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Tumor Discovery

CASE REPORT

Pyogenic granuloma of maxillary median gingiva in a pediatric patient: A case report and literature review

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Abstract

Pyogenic granuloma (PG) is a painless pedunculated granulomatous lesion that develops on the skin and mucous membranes. PG in children is relatively rare. We herein report a case of PG of the maxillary median gingiva in a pediatric patient. The patient was a 6-year-old Japanese boy who presented with a painless mass with bleeding after exfoliation of the left upper deciduous central incisor. The lesion had undergone continuous, rapid growth for approximately 10 days. Intraoral findings revealed a painless, elastic, soft, and non-pedunculated mass, measuring 15 × 10 mm, in the left upper deciduous central incisor area. To differentiate it from malignancy, an excisional biopsy was performed on the patient under general anesthesia. Histological findings confirmed that the patient suffered from PG. At the time of writing this paper, the patient had been clinically followed up for the 18th month after the surgery, showing no signs of tumor recurrence.

Keywords: Pyogenic granuloma; Child; Gingiva

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1. Background

Pyogenic granuloma (PG) is a painless pedunculated granulomatous lesion that develops on the skin and mucous membranes. PG of the oral cavity commonly occurs in the lips, gingiva, and tongue, but its occurrence in children is relatively rare.[1] The etiology of PG has not yet been clearly explained in the literature. Differentiating PG from malignant tumors is necessary as they share a similar trait – rapid mass growth.

We herein report a case of PG of the maxillary median gingiva in a pediatric patient.

2. Case presentation

The patient was a 6-year-old Japanese boy who presented with a painless mass with bleeding after exfoliation of the left upper deciduous central incisor. The patient received anti-inflammatory treatment at a nearby dental clinic. Since the lesion exhibited continuous, rapid growth for approximately 10 days, the patient

was eventually referred to our department for further examination and treatment.

Regarding the intraoral findings, a painless, elastic, soft, and non-pedunculated mass, measuring 15 × 10 mm, was found in the left upper deciduous central incisor area (Figure 1). There was no remarkable personal or family medical history. Panoramic and dental X-ray showed no significant abnormalities. Computed tomography (CT) revealed no obvious bone resorption or destruction (Figure 2). Magnetic resonance imaging revealed a high signal intensity on T2-weighted short tau inversion recovery imaging and a well-defined mass measuring 13 × 10 mm with a contrast effect on T1-weighted imaging (Figure 3). Considering the above findings, the patient underwent an excisional biopsy under general anesthesia in the same month. The excision range was designed with a safety margin of approximately 1 mm around the tumor (Figure 4A), and the lesion was resected as a single mass with the right upper deciduous central incisor (Figure 4B and C). After resection, the healthy gingiva and periosteum at the margins of the resection area were sutured with an absorbable thread. Histologically, a granulomatous lesion with telangiectasia was observed, showing no signs of malignancy (Figure 5). The lesion was a highly vascularized fibrous connective tissue stroma exhibiting numerous dilated blood capillaries, which were of varying sizes and shapes and lined with a single layer of endothelial cells. Based on these findings, the final diagnosis was PG. At the time of writing this paper, the patient had been clinically followed up for the 18th month after the surgery, showing no signs of tumor recurrence. The gingival morphology was also restored to a healthy state. Normal eruption of the successional maxillary anterior teeth was observed (Figure 6).



Figure 1. Pre-operative intraoral findings. A mass measuring 15 × 10 mm was found in the left upper deciduous central incisor area.

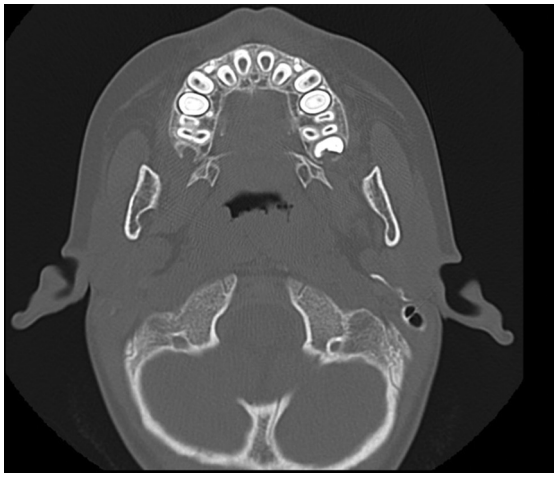


Figure 2. Computed tomography image. No bone resorption and destruction were observed in the upper deciduous central incisor area.

3. Discussion

PG is a granulomatous exophytic lesion that develops on skin and mucous membranes. PG of the oral cavity commonly occurs on the lips, gingiva, and tongue. It occurs more frequently in females than in males, with a ratio of 2:1.[2] The occurrence of PG is most common before the fifth decade of life.[3] The incidence of PG has risen tremendously in the last two decades.[2]

The occurrence of PG is relatively rare in children. To the best of our knowledge, only 26 cases of PG in children below 10 years old, including the current case, have been reported in the English literature. Most of the PG lesions develop on the gingiva, but two cases occurred on the upper lip and one case on the lower lip. The mean maximum diameter of the PG was 16.25 mm. The occurrence of PG was slightly more common in boys. The youngest documented patient who had been diagnosed with PG was 8 weeks old, with the lesion developing on the perigingiva of the congenital tooth. Table 1 presents

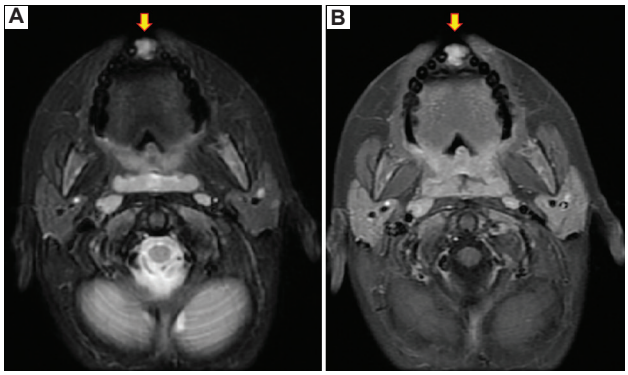


Figure 3. Magnetic resonance imaging images. (A) T2-weighted STIR imaging showed a high signal intensity (arrow). (B) Gadolinium contrast T1-weighted imaging showed a well-defined mass, measuring 13 × 10 mm, with a contrast effect (indicated by arrow).

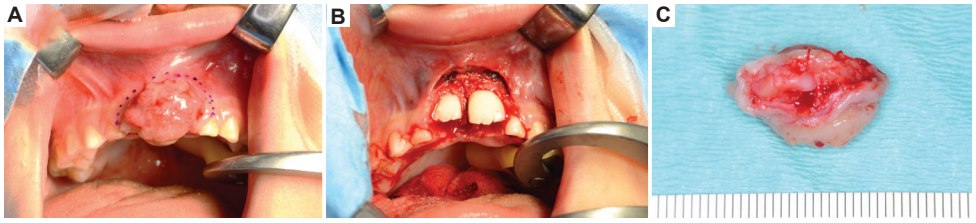


Figure 4. Operative findings. (A) The resection range was designed with a safety margin of approximately 1 mm around the tumor. (B) The lesion was resected in one lump together with the right upper deciduous central incisor. (C) The resected specimen.

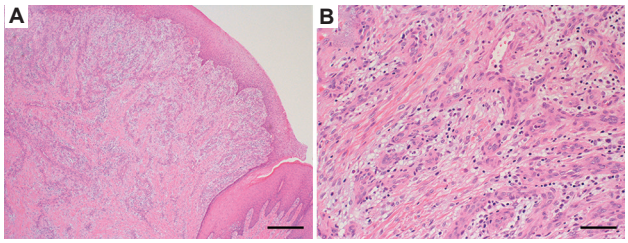


Figure 5. Histopathological findings of a granulomatous lesion with telangiectasia. (A) Low-magnification field (MM ×4); scale bar: 250 μm. (B) High-magnification field (MM ×20); scale bar: 50 μm.



Figure 6. Examination of the patient's oral cavity during follow-up at the 18th month after the surgery. There were no signs of local recurrence.

the baseline characteristics and clinical outcomes of 26 pediatric PG patients under 10 years old, of which 25 of them have been reported in the literature.[4-27]

PG is considered an inflammatory hyperplasia that is unrelated to infection and is caused by various stimuli, including low-grade local irritation, traumatic injury, hormonal factors, drugs, and bone marrow transplant.[28] Of the 26 cases mentioned above, Cheney-Peters and Lund reported the development of PG after bone marrow transplantation for pediatric hematological cancers.[17] Although the mechanism by which PG develops after bone marrow transplantation is unclear, previous reports suggest that inflammation, graft-versus-host disease, and calcineurin inhibitors, such as cyclosporine A and tacrolimus, probably play a role in its development.[29-34] Lindsay and Srivaths reported two cases of PG associated with hemophilia A, in which one case exhibited involuted presentation, with the lesion slowly resolving over the subsequent weeks, and the other demonstrating a lesion which was completely resolved following infusion of recombinant factor 8.[13] However, there is no confirmation on whether the diagnosis of PG was accurate because histopathological examinations were not performed. In addition, Cheney-Peters and Lund concluded that the occurrence of PG in hemophilia patients is grounded in tissue inflammation from vascular trauma and prolonged bleeding, and the resultant increase in circulating systemic inflammatory markers likely promotes the growth of these vascular lesions, which are prone to bleeding, thereby initiating a vicious cycle.[17] Thus, when PG occurs in the oral region, it is important to search for hidden systemic and local factors. In our case, there were few possibilities of PG originating from a systemic disease because there were no abnormalities in the pre-operative examination, and the patient has no relevant medical or family history. Further systemic examination, which was however not conducted, could have identified the etiology in this case.

A definitive diagnosis of PG can be made only with a histopathological examination. Histologically, it is a granulomatous lesion with inflammatory cell infiltration and vascular endothelial cell proliferation. The occurrence of PG is accompanied by the formation of numerous large and small endothelium-lined vessels, which sometimes form lobular aggregates. The arrangement of these lobular aggregates provides invaluable hints for pathologists to make an accurate diagnosis. In some cases, the vascular epithelium may be decimated due to ulceration or trauma or covered by a stratified squamous epithelium.

According to the International Society for the Study of Vascular Anomalies classification, PG lesions exhibiting lobular proliferation of capillaries and endothelial cells are classified as benign vascular tumors.[35] Epivatianos *et al.* reported that PG lesions can be categorized into lobular capillary hemangioma (LCH) and non-LCH types, depending on whether the capillaries proliferated

Table 1. Cases of PG in pediatric patients under 10 years old

Case no.	Year	Authors	Sex	Age	Location	Number of tumors	Treatment	Pathological diagnosis	Size (mm)	Number of recurrences	Final outcome
1	2000	Willies-Jacobo *et al.*[4]	M	Infant	Gingiva	1	Resection	PG	10×8×5	0	—
2	2001	Akyol *et al.*[5]	M	4 months	Tongue	1	Resection	PG	10×8×8	0	Good
3	2001	Milano *et al.*[6]	M	3 years	Gingiva	1	Excisional biopsy/Extraction	PG	—	0	Good
4	2005	Jurkiewicz *et al.*[7]	M	8 weeks	Tongue	1	Resection	PG	30×25	0	Good
5	2006	Shenoy and Dinkar[8]	F	8 years	Gingiva	1	Excisional biopsy	PG	20×10×10	0	Drop out
6	2008	de Souza *et al.*[9]	F	8 years	Lower lip	1	Excisional biopsy	PG	9×8×5	0	Good
7	2009	das Chagas *et al.*[10]	F	2 years	Upper lip	1	Excisional biopsy	PG	16	0	Good
8	2012	An *et al.*[11]	M	8 years	Gingiva	1	Excision	PG	25×20×15	0	Good
9	2013	Ximenes *et al.*[12]	M	4 year	Tongue	1	Excision	PG	8×6×4	0	Good
10	2014	Lindsay and Srivaths[13]	M	4 years	Tongue	1	Spontaneously involuted	None	—	0	Good
11	2014	Lindsay and Srivaths[13]	—	1.5 years	—	1	Recombinant factor 8 infusion	None	—	0	Good
12	2015	Jain *et al.*[14]	M	8 years	Gingiva	1	Excision (advised)	PG	30×30	0	—
13	2016	Agarwal *et al.*[15]	M	8 days	Gingiva	1	Excision	PG	8×5	0	Good
14	2016	Nirmala *et al.*[16]	M	8 years	Gingiva	1	Excision	PG	28×28×9	0	Good
15	2016	Cheney-Peters and Lund[17]	M	1.5 year	Tongue	2	Excision	PG	10×5,10×10	0	Good
16	2018	Peters *et al.*[18]	F	5 years	Tongue	1	Excisional biopsy	PG	6×12	0	Good
17	2019	Lim *et al.*[19]	F	3 years	Tongue	1	Resection	PG	40×30	1	Good
18	2019	Alaoui *et al.*[20]	M	10 years	Tongue	1	Excision	PG	9×6×5	0	Good
19	2020	Agel and Ahluwalia[21]	F	6 years	Gingiva	1	Excisional biopsy/extraction	PG	15×10	0	Good
20	2020	Ahsan Razi *et al.*[22]	M	10 years	Gingiva	1	Excisional biopsy	PG	17×8	0	Good
21	2020	Tazegül and Bodrumlu[23]	M	8 years	Gingiva	1	Excisional biopsy	PG	15	0	Good
22	2020	Tatar *et al.*[24]	M	10 years	Lower lip	1	Excision	PG	3	0	Drop out
23	2021	Kavitha *et al.*[25]	F	8 years	Gingiva	2	Excision	PG	15×10, 20×15	0	—
24	2022	Nassar and Besar[26]	—	10 years	Tongue	1	Excisional biopsy	PG	20×20	0	Good
25	2023	Gera *et al.*[27]	M	6 years	Gingiva	1	Excision	PG	5×10×8	0	Good
26	2023	The current case	M	6 years	Gingiva	1	Excisional biopsy/extraction	PG	8×6	0	Good

Abbreviations: F: Female; M: Male; PG: Pyogenic granuloma.

in a lobular shape based on clinicopathological and immunohistochemical characteristics, and that these two lesion types differ histologically per their pathogenesis.[36] Our case was categorized as non-LCH. However, Kawachi found that capillary hemangiomas and PG differ from each other because of their distinct expression of vascular endothelial growth factors.[37] They also concluded that capillary hemangiomas showed proliferating activity of capillaries, such as endothelial cells and pericyte-like perivascular cells, with the appearance of mast cells and lobules, whereas PG exhibited remarkable proliferative activity mainly of endothelial cells and inflammatory changes. However, it is currently unclear whether capillary hemangiomas and PG are of the same pathological classification.

The development of PG can be categorized according to their respective macroscopic findings into three distinct stages, namely (i) cellular phase, (ii) capillary phase/vascular phase, and (iii) involution phase.[38] Differential diagnoses in the oral region include benign reactive lesions, such as fibromas, hemangiomas, and peripheral giant cell granulomas, as well as malignant tumors, such as metastatic tumors of the oral soft tissue and Kaposi's sarcoma.[2,39] A histopathological examination is essential to distinguish rapidly growing PG from malignant tumors, as in the present case. In our case, an excisional biopsy was performed to avoid having to perform a second procedure if the tumor was not malignant.

Treatment options for PG include electrocautery, electrocoagulation, radiation therapy, and cryotherapy,[28,40] but resection of the lesion together with the surrounding healthy tissue has become a prevailing treatment approach in recent years. However, inadequate resection may result in rapid recurrence. The recurrence rate of PG has been reported to be 6% – 16%.[3,41] Accordingly, a radical cure of the initial resection is important. Furthermore, Asnaashari *et al.* concluded that using lasers for PG lesion removal could reduce stress and fear among pediatric patients and minimize discomfort both during and after surgery.[42] In the aforementioned 26 cases of pediatric PG, resection or excision was performed in most cases, with only one case reporting recurrence within the follow-up period.

For a PG lesion manifesting rapid growth, as in our case, a biopsy is required to rule out malignancy. However, physicians should anticipate tougher restrictions in terms of facilities, examinations, and treatments if the patient is a child. In addition, it is important for health-care personnel to cater to the mental health needs of the affected children and their families. In the present case, the patient was admitted to our hospital because pediatric hospitalization was not feasible at the previous facility which tended to his treatment. We collaborated with the pediatrician who was responsible for the examination and treatment of the patient. We strived to provide prompt yet comprehensive care to the patient while reducing procedures as much as possible. Our tailored treatment plan proved to be effective as a result of the smooth handling of the perioperative treatment and management for the patient.

4. Conclusion

We presented a case of PG in the anterior maxillary region of a child, contributing significantly to the relevant literature. To rule out malignancy, it is important for physicians to collaborate closely with pediatricians to promptly examine and treat the rapidly growing tumors in children, which manifest possible signs of malignancy, as in the present case.

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Conflict of interest

The authors declare no conflicts of interest.

Author contributions

Conceptualization: Takeshi Karube, Seiji Asoda
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Supervision: Taneaki Nakagawa, Seiji Asoda
Writing – original draft: Takeshi Karube, Seiji Asoda
Writing – review & editing: Takeshi Karube, Kaori Yago, Hajime Okita, Seiji Asoda

Ethics approval and consent to participate

Not applicable.

Consent for publication

Informed consent has been obtained from the patient for publication of this case report.

Availability of data

The data can be requested from the corresponding author following reasonable request.

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