

# A Case of Multiple Idiopathic Root Resorption

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How to cite this paper: Konishi, R., Mori, A. and Yoshida, T. (2023) A Case of Multiple Idiopathic Root Resorption. *Open Journal of Stomatology*, **13**, 156-165. https://doi.org/10.4236/ojst.2023.134013

**Received:** March 6, 2023 **Accepted:** April 25, 2023 **Published:** April 28, 2023

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#### Abstract

Tooth root resorption is multifactorial, and its etiology and pathogenesis are poorly understood. Tooth root resorption is often incidentally revealed on radiographic examination. Here, we report a case of root resorption in multiple teeth of unknown etiology. Radiographic examination revealed root resorption in the cervical region of the left lateral incisor, canine, first and second premolars, and first and second molars of the mandible. Panoramic radiographs revealed no mandibular lesions that could cause tooth resorption. The patient did not wish to undergo any treatment and is currently under observation. Tooth root resorption with unknown etiology is rare, and further case collection is needed to determine the cause and treatment.

### **Keywords**

Multiple Teeth, Cement Enamel Junction, External Cervical Resorption

# **1. Introduction**

Resorption is a condition associated with physiological and pathological factors resulting in the loss of dentin, cement, or bone [1]. Various etiological factors for tooth root resorption have been reported, including trauma, pulpal infection, tooth bleaching, and orthodontic treatment [2] [3] [4] [5]. Despite various etiological factors, the etiology of some types of resorptions remains unclear [6] [7] [8]. Multiple idiopathic root resorption (MIRR) was first reported by Mueller [9] in 1930 and is an extremely rare disease of unknown etiology. MIRR is a rare condition in which diffuse external root resorption is observed in multiple teeth. We report a case of MIRR that was discovered incidentally during a radiological examination, along with bibliographical considerations.

# 2. Case Report

A 52-year-old Japanese woman was referred to our hospital for assessment of

cervical root resorption of multiple teeth. Her chief complaint was occlusal pain in the left premolar lesion. She had no significant medical or dental history. Oral examination showed no dental caries in the left mandibular teeth and mild gingivitis at the same site. The root resorption of the surface of the cervical region was not palpable through the gingival sulcus. A panoramic radiograph at the first visit showed root resorption of the cervical region of the teeth (left lateral incisor, canine, first and second premolars, and first and second molars of the mandible), and no abnormal findings were observed in the bone at the same site (Figure 1). We suspected a bone metabolic disease other than dental diseases and consulted the Department of Diabetes, Endocrinology, and Nutrition of our hospital. Blood test results (Table 1) were normal except for  $1\alpha$ , 25-dihydroxy vitamin D3, and tartrate-resistant acid phosphatase. 1a, 25-dihydroxy vitamin D3 was 15 ng/ml (normal, 20 ng/mL), and tartrate-resistant acid phosphatase was 597 mU/dl (normal range, 120 to 420 mU/dl). In addition, Young Adult Mean values of the lumbar spine, femur, and forearm were 54%, 59%, and 85%, respectively. The patient was diagnosed with osteoporosis; however, no systemic disease leading to root resorption was found. Based on the findings, this case was diagnosed as multiple idiopathic root resorption. Since the patient did not wish to undergo treatment, it was decided to follow up until the teeth were fractured. One year later, the oral examination showed unchanged from the first examination (Figure 2). A panoramic radiograph showed that the cervical root resorption



Figure 1. Panoramic radiograph at the initial visit.

Table 1. Blood test results
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Blood test results						
ALP (38 - 113 U/L)	110 U/L					
Ca (87 - 10.1 mg/dl)	9.2 mg/dl					
IP (2.6 - 4.5 mg/dl)	3.3 mg/dl					
urine creatinine (<30 mg/dl)	24 mg/dl					
urine calcium (2.0 - 40 mg/dl)	7.1 mg/dl					
1 <i>a</i> , 25-dihydroxy vitamin D3 (20 ng/ml)	15 ng/ml					
tartrate-resistant acid phosphatase (120 - 420 mU/dl)	597 mU/dl					



Figure 2. Intraoral photographs one year after initial visit. (A) Frontal view; (B) lower view.



Figure 3. Panoramic radiograph one year after initial visit.

of the left lateral incisor, canine, first and second premolars, and first and second molars of the mandible had progressed, and the extent of cervical resorption had expanded to the left central incisor of the mandible (**Figure 3**). Since the patient did not wish to undergo treatment, it was decided to follow up until the teeth were fractured.

#### **3. Discussion**

Root resorption is classified as internal or external [10]. External resorption is classified into five categories: inflammation, replacement, cervical, surface, and

transient apical breakdown [10]. External cervical resorption (ECR) progresses in the cervical region of teeth. Although the etiology of ECR remains unclear, several factors have been suggested [10] including trauma [8] [11] [12], orthodontic treatment [8] [12], periodontal treatment [8] [11], intracoronal bleaching [8] [13], playing wind instruments [14], and idiopathic etiology. In this case, cervical root resorption was observed and classified as ECR. Additionally, idiopathic root resorption was diagnosed because there was no history of trauma, orthodontic treatment, or periodontal treatment. MIRR has been reported in 45 cases between 1970 and 2022 [15]-[49] (**Table 2**). The male-to-female ratio was 1:2.

Number	Author	Sex	Age	General condition	Reason for visit	Starting tooth position	New disease tooth position
1	Kerr <i>et al.</i> (1970)	Female	68	Osteoarthritis. Hypophosphatemia. Hypocalcemia. Low alkaline phosphatase level, returning to normal after 9 years	Routine oral examination	16 - 14, 12, 21 - 26, 42, 43	13, 11, 36 - 41, 44, 46, 47
2	Kerr <i>et al.</i> (1970)	Female	30	Hormone therapy for menstrual disorders	Root resorption	16, 14, 13 - 11, 23 - 27, 37, 33 - 43, 45	/
3	George and Miller (1986)	Female	40	No obvious abnormality	Routine oral examination	11, 12, 13, 32, 33, 34, 35, 36	No
4	George and Miller (1986)	Female	20	Regular review within 2 years without exception follow orthodontic treatment; no new lesions for 4 years after the initial visit and treatment; new root resorption detected after pregnancy	Looseness and pain of teeth	23, 25, 26, 27	No
5	Lydiatt <i>et al.</i> (1989)	Female	39	No obvious abnormality, underwent two gynecological operations	Routine oral examination	26, 27	The entire maxillary dentition
6	Moody <i>et al.</i> (1990)	Male	27	No obvious abnormality	Routine oral examination	17, 16, 25, 26, 27, 31, 33 - 37, 41, 43 - 47	No
7	Moody <i>et al.</i> (1990)	Male	20	No obvious abnormality	Abnormal dental imaging	15, 16, 44, 45, 46	13, 14, 17, 18
8	Moody <i>et al.</i> (1990)	Female	44	Gastric regurgitation	Abnormal dental imaging	25 - 27, 34, 35, 44 - 47	1
9	Liang <i>et al.</i> (2003)	Female	19	No obvious abnormality, no significant progression of root resorption within 2 years after initial visit, new root resorption occurred after 2 years, during which the patient became pregnant	Looseness and pain of teeth	41, 42, 43	15 - 17, 24 - 27, 31 - 33, 44 - 46
10	Liang <i>et al.</i> (2003)	Male	68	No obvious abnormality	Bite discomfort	43 - 45	42, 46, 47
11	Liang <i>et al.</i> (2003)	Male	50	Underwent cholecystectomy 5 years previously	Supplement shedding	34, 35	16, 17, 21 - 25, 42 - 44, 46, 47

Table 2. Clinical features of MIRR cases.

12	Iwamatsu-Kobayashi <i>et al.</i> (2005)	Female	49	Hyper-alkaline-phosphatemia and osteoporosis	Difficulty chewing	11 - 13, 15, 17, 21 - 25, 31 - 35, 41 - 45, 47	No
13	Coyle <i>et al.</i> (2006)	Female	33	The patient had a repair of an incomplete cleft palate with an autogenous bone graft as an infant, and again at 17 years	Routine oral examination	21 22, 23	17 - 14, 24, 25
14	Neely and Gordon (2007)	Male	63	Vitiligo on the skin, after colon cancer surgery, mild heart attack	Root resorption	13, 48	17, 16, 13, 12, 23, 24, 25, 26, 27, 37, 36, 34, 33, 45, 44,
15	Mattar <i>et al.</i> (2010)	Female	23	No obvious abnormality, deep overbite	Crown fracture	46, 45, 32, 31	No
16	von Arx <i>et al.</i> (2009)	Female	50	No obvious abnormality, history of cat exposure	Abnormal dental imaging	12, 11, 21, 22, 23, 24	17, 16, 15, 14, 13, 25, 26, 27
17	von Arx <i>et al.</i> (2009)	Female	58	No obvious abnormality, history of cat exposure	Root resorption	35 - 44	No
18	von Arx <i>et al.</i> (2009)	Male	68	No obvious abnormality, history of cat feces exposure	Root resorption	14, 13, 12, 11, 21, 22, 42, 43, 44, 45, 46	/
19	von Arx <i>et al.</i> (2009)	Male	66	Blind, contact with guide dogs, maybe indirectly exposed to cats	Root resorption	13 - 26, 36 - 44, 47	/
20	Yu <i>et al.</i> (2011)	Male	33	No obvious abnormality	Implanted teeth	15 - 25, 37 - 47	Full mouth teeth
21	Arora <i>et al.</i> (2012)	Female	36	No obvious abnormality	Tooth pain	23, 24, 25, 26	/
22	Roy <i>et al.</i> (2012)	Male	32	No obvious abnormality	Repair missing teeth	13, 23, 24, 25	/
23	Kjær <i>et al.</i> (2012)	Female	28	Pertussis	Root resorption	25, 26, 31, 32	21 - 24, 27, 33
24	Haeberle (2013)	Female	19	No obvious abnormality	Abnormal dental imaging	/	Full mouth teeth
25	Ge and Wang (2013)	Female	27	Discovery of lesions during pregnancy	Tooth pain	22, 23, 26	/
26	Chowdhury and Bashar (2014)	Male	37	Total urine phosphate level is low	Difficulty chewing, tooth pain	31, 41, 42, 43, 48	11 - 18, 21, 23, 27
27	Lu and Li (2014)	Female	30	Healthy	Loose lower front teeth with swollen gums	32 - 42, 35, 37, 45 - 47	/
28	Jiang <i>et al.</i> (2014)	Female	26	No obvious abnormality	Bite discomfort	31 - 46	No
29	Kesary <i>et al.</i> (2014)	Male	17	No obvious abnormality	Swollen gums, loose teeth	17, 16, 14 - 23, 31 - 33, 41 - 47	/
30	Wu <i>et al.</i> (2016)	Female	27	No obvious abnormality, history of cat exposure	Chewing pain, loose teeth	32 - 35	12 - 18, 21 - 26, 31, 36, 37, 48, 41 - 46
31	Neely <i>et al.</i> (2016)	Male	43	Healthy	Root resorption	16	26
32	Neely <i>et al.</i> (2016)	Female	60	Hormone replacement therapy for hypothyroidism as a child	The neck of tooth absorption	23	No

Continued

33	Kumar <i>et al.</i> (2018)	Male	18	History of hepatitis B virus infection	Root absorption	36, 37, 42 - 47	12 - 15, 24 - 26, 33 - 38, 48
34	Dobroś <i>et al.</i> (2018)	Male	41	Celiac disease	Loose teeth	16, 17, 13 - 21	No
35	Shashidara <i>et al.</i> (2018)	Male	32	1	Missing teeth requiring restoration	15, 14, 23 - 25	No
36	Li <i>et al.</i> (2019)	Female	31	No obvious abnormality	Routine oral examination	21, 36	37, 38
37	Llavayol <i>et al.</i> (2019)	Female	26	History of chemotherapy for ovarian cancer	Root absorption	23 - 26, 33 - 35, 43 - 47	36
38	Sharma <i>et al.</i> (2019)	Female	23	No obvious abnormality	Pain and loosening of tooth	23 - 25, 33 - 45	16 - 23, 25 - 28, 34 - 36, 46, 47
39	Deeb <i>et al.</i> (2019)	Female	69	Various systemic diseases, discontinue denosumab for osteoporosis	Pain and sensitivity of the tooth	27, 37	12 - 22, 36, 32 - 44
40	Chen <i>et al.</i> (2020)	Female	29	Discovery of lesions during pregnancy	Loose teeth	17 - 27, 41 - 37, 44 - 47	42, 43, 18, 28, 38, 48
41	Macaraeg <i>et al.</i> (2020)	Female	10	Congenitally missing ossicles, familial expansile osteolysis	Imaging shows root resorption	21, 22, 23, 31, 41	/
42	Samara <i>et al.</i> (2021)	Female	16	History of sepsis	Generalized dull and continuous pain; mild mobility in lower anterior teeth.	14 - 16, 32-36, 41 - 47	37
43	Yilmaz <i>et al.</i> (2022)	Female	36	No systemic health issues or medication use	Unable to chew due to loss of maxillary teeth	11 - 24, 35 - 45	No
44	Mikušková <i>et al.</i> (2022)	Female	74	Arterial hypertension, glaucoma, and postmenopausal osteoporosis with multiple osteoporotic thoracic vertebral fractures	Occasional pain, thermal sensitivity, and slightly increased mobility of the left mandibular second premolar	16, 15, 13 - 26, 37 - 45	/
45	Beaumont <i>et al.</i> (2022)	Female	55	De novo bone metastatic breast cancer	Pain localized to upper right molars	18 - 24, 26 - 28, 38-42, 44 - 48	/

Wang *et al.* [50] reported that the occurrence and progression of MIRR may be related to hormonal changes in females because patients had a history of hormonal changes, including pregnancy, hormone therapy, and gynecological treatment. Considering the patient's age, it is thought that there was a female hormone decrease, and there is a possibility that root resorption and a decrease in hormones are related. The mandibular premolars are most frequently affected during the early stages of MIRR [15]. In this case, the mandibular premolars were within the range of the MIRR. Patients typically experience tooth pain and mobility. The other patients were incidentally diagnosed with MIRR during a radiographic examination. In the present case, occlusal pain was observed, but no other subjective symptoms were reported. The patient was incidentally diag-

nosed with MIRR during a radiographic examination. Resorption often originates from the mesial or distal Cement Enamel Junction, and its image resembles apple cores [17]. Similar characteristics were observed in the present case. Treatment options generally include tooth extraction and replacement with a partial denture, surgical exposure, lesion curettage, and restoration [27] [31]. In this case, because the patient did not wish to undergo treatment, follow-up performs until the teeth will be fractured.

#### 4. Conclusion

In this case, we have performed all possible tests, but have not been able to determine the cause of MIRR. Further case accumulation is needed for MIRR.

#### **Conflicts of Interest**

The authors declare no conflicts of interest associated with this manuscript.

#### **Ethics Approval Statement**

We conducted this study in accordance with the current revision of the 1975 Declaration of Helsinki.

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