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CASE REPORT

Craniofacial/Pediatric

Recurrent Gingival Lesions in a Pediatric Patient

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Summary: We present the case of a 13-year-old girl who developed numerous gingival masses that recurred after two prior resections. Following the initial resection as a child, she reported that there was a period of resolution for several years before recurrence as a teenager. After the second resection, the masses recurred after 4 months. The lesions obscured the majority of her dentition and interfered with speech, eating, and oral hygiene. The patient underwent staged resection of the masses, and the wounds were allowed to heal by secondary intention. The histopathologic findings of the specimens were consistent with a diagnosis of peripheral ossifying fibroma, which is unusual as these are generally solitary lesions. We believe that this case brings attention to an underrecognized and atypical presentation of peripheral ossifying fibroma, and it should be considered in the differential diagnosis of multicentric gingival masses. *(Plast Reconstr Surg Glob Open 2022;10:e4382; doi: 10.1097/GOX.0000000000004382; Published online 15 June 2022.)*

For patients with intraoral soft tissue masses or swellings, there are several different possible diagnoses, one of which is peripheral ossifying fibroma (POF).[1] POF is a benign growth of the gingiva and generally thought to form in response to local irritation although idiopathic presentations have also been described.[2–4] There have been numerous synonymous terms for POF used in the literature, such as peripheral cementifying fibroma, peripheral fibroma with osteogenesis or cementogenesis, and calcifying or ossifying fibrous epulis.[4] They are generally considered to be solitary lesions with the majority found in the maxilla, specifically the incisor and canine areas.[2,3,5]

In this report, we describe an unusual presentation of multicentric POF and review the histologic findings in the resection specimens that are characteristic of this lesion.

CASE PRESENTATION

A 13-year-old girl presented with recurrent maxillary and mandibular gingival lesions. The patient reported that she had soft tissue masses on the maxilla, mandible, and palate early in life that were resected. She had recurrence of the lesions after a period of resolution and underwent reresection of the lesions at the age of 12, with reported complete clearance. After four months, the masses appeared again. The patient now had diffuse hyperplastic gingival tissue across the maxillary and mandibular alveolar segments (Fig. 1). Her teeth were significantly obscured by the soft tissue. Radiographs demonstrated no significant lytic or expansile lesions involving the alveolar or basal bone involvement (Fig. 2). The patient endorsed difficulty with speech, eating, and maintaining oral hygiene. She denied any other medical issues. For symptom relief and diagnostic confirmation, excision of the masses on the anterior maxillary and mandibular alveolar areas was performed. Intraoperatively, the teeth in the involved alveolar segments were grossly mobile. The specimens were sent to pathology.

Histopathologic examination of resections from her original and recurrent lesions all revealed the most frequently reported histopathological features of POF: fibrosis, extraosseous bone formation, and pyogenic granuloma-like overlying granulation tissue (Fig. 3).[2,6] The mineralized components included islands of mature bone and occasional psammomatous calcifications in a background of dense collagenous matrix and cytologically bland spindle cells.

At the 3-week postoperative visit, the patient had recovered well with no evidence of recurrence (Fig. 4). Future treatment will be directed toward further debulking, followed by dentoalveolar reconstruction once a stable soft tissue and bony envelope is created.

DISCUSSION

POF is a benign gingival lesion of uncertain etiology but is thought to be the product of a reactive inflammatory response to local trauma, such as plaque accumulation

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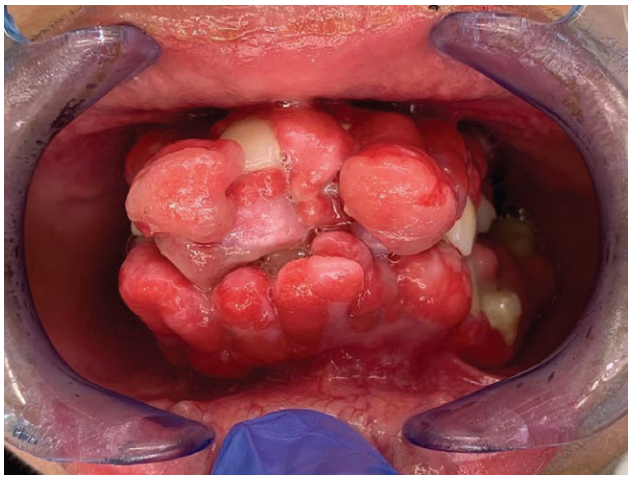

Fig. 1. Intraoral examination reveals diffuse hyperplastic gingival tissue across the maxillary and mandibular alveolar segments.

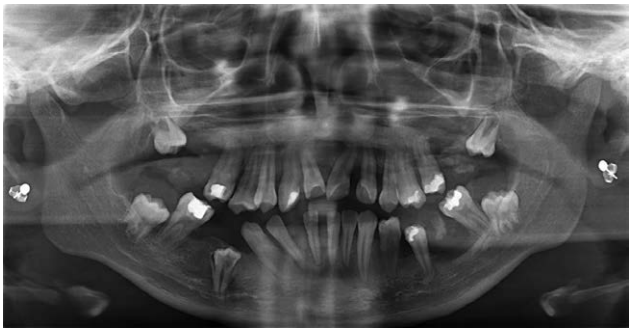

Fig. 2. Panoramic radiograph demonstrating no gross osseous pathology.

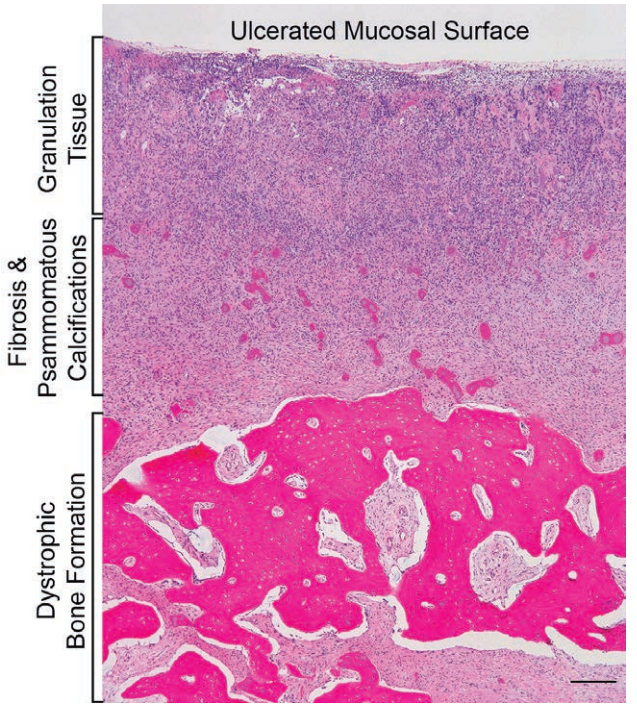

Fig. 3. A low magnification image of the ulcerated gingival surface and underlying stroma reveals typical features of peripheral ossifying fibroma, including a superficial zone of pyogenic granuloma-like inflamed granulation tissue and underlying fibrous stroma with a mixture of psammomatous mineralized bodies and broad zones of mature bone formation. Scale bar = 200 μm.

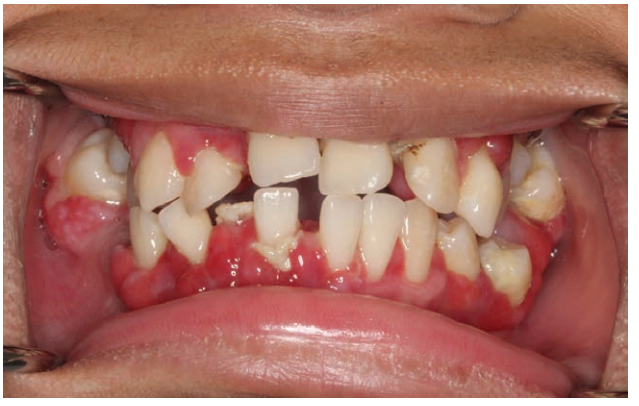

Fig. 4. Intraoral examination at 3-week postoperative visit.

or permanent tooth eruption, associated with the periodontal ligament. POFs are responsible for 3% of all oral tumors and almost 10% of gingival tumors. In one study, 20% of POFs were found in pediatric patients.[3] The lesion occurs most commonly in young women in their first or second decade of life, is typically found on the anterior maxillary alveolus, and is usually solitary.

In considering this case, the differential diagnosis included other reactive gingival lesions, including idiopathic gingival fibromatosis, pyogenic granuloma, peripheral giant cell granuloma, and juvenile ossifying fibroma. Histologic examination was necessary to arrive at a definitive diagnosis given the unusual multicentric presentation. The pathology of the biopsy specimen in this patient was unequivocally diagnostic of POF and not consistent with other diagnoses. Gingival fibromatosis is characterized primarily by accumulation of collagenous extracellular matrix and bland spindle cells.[7] Although small psammomatous calcifications may rarely be seen in gingival fibroma, the large zones of bone formation present in the current case are not. Pyogenic granulomas are characterized by vascular proliferation and granulation tissue and do not contain bone mineralization.[1] Peripheral giant cell granulomas are characterized by giant cells dispersed through stromal tissue.[1] Although the psammomatous calcifications found focally in the patient's lesion bear some resemblance to those found in juvenile (active/aggressive) ossifying fibroma, the latter is an intraosseous lesion, which arises in the native bones, that does not produce the diffuse gingival pathology without bone involvement observed in this patient.

POF generally manifests as a single lesion, less than 2 cm in width, and can be pedunculated or sessile. Other less common presentations have been described, such as giant POFs and those involving the floor of mouth and mandible. Four cases of multicentric POF have been previously published in the literature, and of those, two were

pediatric cases.[8–11] In two of the case reports, the management of the disease was challenging due to multiple recurrences, which similarly occurred in this patient.

Management of POF consists of surgical excision of the mass, including the associated periodontal ligament and periosteum. In the reported cases of multicentric POF, all patients underwent surgical excision. In one patient, corticosteroid infiltration and trichloroacetic acid were also performed in conjunction with several of the excisions but the authors report that the lesions continued to recur.[11] Recurrence rates are reported as high as 20%, so long-term follow-up in these patients is essential.[6] It was discussed that if the lesions should recur in this patient, the surgical plan will be staged total excision of the gingiva and periosteum with reconstruction using allograft and mucosal flaps. The majority of her teeth were periodontally compromised—she will require dentoalveolar reconstruction in the future. This case highlights the importance of early diagnosis and surgical management as untreated lesions can result in permanent damage to surrounding teeth and bone.[3]

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REFERENCES

1. Maymone MBC, Greer RO, Burdine LK, et al. Benign oral mucosal lesions: clinical and pathological findings. *J Am Acad Dermatol.* 2019;81:43–56.
2. Mergoni G, Meleti M, Magnolo S, et al. Peripheral ossifying fibroma: a clinicopathologic study of 27 cases and review of the literature with emphasis on histomorphologic features. *J Indian Soc Periodontol.* 2015;19:83–87.
3. Cuisia ZE, Brannon RB. Peripheral ossifying fibroma–a clinical evaluation of 134 pediatric cases. *Pediatr Dent.* 2001;23:245–248.
4. Yadav R, Gulati A. Peripheral ossifying fibroma: a case report. *J Oral Sci.* 2009;51:151–154.
5. Rallan M, Pathivada L, Rallan NS, Grover N. Peripheral ossifying fibroma. *BMJ Case Rep.* 2013;2013:bcr2013009010.
6. Lázare H, Peteiro A, Pérez Sayáns M, et al. Clinicopathological features of peripheral ossifying fibroma in a series of 41 patients. *Br J Oral Maxillofac Surg.* 2019;57:1081–1085.
7. Gawron K, Łazarz-Bartyzel K, Potempa J, et al. Gingival fibromatosis: clinical, molecular and therapeutic issues. *Orphanet J Rare Dis.* 2016;11:9.
8. Khan FY, Jan SM, Mushtaq M. Multicentric peripheral ossifying fibroma: a case report and review of the literature. *J Indian Soc Periodontol.* 2013;17:648–652.
9. Kumar SK, Ram S, Jorgensen MG, et al. Multicentric peripheral ossifying fibroma. *J Oral Sci.* 2006;48:239–243.
10. Choudary SA, Naik AR, Naik MS, et al. Multicentric variant of peripheral ossifying fibroma. *Indian J Dent Res.* 2014;25:220–224.
11. Lima MD, Teixeira RG, Bonecker M, et al. Recurrent multicentric peripheral ossifying fibroma-like lesion in a child: a case report. *BMC Res Notes.* 2014;7:673.