

Comprehensive oral treatment of drooling factors in child patients with neonatal asphyxia and diagnosed with DDH: case report

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ABSTRACT

Introduction: Drooling is characterized by salivary incontinence, which involves the involuntary leakage of saliva onto the lower lip. It can be caused by anterior sialorrhea and oral motor dysfunction. The instability of the hip region in these patients leads to misaligned body posture, potentially affecting oral motor function. Additionally, rampant caries can contribute to sialorrhea, further worsening the issue of drooling in these individuals. This case report aims to present a comprehensive oral treatment approach for managing drooling in pediatric patients with a history of neonatal asphyxia and a diagnosis of developmental dysplasia of the hip (DDH). Case report: A 6-yearold girl was brought to the Pediatric Dental Clinic at Al Ihsan West Java Provincial Hospital by her mother due to concerns of excessive salivation and frequent toothache. Upon clinical examination, multiple caries was detected. The patient's diagnosis included developmental dysplasia of the hip (DDH), which had led to poor posture due to hip bone dysplasia. Comprehensive dental treatment was administered under general anesthesia, encompassing procedures such as pulpectomy, placement of Stainless-Steel Crowns (SSC), Strip crowns with Glass Ionomer Cement (GIC) restoration, fissure sealant application, topical fluoride treatment, and extractions. During the one-week followup, the patient did not report any pain and experienced a reduction in drooling. The patient was subsequently referred to the medical rehabilitation department for addressing swallowing difficulties. Concurrently, the patient is continuing DDH treatment with an orthopedic specialist at Santosa Hospital in Bandung. Conclusion: Comprehensive oral care and consultation with orthopedic specialists are approaches to mitigate drooling in patients with a history of neonatal asphyxia and a diagnosis of DDH. A multidisciplinary collaboration involving both dental and general medical specialities is essential to address the core issues and enhance the quality of life for these children.

Keywords

comprehensive oral treatment; drooling factors; neonatal asphyxia; developmental dysplasia of the hip (DDH)

Perawatan oral komprehensif faktor drooling pada pasien anak asfiksia neonatorum dan terdiagnosis DDH: laporan kasus

ABSTRAK

Pendahuluan: Drooling adalah inkontinensia saliva atau tumpahan saliva yang tidak disengaja ke bibir bawah karena sialorrhea anterior dan disfungsi motorik mulut. Instabilitas daerah pinggul pada pasien ini menyebabkan postur tubuh menjadi kurang selaras sehingga berpengaruh terhadap motorik oral. Karies rampan menyebabkan sialorrhea yang juga memperparah drooling pada pasien ini. Tujuan dari laporan kasus ini adalah untuk melaporkan perawatan oral yang komprehensif pada pasien anak yang drooling dengan asfiksia neonatal dan didiagnosis displasia perkembangan pinggul (DDH). Laporan Kasus: Pasien anak usia 6 tahun dibawa ibunya ke Klinik Gigi Anak di RSUD Al-Ihsan dengan keluhan mengeluarkan saliva berlebih dan sering sakit gigi. Hasil pemeriksaan klinis didapatkan karies hampir pada seluruh gigi. Pasien didiagnosis dengan Displasia Perkembangan Pinggul (DDH) yang mengakibatkan postur tubuh yang buruk yang dikaitkan dengan displasia tulang pinggul. Perawatan gigi yang komprehensif, dilakukan dengan anestesi umum, meliputi berbagai prosedur seperti pulpektomi, Stainless Steel Crown (SSC), restorasi Strip crown dengan Glass Ionomer Cement (GIC), fissure sealant, perawatan fluoride topikal, dan ekstraksi. Saat kontrol satu minggu, tidak ada keluhan sakit dan drooling berkurang. Pasien dirujuk ke bagian rehabilitasi medik untuk dilakukan perawatan lanjutan pada masalah penelanan. Pasien masih melakukan perawatan DDH dengan dokter spesialis ortopedi di RS Santosa Bandung. Simpulan: Perawatan komprehensif rongga mulut dan perawatan ortopedi merupakan salah satu cara untuk mengurangi faktor penyebab drooling pada pasien yang memiliki riwayat asfiksia neonatorum dan didiagnosa DDH. Kerjasama multidisiplin baik dalam bidang spesialisasi kedokteran gigi maupun dalam bidang spesialisasi kedokteran umum diperlukan agar masalah utama teratasi dan kualitas hidup anak menjadi lebih baik.

Kata kunci

perawatan oral komprehensif; faktor drooling; asfiksia neonatal; developmental dysplasia of the hip (DDH)

INTRODUCTION

The stability of the hip region influences correct posture.¹ One of the abnormalities that can disrupt the stability of the hip region is hip dysplasia or DDH. This condition involves abnormal hip development, resulting in dysplasia, subluxation, and hip joint dislocation in children. Properly developing a child's hip hinges on maintaining the stability of the femoral head within the acetabulum. Acetabular dysplasia leads to anomalies in the development of the socket, encompassing variations in size, shape, and positioning. In the case of hip subluxation, the femoral head is partially displaced from its usual position, yet it maintains some degree of contact with the socket. Conversely, a dislocated hip involves the femoral head losing contact with the socket. Should the hip joint lack stability and anatomical alignment, its proper development will be compromised, potentially resulting in persistent abnormalities as the child reaches the walking stage.²

The prevalence rate for DDH varies across countries and continents, ranging from 1 to 34 cases per 1000 births.³ In a systematic review of unscreened populations, the estimated prevalence of clinically diagnosed established hip dysplasia was 1.3 per 1,000. However, in populations subjected to clinical screening using the Ortolani and Barlow tests, the prevalence ranges from 1.6 to 28.5 cases per 1,000. Furthermore, when ultrasound screening is employed, the prevalence becomes even higher.⁴

The etiology of DDH in children with typical development remains unknown. Genetic and environmental factors influence the development of this condition.⁵ Predictors of DDH include breech presentation, a family history of DDH, and being female. Ligamentous laxity and disorders related to collagen metabolism, estrogen metabolism, pelvic instability associated with pregnancy, and the use of swaddling are strongly correlated with DDH.²

Conservative management is frequently successful when early diagnosis and treatment are implemented. Early diagnosis is a fundamental prerequisite for achieving optimal outcomes and minimizing the potential for long-term complications. The optimal timeframe for diagnosis and treatment has been established as within the sixth week of life; beyond this point, the challenges associated with treatment tend to escalate. DDH can result in compromised hip function and early-onset degenerative joint disease, especially if treatment is postponed. Hip instability, when identified during the neonatal period, can often be effectively managed through non-surgical methods in most cases. However, a delay in diagnosis raises the probability of requiring surgical intervention.

Dislocated hips lose their crucial role in stabilizing and controlling body posture in the sagittal plane, potentially leading to compensatory adjustments in spinal-pelvic alignment to maintain an upright standing position.⁷ Poor posture, including challenges in maintaining an upright or flexed position, represents one of the clinical variables associated with drooling.^{8,9} The typical passage of saliva from the mouth to the esophagus involves several factors, including cognitive perception and control, sensory sensitivity in the mouth, control of facial muscles, coordination between the palate and tongue, coordination of head and neck muscles, and oral motor skills. The occurrence of drooling is typically a result of a combination of disruptions in these factors.⁹⁻¹¹

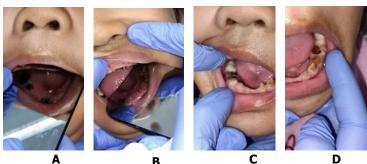
Drooling involves the involuntary leakage or unintended release of saliva onto the lower lip, a condition known as anterior sialorrhea. While drooling is frequently observed in typically developing infants, it tends to diminish between 15 and 36 months of age as salivary control matures. Nevertheless, if this condition persists beyond four years, it is generally considered pathological. The overall prevalence of substantial drooling in childhood is 0.6%. Drooling leads to both physical and psychosocial complications, exerting a substantial negative influence on quality of life. Physical complications encompass skin maceration around the mouth, which can lead to secondary infections, unpleasant odors, dehydration, speech difficulties, and eating disorders. Individuals experiencing drooling also have an elevated risk of aspirating saliva, food, or liquids into the lungs, particularly when typical reflex actions like gagging or coughing are impaired. On the psychosocial front, complications encompass social isolation, barriers to education, heightened levels of dependence and required care, diminished self-esteem, and challenging social interactions. Sialorrhea, or excessive drooling, profoundly affects patients and their caregivers' quality of life. 12

Managing sialorrhea or excessive drooling is most effectively achieved through a multidisciplinary approach. A comprehensive team of pediatricians, physiatrists, neurologists, otolaryngologists, dentists, and speech therapists is essential.^{1,12} The management is carried out in a gradual approach, from conservative to more invasive.¹ Generally, a non-invasive approach that emphasizes oral function and postural control is typically implemented early.⁹ The primary objective in managing drool is to enhance oral motor control over secretions, diminish the social and health repercussions stemming from sialorrhea or drooling, enhance the quality of life for both patients and caregivers and alleviate the burdens encountered by caregivers.¹ Prior to this, no similar case had been published. This case report aims to document the comprehensive oral treatment administered to pediatric patients with drooling tendencies who have a history of neonatal asphyxia and have been diagnosed with developmental dysplasia of the hip (DDH).

CASE REPORT

A 6-year-old girl with multiple dental cavities and excessive drooling sought treatment at the Pediatric Dental Clinic of Al Ihsan Hospital. The parents reported multiple cavities and persistent drooling. A previous dentist had referred her due to her uncooperative behavior during dental procedures. The parents also mentioned that she is their only child.

During the history-taking process, it was discovered that the patient had experienced neonatal asphyxia due to prolonged labour. Additionally, the patient had encountered a tuberculosis infection at the age of one, along with frequent episodes of fever. The developmental milestones for the child were not consistently aligned with the typical developmental trajectory for her age. For instance, the patient initiated prone lying at 7-8 months and achieved sitting at 12 months but bypassed the crawling phase. Instead, she moved with her stomach and stood at 18 months. She commenced walking when she was not around 36 months of age, following traditional walking therapy in Majalaya. When walking for extended durations, she tends to walk on her tiptoes, and there is a noticeable height difference between her left and right feet. She was brought to a doctor and referred to an orthopedic specialist. Following an assessment by the orthopedic specialist, she received a diagnosis of developmental DDH and is currently undergoing treatment. She did not exhibit any learning difficulties and displayed the capacity for regular eye contact. However, she experienced challenges in socializing with unfamiliar individuals and peers. Typically, she cried and became agitated when in a new environment.



A B C D

Figure 1. Intraoral photograph (A-D) shows extensive caries in almost all teeth.

During the extraoral examination, the patient displayed symptoms of drooling and utilized a bib to safeguard her clothing. She required protective measures or frequent clothing changes due to excessive drooling. On intraoral examination, multiple cavities were observed on teeth numbered 55, 54, 62, 63, 64, 65, 75, 74, 73, 72, 82, 83, 84, and 85 (Figure 1).



Figure 2. Panoramic radiograph of the patient.

The patient declined to have intraoral photographs taken; consequently, only a limited number of intraoral photos were acquired, as depicted in the provided images. A panoramic radiograph examination revealed extensive caries in teeth 54, 62, 63, 64, 65, 75, 74, 72, 82, 84, and 85, with pulp involvement (Figure 2). Based on the patient's medical history, intraoral examination, and radiographic findings, the following diagnoses were made: Teeth 55, 75, and 85 exhibited pulpal necrosis, and pulpectomy was recommended as the treatment plan with a favorable prognosis. Teeth 73 and 83 were diagnosed with reversible pulpitis, and treatment involved using Glass Ionomer Cement (GIC) and compomer fillings. Deep fissures in teeth 36 and 46 were identified and were treated with fissure sealants. However, teeth 54, 62, 63, 64, 65, 74, 72, 82, and 84 displayed pulpal necrosis, and the treatment plan recommended was extraction due to a poor prognosis.

Initially, the patient underwent chairside treatment utilizing the behavior guidance technique known as "tell-show-do." Nevertheless, she continued to display uncooperative behavior and became hysterical when the procedure was attempted in the dental chair.



Figure 3. The scene during the operation.

A treatment plan was formulated and subsequently discussed with the patient's mother, who provided her consent by signing an approved informed consent form authorizing the dental treatment to be performed under general anesthesia. The comprehensive dental treatment, conducted under general anesthesia, took place in the operating room of Al Ihsan Hospital and spanned 1 hour and 30 minutes (Figure 3).

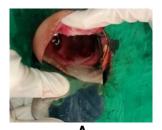




Figure 4. Maxillary and mandibular post-operative intraoral images (A-B)

The dental treatment procedures comprised oral prophylaxis for both the maxilla and mandible, pulpectomy performed on teeth 55, 75, and 85 along with subsequent Stainless Steel Crown (SSC) restoration, Glass Ionomer Cement (GIC) filling with strip crown for tooth 73, dental compomer fillings for tooth 83, fissure sealant application on teeth 36 and 46, as well as topical fluoride treatments. Additionally, extractions were performed as part of the treatment plan (Figure 4).



Figure 5. Post-operative intraoral picture 3 months

During the one-week follow-up examination, the patient reported no toothache and a noticeable reduction in drooling. Additionally, the patient was referred to the medical rehabilitation department to address her swallowing difficulties. At the three-month follow-up, the restorations were still intact, and there were no reports of toothache (Figure 5).

Evidence indicates a noticeable reduction in salivary drooling. As per the therapist's assessment, drooling from the corner of the lip was attributed to the patient's chewing habit, which was influenced by toothache. Due to pain when chewing, the patient refrained from using the left side, especially given that the upper teeth had been extracted. Consequently, a plan was devised to employ an upper denture as a space maintainer for the patient. The patient's mother and the patient received further oral health education at home, aiming to uphold her oral health following the comprehensive dental treatment. The education encompassed guidance on brushing

teeth twice daily with fluoride toothpaste to manage plaque and suggestions for adopting a low-sugar diet. A follow-up appointment is scheduled for the patient six months after the dental treatment is conducted under general anesthesia in the operating room.



Figure 6. Photos of the patient's posture show poor posture due to DDH.

The patient has DDH, and her posture is not optimal, as her head and left shoulder are positioned more forward than her right shoulder (Figure 6). The left and right legs still need to be appropriately aligned. The patient continues wearing three layers of diapers to support the open-leg position. This is necessary because the bones are not correctly positioned, causing her to walk with a somewhat wider stance.

DISCUSSION

In this case report, the patient presented with concerns regarding cavities and drooling. During the history-taking process, the patient's mother conveyed that the patient did not cry immediately after birth and exhibited a blue discoloration due to prolonged labor. This particular symptom is indicative of neonatal asphyxia. According to Tohaga *et al.*, ¹⁴ Asphyxia experienced by infants is among the risk factors for hypocalcemia. The prevalence of hypocalcemia in cases of asphyxia is estimated to be around 30% to 40%. Calcium and phosphorus play a crucial role in forming the mineral salts that constitute bones and teeth within the body. ¹⁴ Inflammation, such as that caused by teething, dental caries, and oral infections, can increase salivary secretion, thereby worsening the issue of drooling. ¹²

The patient underwent dental management to address the issue of drooling. The decision to proceed with oral rehabilitation and treatment under general anesthesia was determined by the severity of caries and the patient's adverse behavior towards dental procedures. Intraoral examinations revealed tooth cavities numbered 55, 54, 62, 63, 64, 65, 75, 74, 73, 72, 82, 83, 84, and 85. The patient's caries risk assessment categorizes her at high risk for developing cavities. The management of children with high caries risk aged over six years, as per AAPD¹⁵ guidelines, includes a control visit every three months, radiographic examinations every six months, brushing teeth with 0.5% fluoride-containing paste or gel, applying topical fluoride every three months, utilizing 38% silver diamine fluoride to halt cavity lesions, offering diet counselling, administering sealants, actively monitoring white spots, and restoring cavity lesions. Therefore, the dental treatment plan devised for this patient at that time involved the application of fissure sealants on teeth with deep fissures, using Glass Ionomer Cement (GIC) and compomer fillings on teeth exhibiting reversible pulpitis, conducting root canal treatment on necrotic teeth with a favourable prognosis, and extracting teeth with a poor prognosis. Additional aspects encompassed topical fluoride application during the intervention and follow-up visits, providing oral health education (DHE) to the patient's parents, offering dietary counselling, and ensuring follow-up examinations every three months.

A comprehensive dental treatment was undertaken due to indications requiring extraction and root canal treatment in several teeth. This included the extraction of teeth numbered 54, 62, 63, 64, 65, 74, 72, 82, and 84, as well as the performance of pulpectomy followed by the placement of Stainless Steel Crowns (SSC) on teeth 55, 75, and 85. Additional dental procedures encompassed dental compomer fillings with crown strips on teeth 73 and 83, fissure sealants on teeth 36 and 46, maxillary and mandibular scaling, and the topical application of fluoride using Clinpro. Stainless steel crowns are widely recognized as one of the most commonly employed and effective devices for restoring primary teeth that have undergone endodontic treatment or display structural defects, such as severe caries. ¹⁶ Vulicevic in Gabrovska *et al.*, ¹⁷ claim, the basic advantage of pediatric crown restorations is to protect the dental pulp from harmful external factors and preserve its vitality.

Compomers, also called dental adhesives, as suggested by their name, exhibit a blend of traits from composites and glass ionomers, combining into a single component. Compomers possess attributes characteristic of glass ionomers, including adhesive properties and the release of fluorine. Furthermore, compomers, which take on a tooth-colored appearance upon hardening, yield an aesthetic outcome. ¹⁸ Fluoride varnish offers several advantages in comparison to other topical fluoride applications. It sets quickly upon application to enamel surfaces and does not necessitate thorough teeth drying. Additionally, it is straightforward, swift, and does not require intricate technique. ¹⁹

During the one-week follow-up visit, there were no reported complaints of toothache, although drooling persisted. Upon observation, it was noted that the patient had developed a habit of infrequently swallowing saliva. Consequently, the patient was referred to the medical rehabilitation department at Al Ihsan Hospital for an assessment and treatment of oral motor skills. According to Hulst K van¹¹, the underlying cause of pathological drooling in young children can be a combination of oral motor difficulties related to swallowing, issues with sensory processing in the oral areas connected to saliva, and an inappropriate sequence of sensations that occur when saliva is involuntarily expelled alongside active swallowing.

Drooling treatment based on the principles of oral recognition, sensory control, and increased motor function is supported by research conducted by Crysdale in Min *et al.*, ⁹ who analyzed the results of drooling management in a Canadian rehabilitation center, found that oral exercise therapy improved oral motor function. An individualized physiotherapy program to rehabilitate orofacial neuromuscular function comprises coordinated and repetitive exercises, often presented as simple games. These exercises aim to enhance lip seals (for instance, blowing through a straw) and incorporate techniques to heighten sensory awareness (such as exposing the mouth to various textures). The overarching goal is to improve chewing capacity, foster the formation of a food bolus, and enhance the swallowing process.

This exercise is most suitable for children who exhibit mild to moderate oral dysfunction, possess solid cognitive skills, and display high motivation.²⁰ This regimen follows the oral motor therapy administered to this patient, which encompasses balloon-blowing exercises and advice to chew food on both sides using a variety of food textures

The patient is afflicted with DDH disorder, contributing to an inadequate posture. As illustrated in Figure 6, the child utilizes three-layer diapers as a makeshift support instead of a cast to maintain the open leg position, given the suboptimal bone alignment. Regrettably, this situation worsens the prevalence of drooling in the patient. According to Hulst $et\ al.$, ¹¹ effective oral function begins with achieving head stability to ensure jaw control. Head stability is affected by torso alignment, which is affected by hip region stability. Based on the research conducted by Harila et al. and Kim $et\ al.$, ²¹ children with congenital hip dislocation are more prone to asymmetric occlusive growth and the development of lateral crossbites. Malocclusion is a common musculoskeletal problem that contributes to drooling.²

The treatment of patients in this case report is conducted through a multidisciplinary approach encompassing various specialized fields. These fields include pediatric dentistry, pediatrics, and anesthesiology, each contributing to dental treatment under general anesthesia. Collaboration among the medical team members is paramount during procedures conducted under general anesthesia, necessitating appropriate equipment and facilities to facilitate swift response during emergencies. Pediatric dentists are responsible for dental care, medical rehabilitation physicians oversee post-treatment oral motor care for drooling, and orthopedic doctors manage DDH cases.

CONCLUSION

Comprehensive oral care and consultation with orthopedic specialists are approaches to mitigate drooling in patients with a history of neonatal asphyxia and a diagnosis of DDH. A multidisciplinary collaboration involving both dental and general medical specialities is essential to address the core issues and enhance the quality of life for these children.

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