A case of peripheral odontogenic fibroma arising in the mandibular premolar region of a teenager

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March 26, 2022

Abstract

Peripheral odontogenic fibroma (POdF) is a rare, benign, and ectomesenchymal tumor. We report a case of a 15-year-old girl who developed POdF in the mandible. The lesion was resected, including the periosteum. Histopathological findings revealed a small mass and cord-like epithelium. No sign of post-operative recurrence appeared after 16 months.

1. Introduction

Peripheral odontogenic fibroma (POdF) is a rare ectomesenchymal, benign odontogenic tumor that most often occurs in adult women, frequently in the mandible and anterior maxilla regions.[1,2]Histopathologically, the tumor consists of fibrotic tissue with odontogenic epithelium and hard tissue showing varying degrees of calcification, but its expression is rare.[3,4]

Along with a review of the literature, here we report a case of POdF arising in the mandibular premolar region of a teenager.

2. Case story / examination

A 15-year-old girl with a chief complaint of swelling in the right mandibular premolar region was referred to our department. She had a history of surgery for a congenital ear fistula. The patient first recognized a painless mass in her mandible at 6 years of age. On this occasion, she was referred to a local dental clinic for the treatment of dental caries and then to our hospital for the treatment of the mandibular tumor. Intraoral examination revealed a well-defined, painless, elastic hard mass, measuring 17×17 mm in size, between the right mandibular canine and first premolar (Figure 1). Panoramic radiography showed a radiolucent area in the alveolar bone at the same location (Figure 2). In addition, sandy calcification was observed inside the lesion. Computed tomography (CT) revealed compressive bone resorption of the alveolar bone between the mandibular right canine and first premolar (Figure 3). Magnetic resonance imaging (MRI) revealed an endophytic appearance of a well-defined enhancing mass on the right side of the mandible on T2-weighted imaging (Figure 4).

A biopsy was performed under local anesthesia and a benign tumor with fibroblast proliferation was diagnosed. Tumor resection was performed under general anesthesia. The tumor was detached from the surrounding bone surface and resected, including the periosteum. A bone depression was observed around the tumor, and the bone surface was smooth. The tumor was 17×17 mm in size and had a smooth surface. Histopathological examination revealed spindle-shaped fibroblast-like cells that spread subcutaneously. Scattered cord-like and lump-like epithelial components were observed in the fibrous tissue near the base of the tumor (Figure

5A). No atypical cells or nuclear fission were observed. Cementum-or bone-like calcifications were observed in the deep area of the tumor. Epithelial components resembled Marasse's remaining epithelial or Hertwig epithelial sheath and were positive for cytokeratin (CK) CK19 staining (Figure 5B, C). There were no signs of post-operative recurrence after 16 months.

3. Discussion

Gardner described POdF in comparison with central odontogenic fibroma and histopathologically separated it into peripheral ossifying fibroma.[5] POdF is a rare odontogenic tumor with gradual growth consisting of a hard, elastic, smooth gingival mass.[1,3,4]

The onset age of POdF is 5 months to 84 years, and there is seemingly a predominant peak in middle age, specifically, during patients' forties.[1,2,6,7] The most common sites to develop POdF are known to be the anterior maxilla and mandible in adults, while some reports show that POdF also occurs frequently in the mandibular canine to premolar region.[7,8] A total of 25 reports of POdF occurring in patients younger than 19 have been published, of which, 12 reported POdF development in the mandible (Table 1).[6,7,9–18] Among those, two cases, including the present case, reported POdF occurring in the mandibular premolar region of teenagers.[10]

Histopathologically, POdF is characterized by odontogenic epithelium scattered to various degrees in the fibrous substrate.[3,5,7] Moreover, bone-like, dentin-like, or cementum-like hard tissue formation is observed, and it is classified into epithelium-poor or epithelium-rich type according to the degree of content of the dentin epithelium.[19] Regarding the hard tissue inside the lesion, bone-like and cementum-like hard tissue formation have a reported frequency of 28.3% and 15.2%, respectively.[6] In the present case, a region was described histopathologically as rich in cellular components as well as a mixture of small mass and cord-like epithelial components similar to Marasse's epithelial remnants and Hertwig epithelial sheath. Moreover, bone-like calcification was observed in the deep areas of the tumor.

In this sense, there are so far only four reports of POdF with hard tissue inside the lesion among teenagers.[17] Although few reports have been published on hard tissue formation in POdF, it has been suggested that the odontogenic epithelium inside the lesion may induce hard-tissue formation, which requires a lengthy time to develop in teenagers.[17] Thus, it has been speculated that the frequency of calcification in POdF might be low.

Some reports of CK-positive cells in the odontogenic epithelium of POdF exist.[20] In the present case, CK19-positive epithelial components resembled Marasse's epithelial remnants and Hertwig epithelial sheath.

POdF was hypothesized to originate from the periodontal ligament, tooth sac, and dental papilla.[17] Central odontogenic fibroma originates from the periodontal ligament or tooth sac according to the 2005 World Health Organization classification, while other authors have reported the possibility of periodontal ligament origin as well.[19] We considered periodontal ligament or tooth sac as the origin of the tumor described here for the following reasons: the tumor was observed during the tooth-replacement period, tumor growth was observed from the neck of the adjacent tooth, fibroblast growth around the tumor did not resemble that of dental papilla, and Marasse's epithelial remnants and Hertwig epithelial sheath-like cells were observed.

The rate of POdF recurrence after surgery is reportedly low.[17,21] However, some studies have reported an early recurrence rate of 50% (29/58 cases).[2,22,23]

4. Conclusions

We resected the tumor along with the periosteum and preserved the adjacent tooth; there were no signs of recurrence at 14 months after the operation. For POdF that develop in young people, long-term follow-up is necessary in consideration of the risk of recurrence.

Authorship list:

Author Contributions:

Kie Yamashiro , Katsuhisa Sekido, Yasushi Hariya contributed to the manuscript preparation. Michiko Okita, Masashi Harada, Yasushi Hariya, and Masaharu Tatetsu contributed to the patient management. All authors read and approved the final manuscript.

Acknowledgments: The authors would like to thank Dr. Eiji Nakayama, Professor at Health Sciences University of Hokkaido, for assistance with diagnostic imaging, and Dr. Tomoyuki Oouchi, Chief of the Department of Pathology at Keiyukai Sapporo Hospital, for assistance with the pathological investigation.

Conflict of interest:

The authors have no conflicts of interest to declare.

Abbreviations: CK, cytokeratin; CT, computed tomography; MRI, magnetic resonance imaging; POdF, peripheral odontogenic fibroma

Consent

Confirmation that the author has obtained written informed consent from patient(s).

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Table 1. Reports of peripheral odontogenic fibroma occurring in patients under 19 years of age.

Figure legends

Figure 1. Intraoral view. (A, B) Intraoral view revealed a well-defined, painless, elastic hard mass, measuring 17×17 mm in size between the right mandibular canine and first premolar.

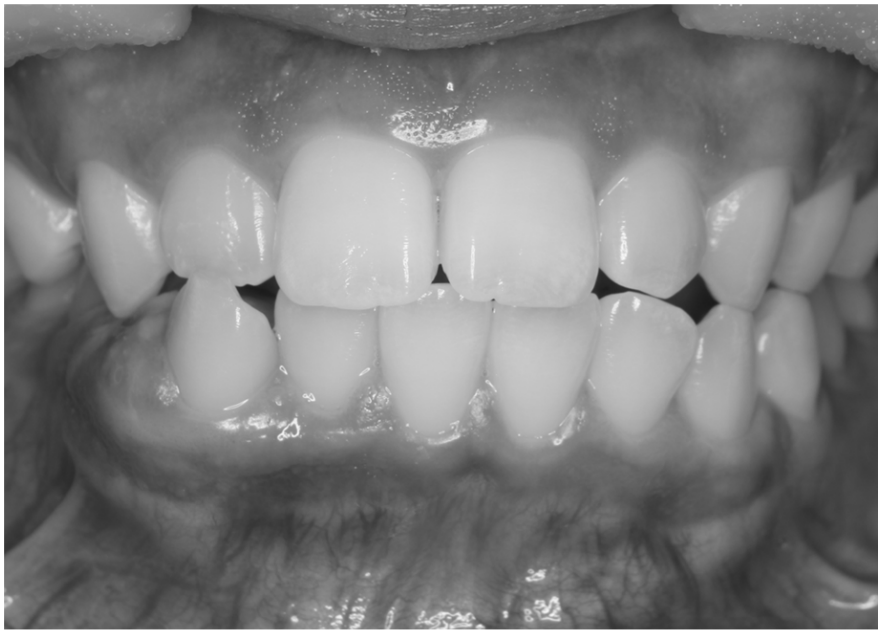
Figure 2. Panoramic X-ray. Panoramic radiography showed a radiolucent area in the alveolar bone between the right mandibular canine and first premolar.

Figure 3. Computed tomography findings . (**A, B**) Compressive bone resorption of the alveolar bone between the right canine and first premolar of the mandible.

Figure 4. Magnetic resonance imaging findings. Endophytic appearance of a well-defined enhancing mass on the right side of the mandible on T2-weighted images.

Figure 5. Histopathological findings. (**A**) Scattered cord-like and lump-like epithelial components were observed in the fibrous tissue near the base of the tumor (H-E staining, ×40).

(**B**) Cementum- or bone-like calcifications were observed. Epithelial components resembled Marasse's remaining epithelial or Hertwig epithelial sheath and were positive CK19 (H-E staining, ×400; CK19 staining, ×40).



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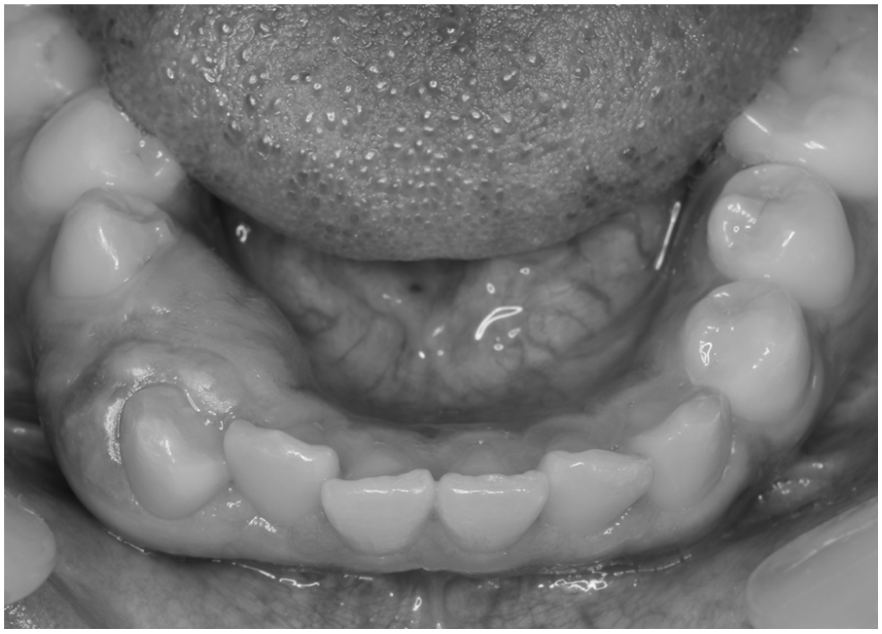

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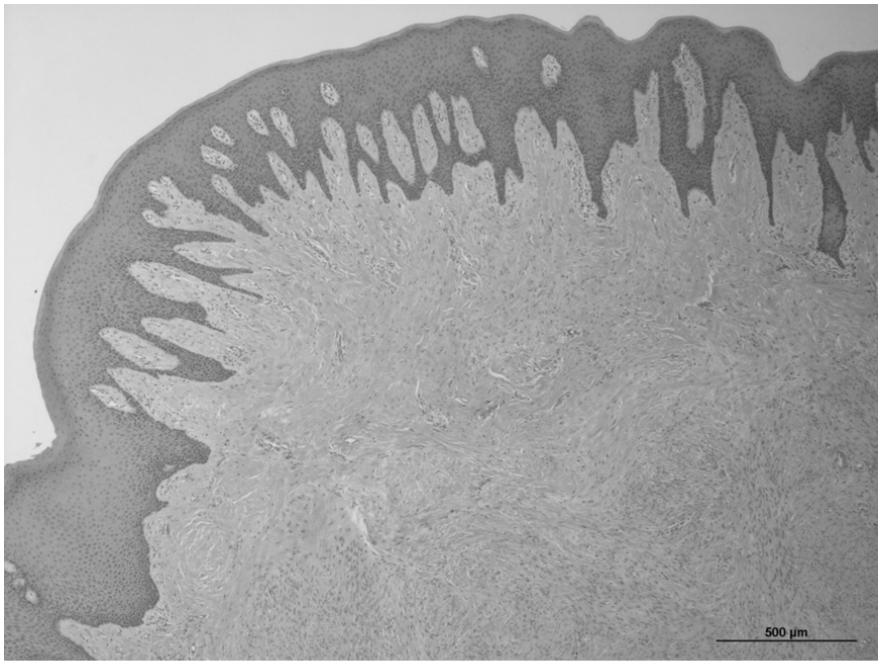

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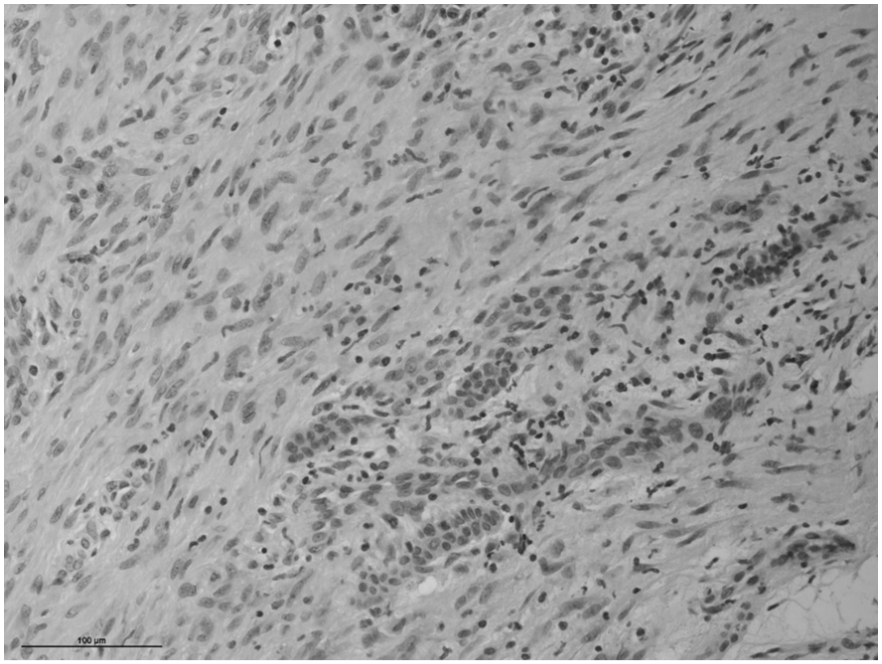

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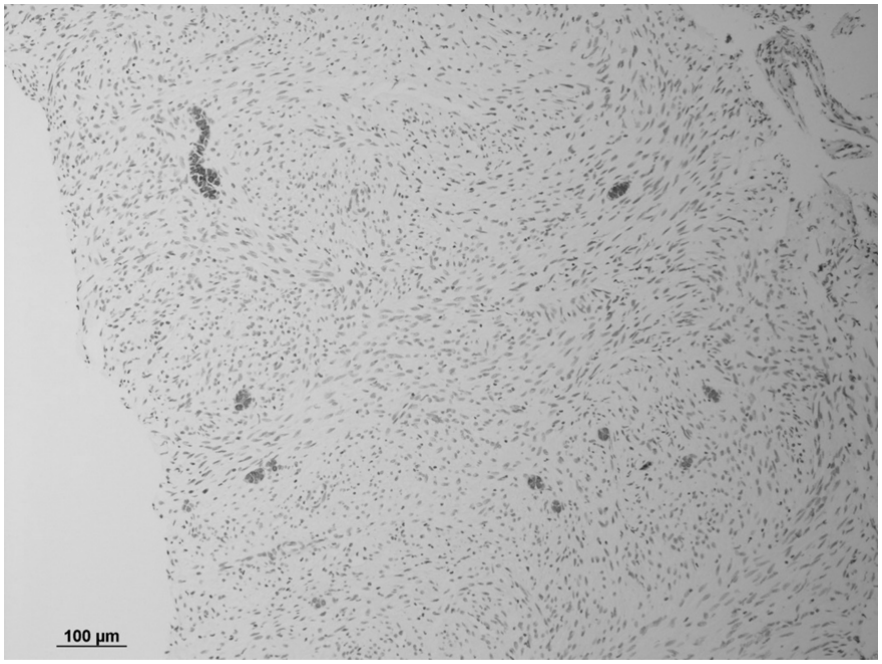

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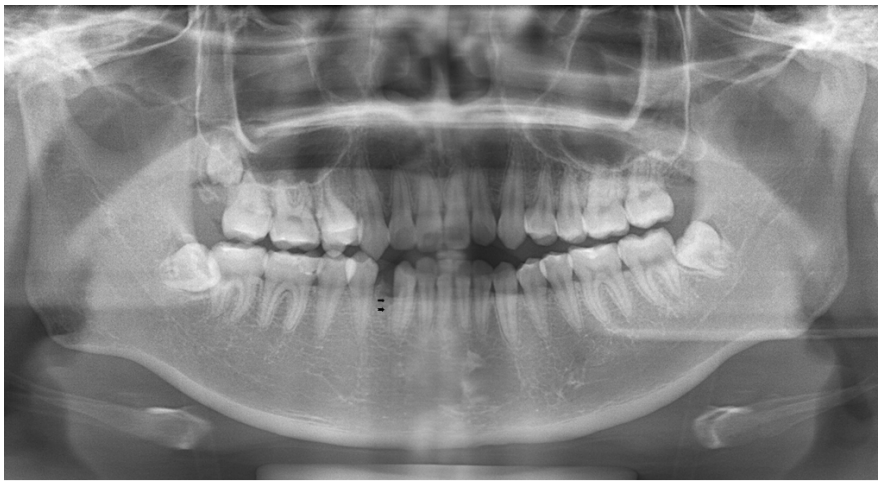
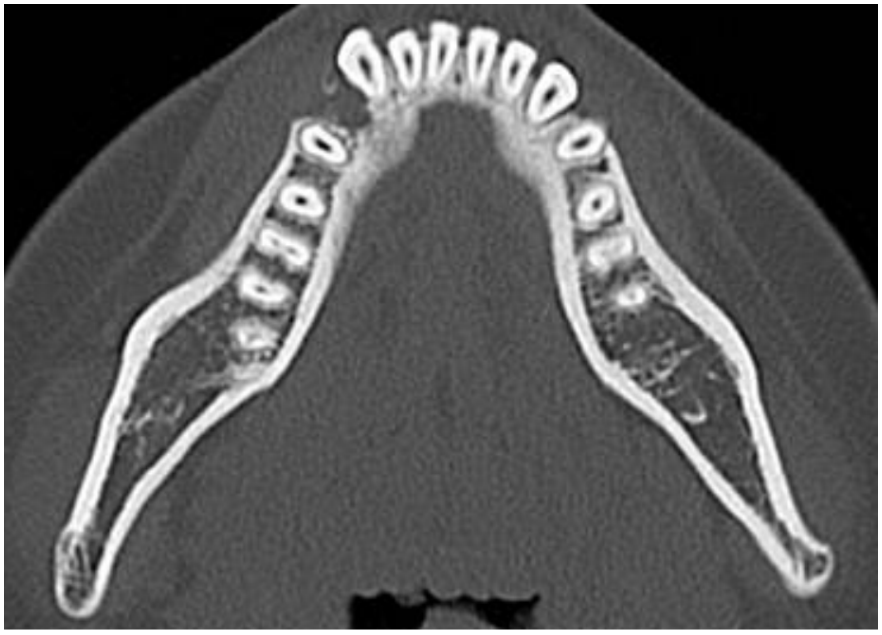


Figure 2



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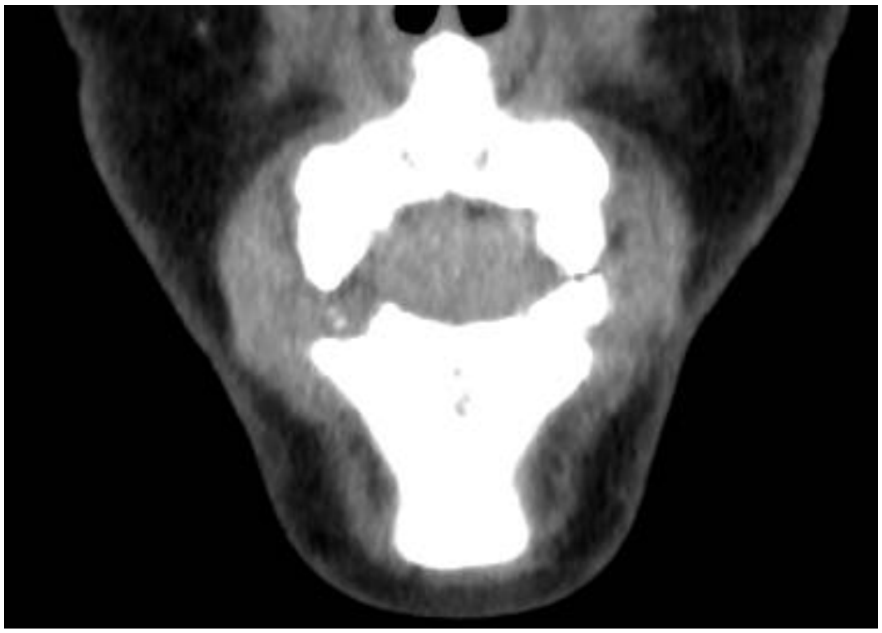

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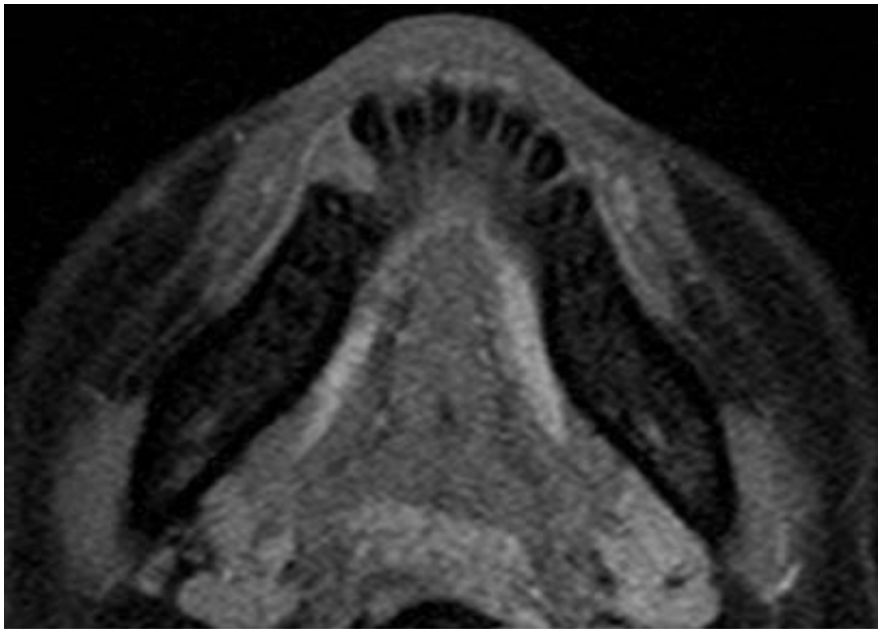

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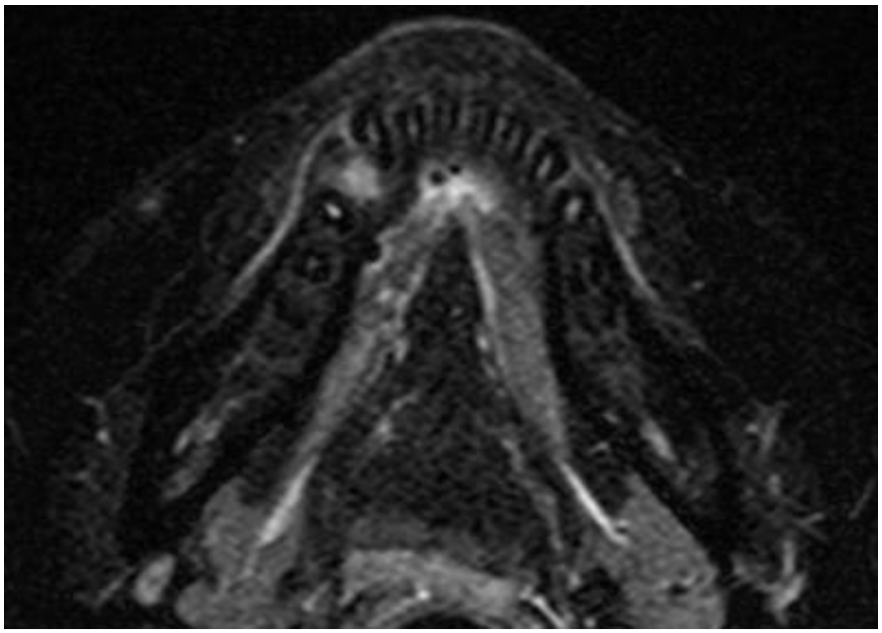

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