

Localized idiopathic root resorption in the primary dentition: Review of the literature and a case report

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ABSTRACT

Idiopathic root resorption (IRR) is an infrequent condition that is usually found as an accidental finding on radiography. A significant number of cases of IRR in permanent dentition have been presented but are rarely reported in primary dentition. The aim of this case report is to present a case of localized IRR in a 7-year-old boy. The patient was referred because of increased mobility of the left mandibular primary second molar. On radiographic evaluation, severe root resorption of that tooth, and mild root resorption of the right mandibular primary second molar were evident; the patient was caries-free. The left affected tooth was lost, and after placing a band and loop space maintainer, the patient was followed for 18 months. A patient with an abnormal pattern of root resorption, especially in the primary dentition, should alert the clinician to rule out the known important local and systemic factors. The exact causes of and treatments for IRR continue to be discovered.

Key words: Idiopathic, localized, primary dentition, root resorption

INTRODUCTION

Root resorption is a multifactorial process that is classified as internal or external according to location. Internal root resorption is less common and occurs isolatedly as a result of chronic inflammation or pulpal infection, orthodontic movement, herpes zoster, or idiopathic factors.^[1,2] External root resorption has several causes, such as mechanical stimulation, inflammatory conditions, luxation injuries, and neoplastic conditions. External root resorption has been also reported to occur in some endocrine disturbances and systemic conditions such as hypoparathyroidism, hyperparathyroidism, hypocalcemia, Gaucher's disease, Paget's disease, hypophosphatemia, Stevens-Johnson syndrome, odontodysplasia, dentin dysplasia, and dentinogenesis imperfect.^[2-8] When none of these conditions are present, root resorption is termed "idiopathic root resorption" (IRR).^[9] IRR is infrequent and was first reported by Mueller and Rony in 1930

in a 37-year-old female.^[10] Several cases related to IRR in permanent dentition have been reported in the literature but cases reported in primary dentition are very few [Table 1]. Therefore, the aim of this review is to report a case of bilateral external IRR of second primary molars in a 7-year-old patient.

Literature review

IRR is an infrequent type of root resorption that can occur in both the cervical and apical regions of the tooth. According to most case reports, it seems that

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Table 1: Reported cases of idiopathic root resorption

Author	Sex	Race	Age	Number of teeth affected	Historical finding	Progression during follow-up period
Mueller and Rony ^[10]	Female	Not reported	36	7	General neurasthenia, vasomotor instability and hepatic functional impairment	+
Carr ^[11]	Female	Not reported	29	>5	Patient was pregnant when resorption was found	+
Kerr <i>et al.</i> ^[12]	Female	White	68	24	Initially high phosphorus and low calcium and alkaline phosphatase levels, returned to normal after 9 years, osteoarthritis	+
	Female	White	30	17	Hormonal therapy for menstrual problems, chronic pyelonephritis, osteoporosis and advanced otosclerosis 2 years later, high level of alkaline phosphatase	Not reported
Soni and La Velle ^[13]	Male	White	34	9	Unremarkable	Not reported
Hopkins and Adams ^[14]	Female	Not reported	20	18	Unremarkable	+
Belanger and Coke ^[15]	Male		14	All permanent teeth	Unremarkable	+
George and Miller ^[6]	Female	Not reported	20	7	Chief complaint of progressive pain and loosening of teeth in the upper left posterior quadrant, history of some bruxism and clenching, mild gingivitis, uncomplicated orthodontic treatment had been completed 2 years before the initial resorption, the patient was pregnant when new resorptions were found	+
	Female	Not reported	40	8	Unremarkable	-
	Female	Not reported	56	1	Except increased mobility, history was unremarkable	-
Pankhurst <i>et al.</i> ^[16]	Male	White	30	11	Marginal gingivitis and early chronic periodontitis, history of narcotic intravenous drug addiction and hepatitis A, low level of parathyroid hormone	-
Saravia and Meyer ^[17]	2 - female (twins)	Black	14	16	Mild generalized gingivitis, their grandmother was reported to be edentulous at an early age	-
Lydiatt <i>et al.</i> ^[18]	Female	White	39	>5	History of tooth loss at early age in her family as a result of gum disease, two gynecological procedures	-
Postlethwaite and Hamilton ^[19]	Male	Not reported	14	20	Unremarkable	Not reported
Moody <i>et al.</i> ^[20]	Male	White	27	17	Unremarkable	+
	Male	White	20	9	Unremarkable	Not reported
	Female	White	44	8	Prolonged problem of gastric regurgitation	Not reported
Moody and Muir ^[21]	Female	Not reported	19	6	Unremarkable	-
Beckett and Gilmour ^[22]	Male	White	57	6	Unremarkable	Not reported
Kim and Hefez ^[2]	Female	Not reported	7	12	Congenitally missing middle ear ossicles, early shedding of multiple primary teeth	Not reported
Di Domizio <i>et al.</i> ^[23]	Female	Not reported	26	All permanent teeth	Unremarkable/increased mobility of involved teeth/not very good oral hygiene was present	Not reported
Liang <i>et al.</i> ^[24]	Female	Latino	19	16	Unremarkable at first visit (3 teeth involved with resorption) and 2 years later, when the patient was pregnant, 13 new teeth were affected	+
	Male	Caucasian	68	6	Unremarkable	+
	Male	Caucasian	50	14	Presence of smoking habit and bruxism, history of allergy to penicillin, cholecystectomy 5 years prior to noticing root resorption	+
	Female	Caucasian	42	8	Orthodontic therapy as an adolescent and again as an adult, slightly high level of parathyroid hormone, osteopenia and presence of generalized gingival enlargement	+

Contd...

Table 1: Contd...

Author	Sex	Race	Age	Number of teeth affected	Historical finding	Progression during follow-up period
Cholia <i>et al.</i> ^[25]	Male	Caucasian	28	16	History of mid-facial fracture in an accident 2 years before initial diagnosis, presence of lateral and anterior open bite as a result of the facial fracture, relatively poor oral hygiene	+
	Male	Caucasian	38	All permanent teeth	Presence of bilateral cleft lip and palate repair and in the past and recently rhinoplasty and scarring from the lip repair, presence of heavily restored dentition, unremarkable familial history	Not reported
	Male	Arabic	37	12	Incisal relationship was class III, congenitally missing maxillary permanent canines	+
	Female	Caucasian	39	14	Increased mobility of lower left third molar tooth, unremarkable medical, dental and familial history	Not reported
Iwamatsu-Kobayashi <i>et al.</i> ^[13]	Female	Japanese	49	21	Osteoporosis, hyper-alkaline phosphatasemia	-
Schätzle <i>et al.</i> ^[26]	Female	White	17	28	Unremarkable	+
Moazami and Karami ^[27]	Male	Iranian	27	17	Unremarkable	Not reported
Gupta and Prakash ^[28]	Female	Not reported	38	10	History of hysterectomy 4 years ago and complaint of pain in the knee joints for the preceding 2 years, unremarkable familial and dental history	Not reported
Khojastepour <i>et al.</i> ^[29]	Male	Iranian	17	8	Unremarkable	Not reported
Sawai and Mehra ^[30]	Female	Not reported	40	28	A complaint of two adjacent painless mobile teeth, noncontributory medical, dental and family history	+
Current case	Male	Iranian	7	2	Unremarkable	+

this type of root resorption is more frequent in young females.^[2,12,24] However, in some literature there has been reported a predominance in men.^[26,27] IRR may affect a single tooth or more. Stafne and Slocumb evaluated 179 cases of IRR in 1944. According to the results of this survey, in most reported cases, only a single tooth was affected and in 19 of 179 cases, more than one tooth was involved.^[2,9] Massler and Perreault, in a study of 301 patients with IRR in at least four teeth, reported that this type of root resorption is mostly found in the maxillary premolars and mandibular incisors and molars exhibiting the least resorption.^[31]

Liang *et al.* reviewed the literature on multiple idiopathic root resorption (MIRR) and found that all cases were asymptomatic and that MIRR was usually detected incidentally on routine radiographs.^[24] However, some patients have reported cold sensitivity, loss of restorations, tooth mobility, and tenderness in the surrounding gingival tissues or involved teeth.^[6,32]

In 1989, Saravia and Meyer reported MIRR in monozygotic twins. Two 14-year, 7-month-old black females were referred to a dental clinic. Clinically and radiographically, no carious lesions were noted, but

root resorption was found in all posterior mandibular teeth and maxillary premolars on panoramic radiographs. Because no possible etiologic factor was found, a diagnosis of MIRR was made for both patients.^[17] Although local environmental factors cannot be completely ruled out, on the basis of this case report and the Newman study,^[33] it seems that genetic factors may also be involved in the pathogenesis of this condition. However, in most case reports and in the case of this study, no familial history of root resorption is reported.^[1,2,17,23,29]

In a large series of case reports of IRR, Stafne and Slocumb failed to find any correlation between this type of root resorption and any specific systemic conditions.^[9]

Kim and Heffez reported a case of MIRR in the primary teeth of a 7-year-old girl who was referred because of early shedding of multiple primary teeth. In her medical history, the parents did not report any systemic disease except for congenitally missing middle ear ossicles. Laboratory findings were normal. Radiographically, there was gross cervical root resorption in all primary teeth.^[2]

CASE REPORT

A 7-year-old boy was referred to Pediatric Department of Mashhad Dental School because of severe mobility in the second primary left mandibular molar. On evaluation of his medical history, the parents reported no systemic disorder. Laboratory findings, which included a complete blood cell count and electrolyte, calcium, phosphorus, and alkaline phosphatase values, were normal. There was no history of dental treatment or trauma. His oral hygiene was excellent, and there was no abnormal finding on extraoral and intraoral examination except for Class II malocclusion [Figure 1]. The patient was caries-free and had no parafunctional habits such as bruxism and no wear facets, or premature contacts were detected; the only chief complaint was increased mobility of the lower left second molar. There was no family history of early exfoliation of primary teeth, abnormal root resorption or spontaneous loss of permanent teeth. There was no history of hypersensitivity of the patient's teeth to thermal stimuli, spontaneous pain, or pain with mastication.

An orthopantomogram (OPG) X-ray was taken for thorough evaluation of the patient's dentition, and posteroanterior (PA) views were obtained for a more detailed examination of the affected tooth. On radiographic examination, there was no sign of caries or any other abnormal finding except root resorption in both of the second mandibular molars. OPG and PA views showed extensive root resorption of the left second mandibular molar and mild external root resorption of the right second mandibular molar [Figure 2].

On clinical examination, the color and texture of the gingival tissue around the involved teeth were normal. Except for the right mandibular primary second molar that had severe mobility, the mobility of the other teeth was within normal range. The pulp test revealed pulp vitality of the right mandibular primary second molar, but we could not test the left affected tooth because as we were taking the familial history, the patient wiggled and pulled the tooth out with his hand. On macroscopic examination of the exfoliated second molar, the roots were thoroughly resorbed, and the caries-free crown was undermined [Figure 3].

Histological evaluation of the exfoliated tooth was impossible due to complete resorption of roots, but histological evaluation of the soft tissue removed from the socket of the exfoliated tooth showed nonspecific

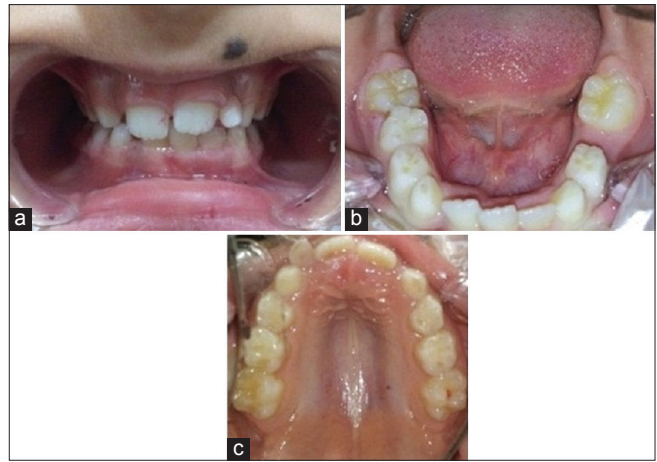


Figure 1: Intraoral examination showed no abnormal findings except for increased mobility of the left second lower molar (the tooth was removed prior to photography). (a) Frontal view of patient's occlusion (b) occlusal view of mandibular arch (c) occlusal view of maxillary arch

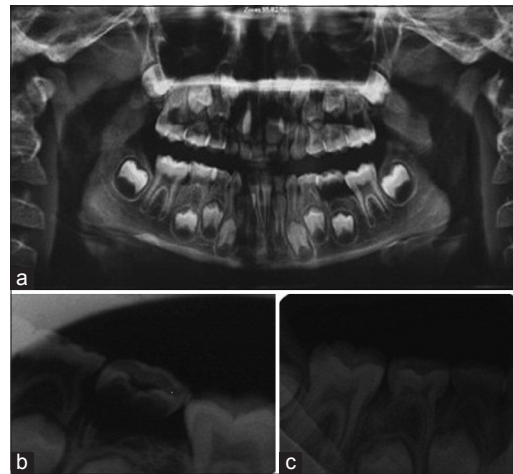


Figure 2: (a) Orthopantomogram X-ray showed severe inflammatory root resorption of left second mandibular molar and mild replacement of root resorption of right mandibular molar. (b and c) posteroanterior X-rays of affected areas

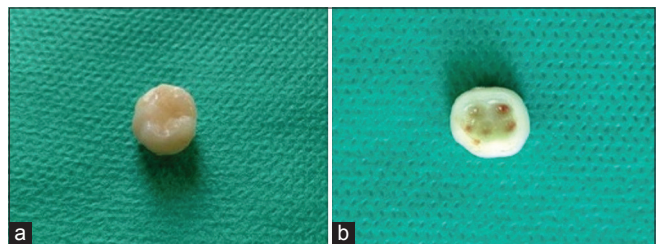


Figure 3: Gross examination of exfoliated left mandibular molar showing a complete root resorption and undermined crown with resorbed dentin and intact thin enamel. (a) occlusal view of exfoliated crown (b) internal view of exfoliated crown

chronic inflammation [Figure 4]. Langerhans cells were found on microscopic examination, so immunohistochemical staining with CD1A was performed. However, a negative result for this test ruled out Langerhans cell disease.

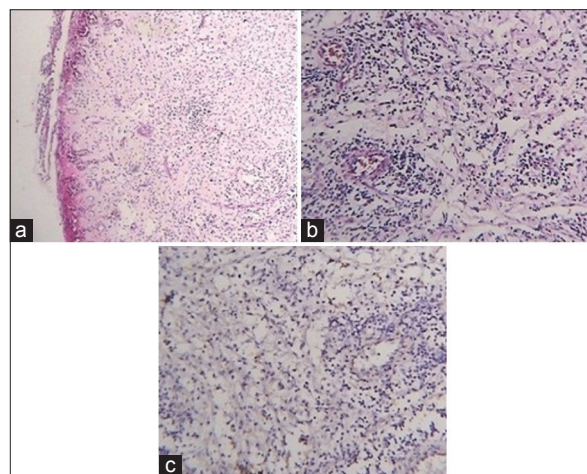


Figure 4: Histological view of soft tissue removed from socket of exfoliated tooth. (a) ×10 exhibits chronic inflammation. (b) ×40 reveals chronic granuloma-like centers. (c) CD1A staining (negative)

On the basis of the history, oral examination, and radiographic evaluation, and because there was no specific cause for this condition, a diagnosis of localized IRR was made, and after placement of a band and loop space maintainer, the patient was followed for 18 months [Figure 5].

In the follow-up period over 18 months, the right involved tooth was clinically asymptomatic; hence, the patient's father did not agree to take new PA radiographs to determine if there was any progression of the root resorption.

DISCUSSION

Cervical IRR begins in the cemento-enamel junction area of the teeth, exhibiting an irregular radiolucency initiating in the periodontal ligament (PDL) on radiography. In the apical types, the resorption starts apically and progresses coronally and radiographically is characterized by gradual shortening and rounding of the remaining root, caused by replacement of the root with normal-appearing trabeculated bone, but ankylosis (fusion of tooth to bone) does not occur and the PDL space is usually visible throughout the entire root surface except for the most apical part.^[29,32,34] Apical IRR has been reported more frequently in males while cervical IRR is more common in females.^[24,25]

The localized type IRR is defined as one to three posterior teeth while the multiple type occurs in more than three teeth; eventually, most of the dentition is involved in a symmetric pattern.^[9] In the multiple tooth cases, the process usually progresses and eventually



Figure 5: Intraoral photograph after 18 months of idiopathic root resorption diagnosis. There were no abnormal signs or symptoms in the follow-up visit

may lead to the loss of several teeth, while the localized types appear to be self-limiting.^[32] However, this is not always the case. In our patient, despite root resorption being localized, it had progressed to the point that the left mandibular primary second molar could be removed by hand. Furthermore, In 2005, Iwamatsu-Kobayashi *et al.* reported a case of multiple cervical IRR with no progression during the follow-up period.^[3]

Hopkins and Adam reported that the interproximal areas appeared to be involved more severely than the buccal and lingual aspects. A higher level of attachment or a higher level of PDL activity in this region may be a possible etiologic factor for these findings.^[14] Lindskog and Hammarström have shown that precementum has anti-collagenase factors that can prevent the enzymatic destruction of this tissue.^[35] Consequently, it is suggested that any trauma to the cervical area or developmental defects such as hypoplasia or hypomineralization of the cementum can be considered etiologic factors for cervical IRR. However, in this case, there was no history of trauma in the affected regions.

In the absence of occlusal function, some atrophic changes in the PDL may occur. Elimination of the cushioning effect of the PDL can lead to increased occlusal stress to the tooth and can induce inflammation. Following the release of inflammatory mediators of local cells, the process of resorption begins.^[28] It has been suggested that a long-term hypofunctional condition may be a possible etiological factor in root resorption.^[36] However, it does not appear to play a role in our case.

The process of root resorption involves a complex interaction of inflammatory cells, resorbing cells, hard tissue, cytokines, and enzymes.^[28] Therefore, any factor that incites the inflammatory process may initiate the process of root resorption. Furthermore, it has been reported that the process can result from acute microbiologically induced osteoclastic activation.

Histological examination of any soft tissue or bone removed from this type of root resorption represents nonspecific chronic inflammation. In the histological evaluation of tissue removed from the socket in our case, nonspecific chronic inflammation was evident. Langerhans cells were also observed on the initial microscopic evaluation, but after immunohistochemical staining with CD1A, Langerhans cell disease was ruled out. However, in the affected right molar, it seems that there was replacement root resorption. We can speculate that there was replacement root resorption at the affected left tooth first, and after mobility was increased to some extent so that bacteria could penetrate into the PDL, it was contaminated by microorganisms and, therefore, became infection-related resorption.

Although many treatments have been proposed to arrest the process of IRR and a number of these methods have been reported useful in animal studies, none of these treatment options are effective clinically in humans, and no interceptive therapeutic regimens are fully indicated.^[21,29] However, based on presented case reports, treatment options, depend on symptoms, and the extension and severity of root resorption, include: Observation, endodontic therapy (in cases with pulpal involvement or cases with root resorption near the pulp), curettage, restoration of cervical lesions and extraction.

In general, a patient with an abnormal pattern of root resorption, especially in the primary dentition, should alert the clinician to rule out all important causes of resorption. More studies should be conducted to further facilitate the clinical management of this pathologic process.

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Conflicts of interest

There are no conflicts of interest.

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