sepsis, hypoxia, and pulmonary hyperplasia, which have been identified as contributing factors in infant mortality. Viral infection increases the risk of poor outcomes for both the mother and infant (Akter et al., 2022). These findings are consistent with this report. Furthermore, COVID-19 infection may lead to the development of Bell's palsy in the form of a primigravida in pregnant women with a confirmed diagnosis of COVID-19 (Kumar et al., 2021). Other risk factors that contributed to the development of facial palsy in this report was the anatomical position of the fetus during pregnancy. It is possible that the extremities of the fetus might have compressed the mastoid region, causing compression of the facial nerve or arteries that supply oxygen to the facial nerve, thereby leading to ischemia. Facial palsy might also result from inflammation and ischemia (secondary, tertiary or the squamous), which in turn thickens the facial nerve sheath and forms fibrotic bands that cause nerve transection and compression (Zhang et al., 2020). Moreover, the prolonged labor process exerted pressure. The mother of this patient received labor induction for 24 hours without any progress in the opening of the cervix. As a result, it was decided to proceed with a cesarean section, which itself carries an inherent risk factor. This can occur due to the first pregnancy, whereby the cervix has not dilated properly. A case report of a similar case (male infant with a birth weight of 2.575 g and APGAR scores of 9 and 10 during the first and five minutes after delivery, respectively) indicated that labor induction resulted in facial palsy due to cerebellar hemorrhage (Coviello et al., 2020).

The physical examination revealed no evidence of syndromes related to facial palsy, such as microtia and absent pectoralis major. In addition, the cranial nerve examination revealed no abnormalities other than the nervus facialis. Furthermore, the examination of the development of language and pronunciation revealed no hearing impairment. Therefore, it is highly likely that this case is congenital non-syndromic facial palsy. Normal functional muscle strength with no pathological reflexes indicated that the brain is functioning normally. The cerebral cortex, particularly the motor cortex, was believed to be essential for muscle movement and control (Clark et al., 2014).

The symptoms of facial nerve palsy vary depending on the location of the damage. Infranuclear lesions form distal to the facial nerve nucleus, causing peripheral facial palsy in the ipsilateral upper and lower face (Ho et al., 2015). Once the facial motor nerve has emerged from the skull, it splits into two primary terminal branches at the stylomastoid foramen, producing the posterior auricular branch. The inferior cervicofacial branch, which descends along the mandible, supplies the muscles in the lower part of the face. It gives rise to the buccal, mental, and cervical branches, which in turn supply the risorius, buccinator, orbicularis oris, depressor anguli oris, depressor labii inferioris, mentalis, and platysma muscles. The temporofacial branch runs in a horizontal anterior direction, producing frontal branches which innervate the frontalis and orbicularis oculi muscles; suborbital branches which innervate the levator labii superioris, zygomaticus, levator anguli oris, and dilatator naris muscles; and buccal branches which innervate the buccinator and orbicularis oris muscles (Renault and Quijano-Roy, 2015). In this case, the inferior cervicofacial and superior temporofacial branches may also be affected.

The House-Brackmann scale was used to evaluate this patient. The Facial Nerve Dysfunction Committee of the American Academy of Otolaryngology-Head and Neck Surgery has recognized this scoring system as the standard for evaluating facial palsy. The scale employs a six-point grading system, with the first grade denoting normal function and the sixth grade denoting total flaccid paralysis (Morea and Jessel, 2020). There was a progression from a severe disfunction (grad V as illustrated in Figures 1 and 2) to a mild dysfunction (grade II as illustrated in Figure 3) following a rehabilitation program involving NMES and laser therapy, as well as massage and biofeedback, for 18 months. This report is consistent with a study by Dewi et al. (2020), which found that NMES, biofeedback, and massage therapy can improve peripheral facial palsy. It is well known that a small electrical current is used in NMES. This therapy is used for neurologic illnesses causing paralysis by stimulating neurons that supply the affected muscles. NMES is useful for neuromuscular rehabilitation, muscle strengthening, avoiding muscular atrophy, and restoring facial muscle strength in individuals with stroke who are facially paralyzed (Marotta et al., 2020). Electrical stimulation has been used since the 1950s with the objective of improving facial function, minimizing sequelae, stimulating nerve regeneration, and



Surabaya Medical Journal (SMJ IDI Surabaya)

e-ISSN: 2986-7584; p-ISSN: 2986-2469

maintaining muscle bulk and contractile properties. In addition, the cellular quality of muscle reinnervation has been investigated in animals, where electrical stimulation was used to reduce the incidence of polyinnervation, fibrosis, Wallerian degeneration, and demyelination (Fargher and Coulson, 2017).

Another rehabilitation program performed on this patient was low-level laser therapy (LLLT). It is regarded as a non-painful and non-invasive treatment approach suitable for all kinds of patients, including those who are unable to take corticosteroids. By stimulating intracellular signaling pathways associated with nucleic acid synthesis, protein synthesis, enzyme activation, and cell cycle progression, laser prevents cell death, reduces inflammation, and promotes cell regeneration (Kandakurti et al., 2020). A study have demonstrated that it enhances axonal development and myelinization, as well as boosting the functional activity of the wounded peripheral nerve (Kim et al., 2020). However, only a few randomized control trials have evaluated the improvement in outcomes following physical therapy (Bharti, 2021). In addition, this patient was administered home therapies, including facial massage, biofeedback, and warm compresses three times a day with a duration of 10 minutes. Studies have demonstrated that these therapies can facilitate muscle relaxation and improve muscle tone, as well as stimulate muscle strength through specific movements in front of a mirror (Baba et al., 2021; Dewi et al., 2020). In this case, the parents have high expectations that their daughter will achieve symmetrical lips similar to those of other children, given her psychosocial development. The prognosis for peripheral facial palsy in children is likely to be more favorable than in adults (Baba et al., 2021). Should the patient's condition at the end of therapy fall short of parental expectations, it is recommended that they undergo dynamic facial reanimation surgery, with a recovery outcome of more than 80%, and crossfacial nerve grafting, with a recovery outcome of more than 83% (Ciorba et al., 2015). Parental involvement is crucial in the successful therapy. It is essential that doctors provide encouragement to the parents to regularly adhere to therapies at the hospital and continue performing home treatments, including the use of warm compresses, biofeedback, and massage therapy.

Limitations

Due to the rarity of non-syndromic facial palsy, it was challenging for the authors to find comparable case reports involving NMES and laser therapy.

CONCLUSION

In cases of congenital facial palsy, a comprehensive history and physical examination is necessary to determine if the facial palsy is part of a syndrome or a standalone condition. Following this, the physician can prescribe an appropriate comprehensive treatment with a rehabilitation program that aims to maximize the function of the affected facial muscles. NMES and laser therapy can be considered as forms of rehabilitation therapy, in addition to home therapies such as massage, biofeedback, and warm compresses. Such an intervention will not only support the patient, but it will also facilitate the psychosocial development of the patient and their family.

Acknowledgment

The author would like to thank the patient and her guardians for their consent to publish the case.

Conflict of Interest

All authors have no conflict of interest to declare.

Funding

None.

Author Contribution

The authors contributed to all stages of this report, including preparation, data collection and analysis, drafting the manuscript, and approval for the publication of the manuscript.



Declaration of patient consent

The authors certify that they have obtained all necessary patient consent forms. In the form, the guardians of the patient have provided images and other clinical information for publication in the journal. The guardians understand that the names and initials of both the patient and the guardians will not be published and that reasonable efforts will be made to conceal their identities.

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Surabaya Medical Journal (SMJ IDI Surabaya)

e-ISSN: 2986-7584; p-ISSN: 2986-2469

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