Submitted: 07 February, 2025 Accepted: 18 April, 2025 Published: 03 November, 2025 DOI:10.22514/jocpd.2025.145

CASE REPORT

Journal of Clinical Pediatric Dentistry

Repeated facial swelling caused by pericoronitis in a patient with megacystis microcolon intestinal hypoperistalsis syndrome: a case report

Chihiro Takasaki[1,*], Toshihiro Yoshihara[1], Aya Matsuda[2], Yasutaka Yawaka[1]

[1]Dentistry for Children and Disabled Persons, Department of Oral Functional Science, Faculty of Dental Medicine, Hokkaido University, 060-8586 Sapporo, Japan
[2]Vascular Biology and Molecular Pathology, Department of Oral Pathobiological Science, Faculty of Dental Medicine, Hokkaido University, 060-8586 Sapporo, Japan

***Correspondence**
chihiro@med.hokudai.ac.jp
(Chihiro Takasaki)

Abstract

Background: Facial swelling due to pericoronitis associated with tooth eruption commonly involves the wisdom teeth, but can rarely involve deciduous teeth or permanent premolars. We report a very rare case of repeated facial swelling caused by pericoronitis associated with the eruption of deciduous molars and a premolar. **Case**: The patient was a 13-year 7-month-old Japanese girl with megacystis microcolon intestinal hypoperistalsis syndrome (MMIHS), gingival fibromatosis and numerous impacted teeth. She visited our clinic for investigation and treatment of left facial swelling. Extraoral examination revealed swelling of the left mandibular angle accompanied by a feeling of heat, and two submandibular lymph node masses were palpable. Intraoral examinations revealed that the lower left deciduous molars had started erupting and the gingiva on the buccal side of the deciduous molars was red and swollen, while gingiva still covered the first premolar. We diagnosed facial swelling due to pericoronitis of the lower left deciduous molars and first premolar. We disinfected the periodontal pockets, then injected antibacterial ointment into them. Systemic antibiotic was also administered and facial swelling disappeared. However, the patient stopped taking the antibiotic and swelling of the left face recurred. We therefore resected the gingiva covering the deciduous molars and first premolar to expose the crowns after the inflammatory response subsided. No further swelling has occurred since the tooth crowns were exposed. **Conclusions**: Gingival excision exposing tooth crowns after the inflammatory response subsides appears effective for improving facial swelling associated with tooth eruption in patients with MMIHS, by removing the bacterial flora causing inflammation.

Keywords

Facial swelling; Pericoronitis; Gingival fibromatosis; Megacystis microcolon intestinal hypoperistalsis syndrome (MMIHS)

1. Introduction

Pericoronitis is an inflammation of the gingiva and soft tissues surrounding partially erupted teeth [1]. This inflammation commonly involves the wisdom teeth, but can rarely involve the premolars or primary teeth [2]. Pericoronitis is caused by bacterial overgrowth in a confined space, exacerbated by reduced cleaning of the periodontal pockets [3]. The periodontal pockets in patients with pericoronitis harbor lower levels of periodontal pathogens than those in patients with periodontitis, and the microbiota in pericoronitis is highly diverse [4]. As severe pericoronitis causes facial swelling, a common treatment to relieve facial swelling is antibiotics, extraction of the offending tooth or gingivectomy after the clinical symptoms subside [1, 5].

Megacystis microcolon intestinal hypoperistalsis syndrome (MMIHS) is a rare congenital gastrointestinal disorder with poor prognosis [6–10]. The syndrome is characterized by microcolon, decreased intestinal peristalsis and distended non-obstructive bladder [7]. Since we reported oral findings from a patient with MMIHS in 2018 [11], similar findings do not appear to have been reported. We describe here a case with repeated facial swelling due to pericoronitis involving deciduous molars and a premolar in a patient with MMIHS, and outline the steps we took for treatment.

2. Case report

A 13-year 7-month-old Japanese girl with MMIHS visited Dentistry for Children and Disabled Persons at Hokkaido University Hospital Dental Clinic in Sapporo, Japan. Her chief complaint was left lower facial swelling that had started the previous day. She also showed gingival fibromatosis, multiple impacted teeth, mental retardation, motor retardation and

epilepsy. She has been taking levetiracetam, an anti-epileptic agent, since she was 4 years 5 months old. Levetiracetam has not been reported to have gingival proliferative effects. She had no history of taking any other anti-epileptic drugs with gingival proliferative effects. The gingiva appeared hypertrophic at birth. Her family medical history was unremarkable. She was always fed via nasogastric tube. She was unable to independently brush her teeth adequately due to mental and motor retardation, but her oral cavity was cleaned daily by her mother. Extraoral examination revealed swelling of the left mandibular angle accompanied by a feeling of heat, and two submandibular lymph node masses were palpable, each the size of a broad bean (Fig. 1A,B). Body temperature was normal. Intraoral examination revealed that the lower left deciduous molars, which had been completely impacted since birth, had started erupting (Fig. 1C, arrows). In addition, the gingiva on the buccal side of the second deciduous molar appeared red and swollen (Fig. 1C, arrowhead). X-ray imaging showed that the dental crown of the lower left first premolar overlapped with the crowns of the deciduous molars (Fig. 1D). The oral examination and X-ray image showed that the first premolar was present on the buccal side of the deciduous molars, covered by the red and swollen gingiva. The depth of the periodontal pocket for the lower left second deciduous molar was 5 mm. The patient reported pain in the gingiva around the lower left molars during brushing over the past few days. We diagnosed facial swelling due to pericoronitis of the lower left deciduous molars and first premolar. The periodontal pockets of the deciduous molars and first premolar were disinfected using cotton plugs with iodine tincture, then antibacterial ointment containing minocycline hydrochloride was manually injected into the pockets using a syringe with nozzle. After administration of amoxicillin three times a day after meals for one week through a transudate tube, facial swelling disappeared. However, swelling of the left face recurred three weeks after stopping amoxicillin (Fig. 2A). Facial swelling recurred three times over a 3-month period. Once the inflammatory response subsided, we incised and resected the gingiva covering the deciduous molars and first premolar to expose the crowns (Fig. 2B). First, surface anesthesia (20% ethyl aminobenzoate dental jelly) was applied to the gingiva around the lower left molar, then a local anesthetic agent

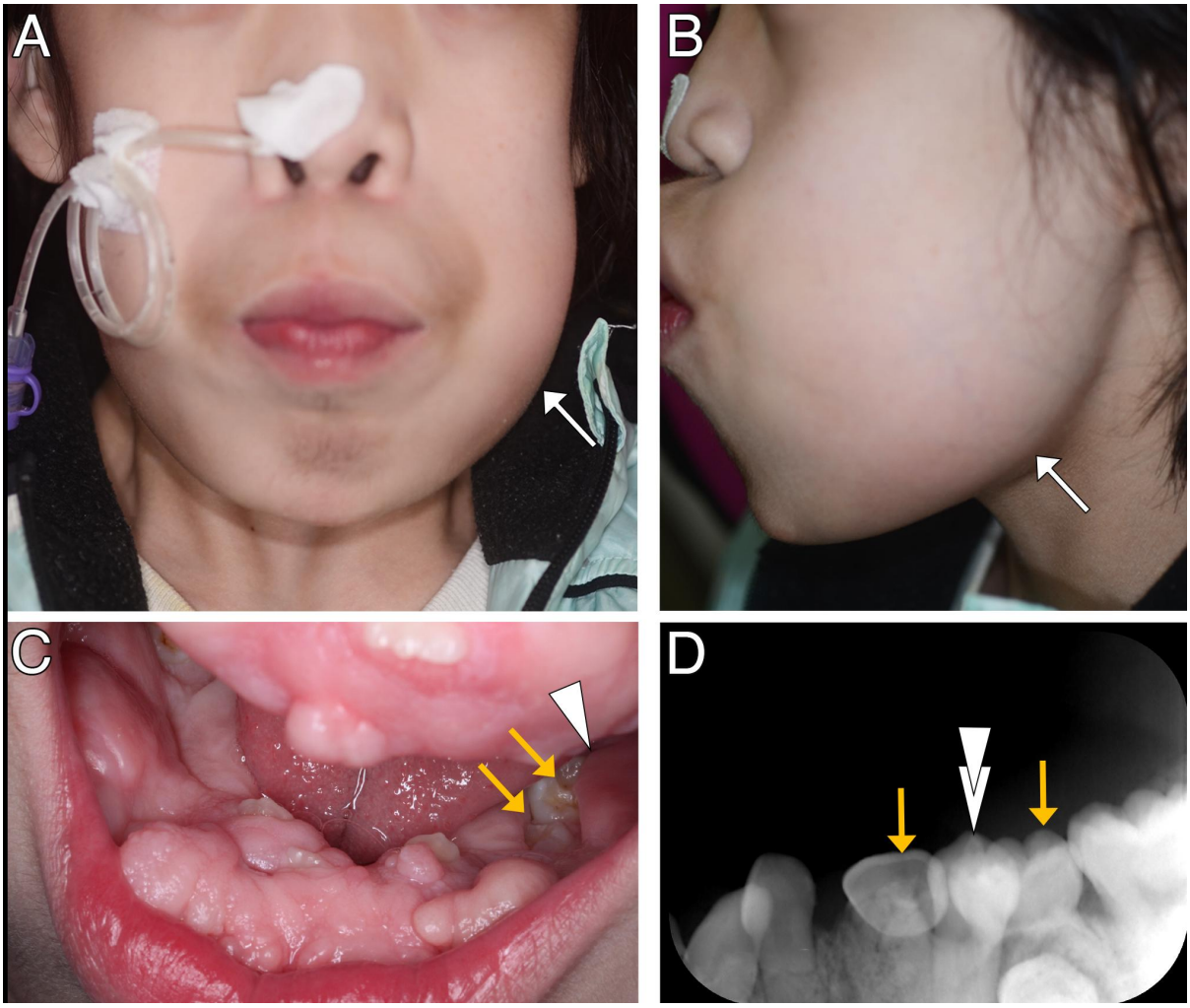

FIGURE 1. Clinical and X-ray photographic findings at first facial swelling. (A) Frontal view of the face. (B) Lateral view of the face. (C) Intraoral view. (D) X-ray image. White arrow indicates facial swelling. Yellow arrows indicate lower left deciduous molars. Arrowhead indicates redness and swelling of the gingiva in (C). Double arrowhead indicates the lower left first premolar in (D).

(2% lidocaine with 1:80,000 epinephrine) was injected into the gingiva. Next, to remove the gingiva covering the distal buccal side of the lower left second deciduous molar, incision lines were made from lingual to buccal and from distal to proximal along the crown of the second deciduous molar, and the marginal gingiva was removed. Incision lines were then made along the buccal aspect of the first premolar from distal to proximal side, and the marginal gingiva was removed to expose the crown of the first premolar. Finally, incision lines were made along the lingual crown surface of the first deciduous molar, and the marginal gingiva was removed to expose the crown of the first deciduous molar.

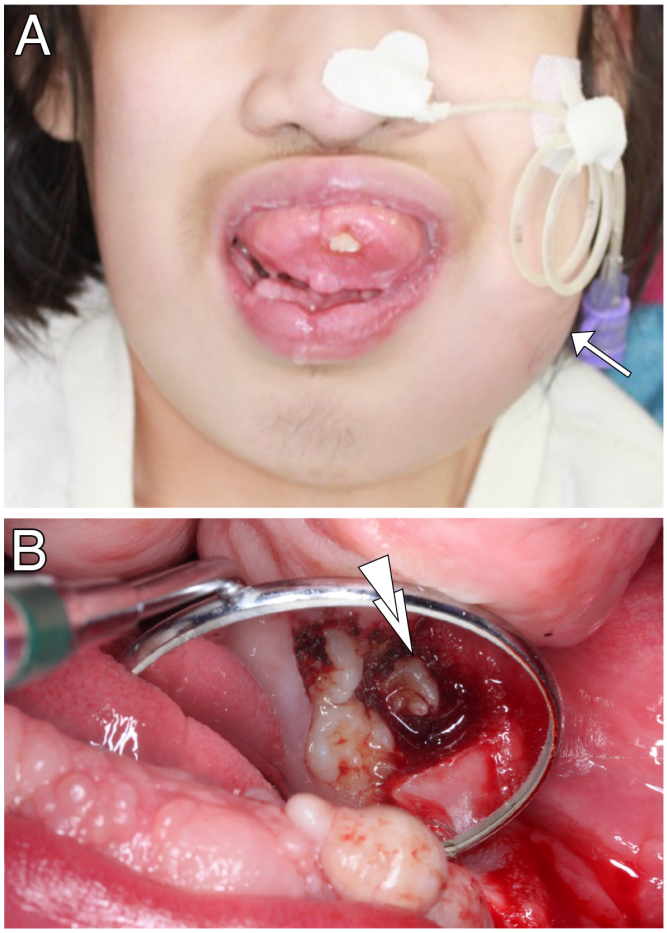

FIGURE 2. Clinical findings with the third episode of facial swelling (A) and at surgery (B). (A) Recurrent facial swelling. (B) Mirror image of the intraoral view immediately after surgery. White arrow indicates facial swelling. Double arrowhead indicates the lower left first premolar with central tubercle.

Histopathological examination showed non-specific inflammation of the resected gingiva derived from pericoronitis of the lower left second deciduous molar (Fig. 3). The lower left first premolar showed an unbroken tubercle on the occlusal surface. We covered the tubercle with glass ionomer cement to prevent pathological exposure of the pulp due to fracture or attrition. As of 2 years 11 months postoperatively, we have been providing oral care every 3 months. The patient has shown no further swelling of the face since the crowns of the teeth were exposed (Fig. 4A,B). Oral care has also continued to be provided daily by her mother.

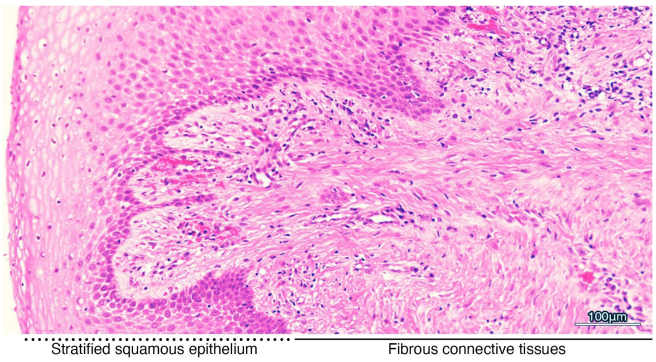

FIGURE 3. Histological image of the resected gingiva at the time of surgery (hematoxylin and eosin, original magnification ×4). Infiltration of inflammatory cells is apparent. Dotted line indicates stratified squamous epithelium. Solid line indicates fibrous connective tissues. Scale bar; 100 μm.

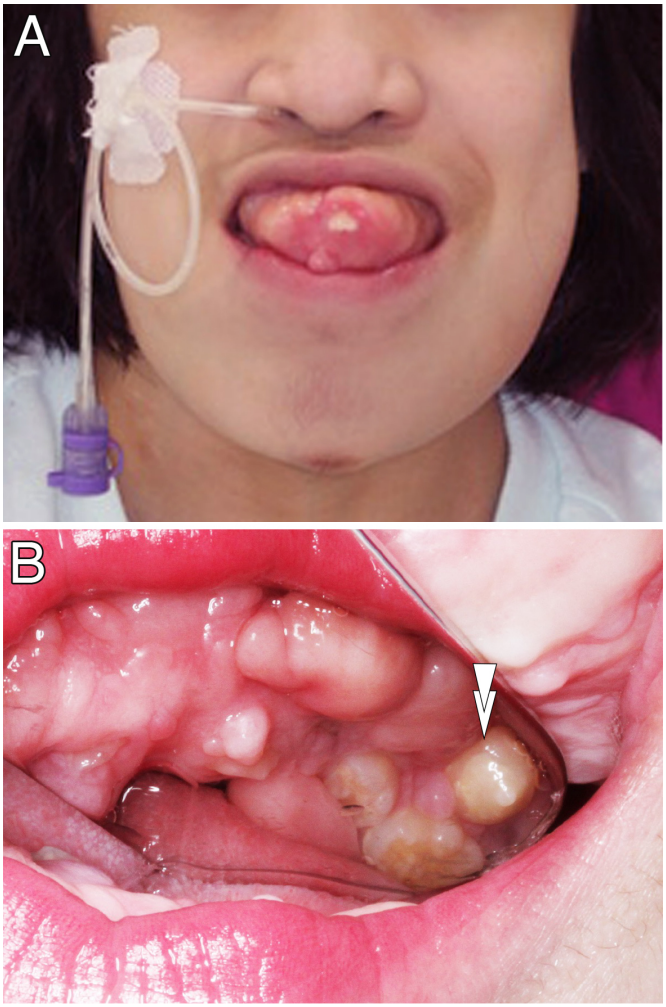

FIGURE 4. Clinical findings more than 1 year after surgery. (A) Facial view. (B) Mirror image of the intraoral view. Double arrowhead indicates the lower left first premolar. A tubercle on the occlusal surface is covered with glass ionomer cement.

3. Discussion

In the present report, we described a rare case of facial swelling due to pericoronitis caused by deciduous molars and a premolar in a patient with MMIHS and gingival fibromatosis. Tooth-related facial swelling in children is often due to severe caries or fracture of a central tubercle, but rarely due to pericoronitis associated with tooth eruption [2, 12–16]. The facial swelling in this case was due to pericoronitis of the deciduous molars and a premolar without a broken central tubercle. The optimal treatment was considered to be resection of the gingiva covering the tooth crowns.

Gingival fibromatosis is a rare disease that involves slowly progressive, local or diffuse enlargements of gingiva as a non-syndromic hereditary gingival fibromatosis, part of a syndrome, or a side effect of certain pharmacotherapies [17–19]. Gingival fibromatosis was also present in this case. As we have reported previously, gingival fibromatosis in this case was probably part of a syndrome, because gingival proliferation had been observed more than 4 years before the patient started taking anti-epileptic drugs [11]. The fact that no one in her family had gingival fibromatosis also ruled out hereditary gingival fibromatosis. Gingival fibromatosis has been reported to occur in several syndromes, including Cowden syndrome, Jones syndrome, Murray-Puretic-Drescher syndrome, Ramon syndrome, Rutherfurd syndrome and Zimmermann-Laband syndrome [19–21]. Although there have not been any definitive reports on MMIHS with gingival fibromatosis, it is suggested that some MMIHS cases are accompanied by gingival fibromatosis [11]. Among patients with gingival fibromatosis, aggressive periodontitis has been reported on rare occasions [22–26], but no reports appear to have described patients suffering from pericoronitis with associated facial swelling. Bacterial infection is the main cause of periodontal disease [26, 27]. In this case, significant gingival hyperplasia was present and prevented easy plaque removal by applying a toothbrush to the gingival sulcus of the erupting molars. Inflammation was considered to be spreading from bacteria retained in the gingival sulcus, leading to swelling of the submandibular lymph nodes. During the eruption of teeth in patients with gingival fibromatosis and systemic disease, removal of plaque from the gingival sulcus must be performed more carefully or mechanically at a dental clinic if the teeth cannot be sufficiently brushed at home. After gingivectomy, it is important for patients with gingival fibromatosis to regularly visit the dental clinic for plaque control, and correct oral hygiene is considered the basis for preventing relapse, with sites of poor oral hygiene showing faster relapse [28–31].

Histopathological examination showed non-specific inflammation of the resected gingiva derived from pericoronitis, consistent with inflammation due to pericoronitis. Clinically, these histopathological findings would manifest as pain, gingival redness, and swelling, all of which were observed in the present case.

In this case, amoxicillin as a systemic antibiotic was used to relieve the symptoms of facial swelling. This was effective, but the facial swelling recurred after the patient stopped taking the antibiotic. According to a report that investigated whether systemic antibiotics should be prescribed for pericoronitis, no single randomized or non-randomized controlled trials have assessed the harms and clinical effectiveness of systemic antibiotics in adults with pericoronitis [32]. In children, appropriate prescription of antibiotics for orofacial infections in pediatric outpatient settings has been reported [33]. Amoxicillin was shown to be the most commonly recommended antibiotic for short durations of 3–5 days, with metronidazole or azithromycin as alternative antibiotics for penicillin-sensitive patients [33]. Proper use of antibiotics is necessary to prevent antimicrobial resistance. When systemic administration of antibiotics alone fails to achieve complete cure, other treatment options need to be considered, such as gingivectomy in the present case.

Many patients with MMIHS die in infancy from sepsis [8]. This syndrome may thus predispose patients to infection. Some points about immune function in patients with MMIHS remain unclear. MMIHS is a variant of Hirschsprung's disease [34] and the composition of the microbiota in patients with Hirschsprung's disease is known to differ from that in healthy individuals, showing a decreased richness of the overall microbiota [35]. In recent years, intestinal bacteria have been reported to affect the host immune system [36–40]. We acknowledge that this discussion is based on general knowledge of MMIHS and remains limited by the fact that no specific immunological evaluations were performed in this case. Healthy children rarely experience facial swelling due to gingival inflammation [2]. Patients with MMIHS may thus have reduced immune function compared to healthy children, and the oral and intestinal flora of patients with MMIHS may differ from those of healthy subjects. In the present case, we did not examine the bacterial groups present in the periodontal pockets or gut. One challenge for the near future is thus to clarify the immune function and bacterial flora of MMIHS patients. Evaluating the bacterial flora may enable early detection of risks for infection, inflammation, and digestive disorders, prediction of disease risk and implementation of preventive measures. Children with MMIHS are currently very rare, but will live longer as medical care advances. Patients with this disease are thus likely to be encountered more frequently in the near future. Collaboration between pediatricians, gastroenterologists, and dentists may provide a more comprehensive approach to the care of patients with MMIHS. The present case report may help improve oral management for patients with MMIHS.

4. Conclusions

Facial swelling due to pericoronitis of deciduous molars and a premolar is rare. Resection of the gingiva covering the tooth crowns may be a useful option to improve facial swelling due to pericoronitis of deciduous molars and premolars.

ABBREVIATIONS

MMIHS, megacystis microcolon intestinal hypoperistalsis syndrome.

AVAILABILITY OF DATA AND MATERIALS

All data generated during this case report are included in this published article.

AUTHOR CONTRIBUTIONS

CT, TY and YY—contributed to the drafting of the manuscript. CT and YY—contributed to the oral examination and dental treatment. AM—contributed to the pathological analysis. All authors approved the version submitted for publication. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

As a case report, this study was exempted from requiring ethics approval by the Ethical Review Board for Life Science and Medical Research at Hokkaido University Hospital. Informed consent for publication of clinical details and images was obtained from the mother of the patient.

ACKNOWLEDGMENT

The authors thank FORTE Science Communications (https://www.forte-science.co.jp/) for English language editing.

FUNDING

This research received no external funding.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

REFERENCES

[1] Douglass AB, Douglass JM. Common dental emergencies. American Family Physician. 2003; 67: 511–516.

[2] Folayan MO, Ozeigbe EO, Onyejaeka N, Chukwumah NM, Oyedele T. Non-third molar related pericoronitis in a sub-urban Nigeria population of children. Nigerian Journal of Clinical Practice. 2014; 17: 18–22.

[3] Kwon G, Serra M. Pericoronitis. StatPearls Publishing: Treasure Island (FL). 2025.

[4] Ribeiro MHB, Ribeiro PC, Retamal-Valdes B, Feres M, Canabarro A. Microbial profile of symptomatic pericoronitis lesions: a cross-sectional study. Journal of Applied Oral Science. 2020; 28: e20190266.

[5] Sands T, Pynn BR, Nenniger S. Third molar surgery: current concepts and controversies. Part 1. Oral Health. 1993; 83: 11–14, 17.

[6] Berdon WE, Baker DH, Blanc WA, Gay B, Santulli TV, Donovan C. Megacystis-microcolon-intestinal hypoperistalsis syndrome: a new cause of intestinal obstruction in the newborn. Report of radiologic findings in five newborn girls. American Journal of Roentgenology. 1976; 126: 957–964.

[7] Puri P, Shinkai M. Megacystis microcolon intestinal hypoperistalsis syndrome. Seminars in Pediatric Surgery. 2005; 14: 58–63.

[8] Gosemann JH, Puri P. Megacystis microcolon intestinal hypoperistalsis syndrome: systematic review of outcome. Pediatric Surgery International. 2011; 27: 1041–1046.

[9] Mc Laughlin D, Puri P. Familial megacystis microcolon intestinal hypoperistalsis syndrome: a systematic review. Pediatric Surgery International. 2013; 29: 947–951.

[10] Wymer KM, Anderson BB, Wilkens AA, Gundeti MS. Megacystis microcolon intestinal hypoperistalsis syndrome: case series and updated review of the literature with an emphasis on urologic management. Journal of Pediatric Surgery. 2016; 51: 1565–1573.

[11] Takasaki C, Yoshihara T, Yawaka Y. Oral findings in a patient with megacystis microcolon intestinal hypoperistalsis syndrome: a case report. Pediatric Dentistry Journal. 2018; 28: 57–61.

[12] Janakarajah N, Sukumaran K. Orbital cellulitis of dental origin: case report and review of the literature. British Journal of Oral and Maxillofacial Surgery. 1985; 23: 140–145.

[13] Chen CM, Lee KT, Chuang FH, Hong YY, Chen HC, Hsu KR, *et al.* Facial cellulitis arising from dens evaginatus: a case report. The Kaohsiung Journal of Medical Sciences. 2005; 21: 333–336.

[14] Michael JA, Hibbert SA. Presentation and management of facial swellings of odontogenic origin in children. European Archives of Paediatric Dentistry. 2014; 15: 259–268.

[15] Abdul Satar H, Yaakub A, Md Shukri N, Ahmad Tajudin LS. Orbital cellulitis secondary to dental abscess in children. Cureus. 2021; 13: e14392.

[16] Słotwińska-Pawlaczyk A, Orzechowska-Wylęgała B, Latusek K, Roszkowska AM. Analysis of the clinical status and treatment of facial cellulitis of odontogenic origin in pediatric patients. International Journal of Environmental Research and Public Health. 2023; 20: 4874.

[17] Anavi Y, Lerman P, Mintz S, Kiviti S. Idiopathic familial gingival fibromatosis associated with mental retardation, epilepsy and hypertrichosis. Developmental Medicine & Child Neurology. 1989; 31: 538–542.

[18] Bittencourt LP, Campos V, Moliterno LF, Ribeiro DP, Sampaio RK. Hereditary gingival fibromatosis: review of the literature and a case report. Quintessence International. 2000; 31: 415–418.

[19] Gawron K, Łazarz-Bartyzel K, Potempa J, Chomyszyn-Gajewska M. Gingival fibromatosis: clinical, molecular and therapeutic issues. Orphanet Journal of Rare Diseases. 2016; 11: 9.

[20] Takagi M, Yamamoto H, Mega H, Hsieh KJ, Shioda S, Enomoto S. Heterogeneity in the gingival fibromatoses. Cancer. 1991; 68: 2202–2212.

[21] Hart TC, Pallos D, Bozzo L, Almeida OP, Marazita ML, O'Connell JR, *et al*. Evidence of genetic heterogeneity for hereditary gingival fibromatosis. Journal of Dental Research. 2000; 79: 1758–1764.

[22] Casavecchia P, Uzel MI, Kantarci A, Hasturk H, Dibart S, Hart TC, *et al*. Hereditary gingival fibromatosis associated with generalized aggressive periodontitis: a case report. Journal of Periodontology. 2004; 75: 770–778.

[23] Chaturvedi R. Idiopathic gingival fibromatosis associated with generalized aggressive periodontitis: a case report. Journal of the Canadian Dental Association. 2009; 75: 291–295.

[24] Japatti S, Bhatsange A, Reddy M, Patil S, Chidambar, Waghmare A. Bimaxillary unilateral gingival fibromatosis with localized aggressive periodontitis (eating the tooth at the same table). Contemporary Clinical Dentistry. 2013; 4: 366–370.

[25] Gupta ND, Goyal L. Recurrent idiopathic gingival fibromatosis with generalized aggressive periodontitis: a rare case report. Journal of Indian Society of Periodontology. 2015; 19: 610.

[26] Darveau RP. Periodontitis: a polymicrobial disruption of host homeostasis. Nature Reviews Microbiology. 2010; 8: 481–490.

[27] Arweiler NB, Netuschil L. The oral microbiota. Advances in Experimental Medicine and Biology. 2016; 902: 45–60.

[28] Arduino PG, D'Aiuto F, Cavallito C, Carcieri P, Carbone M, Conrotto D, *et al*. Professional oral hygiene as a therapeutic option for pediatric patients with plasma cell gingivitis: preliminary results of a prospective case series. Journal of Periodontology. 2011; 82: 1670–1675.

[29] Ferreira Gonçalves C, Mundim AP, Martins RFS, Gagliardi RM, Santos PSS, Ayrton de Toledo O. Hereditary gingival fibromatosis: a case report with seven-year follow-up. Acta stomatologica Croatica. 2018; 52: 254–258.

[30] Bello L, Romano F, Gaido C, Defabianis P. The effect of an oral spray containing an aqueous extract of Triticum vulgare on dental plaque and gingival inflammation in schoolchildren: a randomized controlled trial. European Journal of Paediatric Dentistry. 2020; 21: 110–114.

[31] Berta G, Romano F, Vallone R, Abbadessa G, Di Scipio F, Defabianis P.

An innovative strategy for oral biofilm control in early childhood based on a resveratrol-cyclodextrin nanotechnology approach. Materials. 2021; 14: 3801.

[32] Leroy R, Bourgeois J, Verleye L, Toma S. Should systemic antibiotics be prescribed in periodontal abscesses and pericoronitis? A systematic review of the literature. European Journal of Oral Sciences. 2022; 130: e12884.

[33] Dar-Odeh N, Fadel HT, Abu-Hammad S, Abdeljawad R, Abu-Hammad OA. Antibiotic prescribing for oro-facial infections in the paediatric outpatient: a review. Antibiotics. 2018; 7: 38.

[34] Friedmacher F, Puri P. Classification and diagnostic criteria of variants of Hirschsprung's disease. Pediatric Surgery International. 2013; 29: 855–872.

[35] Neuvonen MI, Korpela K, Kyrklund K, Salonen A, de Vos W, Rintala RJ, *et al*. Intestinal microbiota in Hirschsprung disease. Journal of Pediatric Gastroenterology and Nutrition. 2018; 67: 594–600.

[36] Rooks MG, Garrett WS. Gut microbiota, metabolites and host immunity. Nature Reviews Immunology. 2016; 16: 341–352.

[37] Lin L, Zhang J. Role of intestinal microbiota and metabolites on gut homeostasis and human diseases. BMC Immunology. 2017; 18: 2.

[38] Fung TC, Olson CA, Hsiao EY. Interactions between the microbiota, immune and nervous systems in health and disease. Nature Neuroscience. 2017; 20: 145–155.

[39] D'Amelio P, Sassi F. Gut microbiota, immune system, and bone. Calcified Tissue International. 2018; 102: 415–425.

[40] Dang AT, Marsland BJ. Microbes, metabolites, and the gut-lung axis. Mucosal Immunology. 2019; 12: 843–850.

How to cite this article: Chihiro Takasaki, Toshihiro Yoshihara, Aya Matsuda, Yasutaka Yawaka. Repeated facial swelling caused by pericoronitis in a patient with megacystis microcolon intestinal hypoperistalsis syndrome: a case report. Journal of Clinical Pediatric Dentistry. 2025; 49(6): 225-230. doi: 10.22514/jocpd.2025.145.