

Case Study

Management of Drug Induced Gingival Overgrowth (Digo) in a Patient with Cerebral Palsy Type Spastic Quadriplegia: A Case Report

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ABSTRACT

Drug Induced Gingival Overgrowth (DIGO) can be defined as local or general enlargement of the gingival tissue that occurs as a side effect of certain drugs such as immunosuppressive drugs, anticonvulsants and calcium channel blockers. Epilepsy is known to be closely related to cerebral palsy; 15-60% of children with cerebral palsy report having epilepsy requiring anticonvulsant drugs as routine therapy. When gingival enlargement occurs, it interferes with normal oral hygiene practices and can interfere with masticatory function. Gingival hyperplasia can be treated by stopping or changing drugs. In more severe cases, surgical treatment is required. A 6 year old boy patient with a history of epilepsy and cerebral palsy came with complaints of lumps in the gums of the right and left lower jaw. The patient started taking epilepsy medication about 3 years after being diagnosed with epilepsy. After about 1.5 years of taking epilepsy drugs, the patient experienced enlargement of all of his maxillary and mandibular gingiva. The patient was consulted to the Oral and Maxillofacial Surgery Department of Hasan Sadikin General Hospital, on intra-oral examination we found there was gingival enlargement that covered almost the entire tooth surface, the treatment plan was carried out by a surgical procedure in stages under general anesthesia. The results obtained after surgery showed that the shape of the gingiva was normal and the patient's masticatory function had improved. In cases where there is overgrowth of the gingiva to cover the entire tooth surface, surgical procedure is a standard procedure to restore the normal shape of the gingiva.

Keywords : *Gingival Hyperplasia, Gingival Enlargement, Epilepsy, Cerebral palsy*

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INTRODUCTION

Cerebral palsy (CP) is a group of clinical syndromes characterized by non-progressive central motor deficits caused by damage to the immature brain. CP is a non-specific descriptive term used for motor function disorders that arise in early childhood and are characterized by altered muscle tone (usually spasticity), involuntary movements, ataxia, or a combination of all these abnormalities that are neither episodic nor progressive. It most

commonly affects the extremities, but can also affect other parts of the body.¹

Patients with CP often get comorbidities, either as etiology or as complications of CP. Things that are often found as complications or comorbidities in CP are disorders of brain function, including epilepsy. Seizure attacks in CP patients vary and can be found at an early age. Seizure attacks are more frequent in severe CP. In CP with epilepsy, usually the onset of seizures at an early age and the need to use anti-epileptic drugs of more than one type and the

risk of relapse after discontinuation of anticonvulsant drugs is higher.^{2,3}

Gingival hyperplasia is an overgrowth of the gingiva (gum tissue) characterized by enlarged, inflamed and bleeding gums. Drug induced gingival overgrowth (DIGO) is an enlargement (hyperplasia) of the gingiva that occurs due to the side effects of taking certain drugs such as anticonvulsants, immunosuppressants, and calcium channel blockers (CCBs). Anticonvulsant (antiepileptic) drugs include valproic acid and carbamazepine. Valproic acid and carbamazepine are still the drugs of choice in cases of epilepsy despite their known side effects of gingival enlargement.^{4,5}

Gingival enlargement is usually present in 50% of patients taking the drug and usually occurs in young patients. Excessive gingival growth is related to drug dose, duration of treatment, and the presence of plaque. The clinical features of gingival enlargement induced by valproic acid and carbamazepine are that the marginal gingiva and interdental papilla appear enlarged and rubbery, the surface appears smooth, accompanied by stippling or lobed, and may also be accompanied by slight inflammation or no inflammation.^{4,6}

METHODS

The research employed a qualitative research design, aiming to delve deeply into the contextual conditions by providing a detailed and comprehensive description of the circumstances within a natural setting. This approach seeks to uncover the reality of what occurs within the study field. The chosen research method was a case study.¹⁸

The data collection method employed involves utilizing literature and secondary data sources. Literature review entails gathering information and data from diverse materials available in the library, including reference books, previous research findings, articles, notes, and relevant journals pertaining to the research problem. Following data acquisition, analysis proceeds through three stages: data reduction, data presentation, and drawing conclusions.¹⁹

RESULT AND DISCUSSION

Case Report

A 6-year-old boy came to the Department of Oral and Maxillofacial Surgery, Hasan

Sadikin Hospital (RSHS), Bandung with a complaint of a lump on the right and left lower jaw gums. The patient had a history of Cerebral palsy Type Spastic Quadriplegia and began routinely taking valproic acid and carbamazepine after being diagnosed with epilepsy from a pediatrician 3 years ago (Figure 1). The patient had enlarged gingiva in both maxilla and mandible after 1.5 years of epilepsy medication. The patient's parents complained about the contours of their child's gingiva which had enlarged to cover almost the entire tooth surface, making it difficult for the patient to chew. The patient then came to RSHS for further treatment.

Intraoral examination showed enlarged upper and lower jaw gingiva covering almost the entire tooth surface, pink gingival color, rubbery consistency, not easy to bleed (Figure 2). Then the patient was planned for excisional biopsy under general anesthesia in stages to minimize the risks arising from surgery (Figure 3 and Figure 4).



Figure 1. Pre-operative profile photo

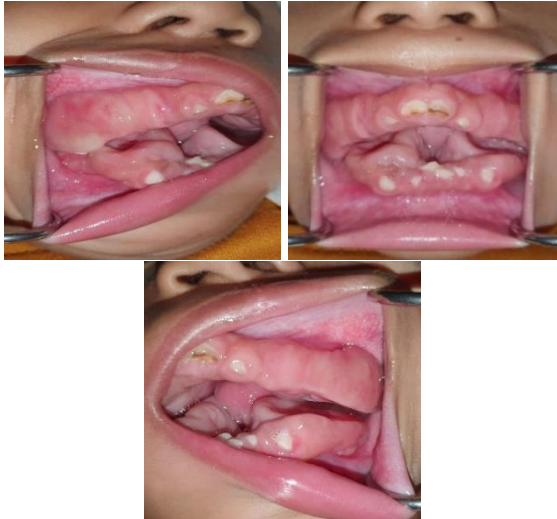


Figure 2. Intraoral enlargement of the maxillary and mandibular gingiva covering 2/3 of the tooth surface.



Figure 3. Intra operative view of excisional biopsy performed on the right jaw, postoperative wound closed with periodontal pack.

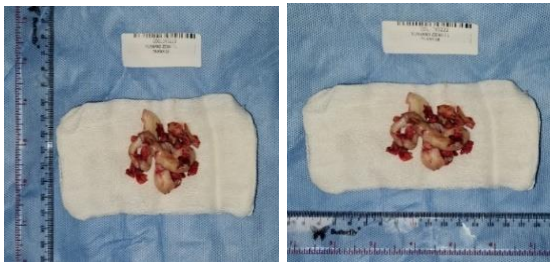


Figure 4. Photograph of gingival overgrowth tissue after excisional biopsy



Figure 5. Intraoral condition of the patient 1 day after excisional biopsy

The patient's intraoral state was monitored and the periodontal pack was maintained from day 1 to day 7 postoperatively (Figure 5). After 7 days postoperatively the patient came back for control. Complaints were denied and there was no bleeding. Then the periodontal pack was removed, the gingival contour was found to be smaller on the operated side of the jaw (Figure 6). The anatomical pathology examination concluded Diffuse fibrous hyperplasia a.r gingiva of teeth 51-55, 81-85. Furthermore, observation was carried out on the patient's gingival condition and planned to perform excision on the left jaw side 3 months later.



Figure 6. Intraoral condition of the patient 7 days after excisional biopsy. The maxillary and mandibular right gingival contours were visibly reduced.

Discussion

Cerebral palsy (CP) is a chronic condition characterized by non-progressive impairment of posture and movement. Spasticity leads to impairments in posture, movement control, balance and coordination, which ultimately affects the functional activities of a child with CP. It is also referred to as Little's disease as the term was first introduced by William John Little in 1843, where he attributed the rigidity that occurs due to damage to the brain, whether it occurs in infancy, premature birth, or asphyxia at birth.²⁵ Children with cerebral palsy may have permanent nerve damage, causing impairments in gross motor function, fine motor function, speech, and other aspects as cerebral palsy impairs coordination of functions. Variations of cerebral palsy are based on the location of the brain damage, which includes spastic, ataxic, athetoid, and hypotonic types. Meanwhile, based on the area of the body affected, the condition is classified into diplegia, monoplegia, hemiplegia, quadriplegia, and tetraplegia.^{7,8}

Cerebral palsy is a progressive non-developing condition that can cause movement

restrictions and postural changes in children, potentially affecting all psychosocial aspects and, therefore, impacting on the child's quality of life. Evaluation of quality of life is important to assess the most appropriate interventions for children with cerebral palsy. Cerebral palsy (CP) is a disorder characterized by muscle tension, unnatural postures and abnormal movements. The condition is clinically categorized based on the most predominant motor syndrome, such as spastic hemiplegia, spastic diplegia, spastic quadriplegia, and extrapyramidal or dyskinetic.^{16,17}

In a previous study conducted by Zufahmi and colleagues, the results showed that after undergoing physiotherapy treatment three times a week for four weeks, there was an improvement in motor skills as measured by Gross Motor Functional Measurement. The improvement was mainly seen in the ability to sit, crawl, and stand. This study provides strong support for the effectiveness of physiotherapy as an intervention that can help overcome motor disorders in patients with Cerebral Palsy.²⁰

Children with Cerebral Palsy generally experience a range of limitations, including impairments in sensation, cognition, communication, spasticity, perception and behavior. These changes can limit a child's motor abilities, which in turn will affect their ability to perform functional activities and daily living. Children with cerebral palsy can be classified based on their level of physical motor impairment, which includes the ability to walk without support, using assistive devices, or not being able to walk at all. These physical motor skills also relate to small muscle movement and fine motor skills, which have far-reaching effects. Fine motor skills are influenced by gross motor skills, and because of their link to a child's health and coordination of movements, factors such as health and fitness can also affect coordination between motor movements.^{21,22}

Quadriplegia cerebral palsy is a stiffness that affects both limbs, namely both hands and both feet. It is the most severe form of cerebral palsy as it involves many areas of the body. Children with spastic quadriplegia cerebral palsy are generally unable to walk, and they often experience some associated conditions, such as speech difficulties or seizures. Epilepsy is commonly present in patients with cerebral palsy, occurring in 30%-40% of cases. Epilepsy tends to be more common in CP children with quadriplegia (75%), ataxia (83%), and mixed

forms (80%) compared to diplegia (32%) and hemiplegia (38%).^{8,9}

Gingival enlargement as a side effect of drug use is one of the problems often encountered in the oral cavity of patients with certain conditions such as Cerebral Palsy. In Cerebral Palsy patients who also suffer from epilepsy, routine use of anticonvulsant drugs can change the shape of the gingiva to be larger than normal, which is clinically visible. Factors leading to gingival enlargement can be local, such as plaque accumulation accompanied by poor oral hygiene. Additionally, systemic factors can also play a role, where the consumption of certain medications can trigger a condition known as drug-induced gingival overgrowth (DIGO).^{9,10}

Drug-induced gingival overgrowth (DIGO) emerges as a side effect attributed to specific therapeutic medications, notably phenytoin, nifedipine, and cyclosporine A. Primary drug categories associated with DIGO comprise anticonvulsants, immunosuppressants, and calcium channel blockers. This review examines the etiology, pathophysiology, and diagnostic considerations of gingival overgrowth, emphasizing the collaborative efforts of the interprofessional team in its treatment.²³

Previous research by Rinawati S. and colleagues showed an association between anticonvulsant drug consumption and gingival enlargement. Management of gingival enlargement in the previous study involved patient education about the importance of maintaining oral hygiene and consultation with a neurologist for consideration of anticonvulsant drug replacement. Operative measures such as surgery have not been performed for fear of triggering epileptic seizures, so the results achieved have not been optimal. The patient is required to have regular control every month for further observation. However, in this case, the patient underwent operative measures in the form of gradual gingival excision under general narcosis, with the hope that the results achieved will be more optimal.

The severity of gingival enlargement is classified into Grade I: enlargement limited to the interdental papilla. Grade II: Enlargement covers the interdental papillae and gingival margin. Grade III: enlargement covers one third or more of the clinical crown of the tooth.¹¹ The enlargement in this case was caused by the

induction of valproic acid and carbamazepine. The gingival enlargement in this case is included in Grade III, which is gingival enlargement that covers one third or more of the clinical crown of the tooth.

Anticonvulsant medications exert their effects on a cellular level by impeding the influx of intracellular calcium ions. Similarly, the mechanism of action at the cellular level for the three distinct drug categories anticonvulsants, immunosuppressants, and calcium channel blockers involves the inhibition of cation influx, particularly sodium and calcium ions.^{10,12}

Gingival overgrowth is influenced by multiple factors. One potential cause is the accumulation of bacterial plaque, with the severity of overgrowth directly linked to the extent of plaque buildup and the resulting inflammation. Reduced activity of folic acid (FA)-dependent transport in gingival fibroblasts can lead to diminished FA uptake by cells. This interference with matrix metalloproteinase metabolism and inhibition of collagenase activation contribute to the accumulation of connective tissue and collagen, exacerbating the condition.^{13,14}

Anticonvulsant drugs inhibit the absorption of folic acid, thereby limiting the production of active collagenase. Fibroblasts activated by anticonvulsant drugs produce large amounts of interleukin (IL)-6, IL-1, and IL-8. These mediators can stimulate T cell proliferation and the presence of neutrophils in the tissue, creating a direct interaction between the immune system and connective tissue.^{12,15}

Management of patients with drug-induced gingival enlargement includes several steps: discontinuing or changing the drug, for example by changing carbamazepine and valproic acid to other anticonvulsant drugs; plaque control, where pseudo or false pocket formation often occurs due to plaque build-up; surgical therapy, which may include gingivectomy or gingival excision procedures.¹⁵

After initial therapy, there was an improvement in the gingival contour especially in the maxilla. Management of gingival overgrowth after surgery focuses on maintaining oral health to control gingival inflammation. The interaction between the drug and the gingival tissue may become worse due to gingival inflammation caused by poor oral care. Surgery on the enlarged tissue is often required to achieve good functional and aesthetic results. Surgical treatment may

include gingivectomy or gingival excision. Discontinuation of medication is intended to monitor the decrease or reduction in size of the gingival overgrowth, but if the medication is re-administered, it may lead to recurrence of the gingival overgrowth.^{12,16}

The aim of the surgical procedure in this case is to restore the gingival contour to normal so that optimal masticatory function can be achieved. To achieve the best outcome, it is important to consider drug discontinuation, but the decision should be discussed with the patient's treating paediatrician to ascertain whether it is possible to discontinue or replace valproic acid or carbamazepine with another anticonvulsant drug. This is a challenge in dealing with potential recurrences, as there is still the possibility of gingival tissue regrowth. Therefore, gradual evaluation and surgical procedures are required to observe gingival growth in the patient's oral cavity.

Management of Drug Induced Gingival Overgrowth (DIGO) in patients with spastic quadriplegia cerebral palsy involves careful evaluation of the risks and benefits of various therapeutic approaches. The use of drugs such as cyclosporine, phenytoin and nifedipine, which are the main triggers of DIGO, should be carefully considered. While such medications may be necessary to manage the patient's condition, their adverse impact on gum health should be anticipated. Preventive measures, such as dose adjustments or replacing medications with gum-safer alternatives, can reduce the risk of DIGO. However, it is important to remember that discontinuing such medications may not always be possible and may negatively impact the management of the patient's underlying disease. Therefore, a holistic and coordinated approach between dentists, cerebral palsy specialists, and medical doctors should be implemented to assess and manage the risk of DIGO while maintaining essential therapeutic benefits for the patient.

CONCLUSION

The management of DIGO depends on the severity of the gingival hyperplasia. In this case, gingival hyperplasia was categorized at grade 3, where the surgical procedure improved masticatory function and provided good aesthetics. Further action is required to achieve a normal gingival contour in the patient's entire jaw.

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