



**Research Article**

**LOCALIZED AGGRESSIVE PERIODONTITIS AND CHILDHOOD HYPOPHOSPHATASIA WITH CONCOMITANT EXTERNAL ROOT RESORPTION: IS IT A CAUSE AND EFFECT RELATIONSHIP?**

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**ABSTRACT**

**Introduction:** Localized Aggressive Periodontitis (LAgP) is characterized by the early age of onset, the rapid rate of alveolar bone destruction and localized molar-incisor involvement. Till date, there are very rare evidences of LAgP occurring along with external root resorption (ERR) with underlying systemic disorder. Presented case report highlights the severe ERR that occurred along with LAgP in the presence of an underlying systemic disorder -Childhood Hypophosphatasia.

Childhood hypophosphatasia is detected at a later age with a consistent feature of premature loss of primary teeth without evidence of a significant inflammatory response. Teeth may show enlarged pulp chamber and significant degree of alveolar bone loss may be seen.

**Case Report:** An 18 year old male patient presented with the complaint of poor aesthetics and replacing his missing anterior teeth. There was no clinically detectable physical abnormality. Clinical examination of the oral cavity was conducted along with biochemical, haematological and radiological investigations. Radiographs revealed vertical bone loss (arc shaped) and severe external root resorption with teeth 26, 36 and 46. Biochemical investigations presented with a just below normal serum profile for alkaline phosphatase, parathormone and serum calcium suggestive of Hypophosphatasia (Childhood Form). No similar family history was reported. A diagnosis of localized aggressive periodontitis was established. However, there were no local or systemic disorders detected that could cause such severe amount of external root resorption. Histopathology report of Extracted molar showed rough cemental surface with hypo-plastic dentinal tubules suggestive of Hypophosphatasia. The differential diagnoses for the etiology of root resorption were LAgP & hypophosphatasia (Childhood form).

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**INTRODUCTION**

Localized Aggressive Periodontitis (LAgP) is characterized by an age of onset at about puberty. It clinically presents with a localized first molar/incisor involvement with interproximal attachment loss on at least two permanent teeth. One of the involved teeth is a first molar, and involving no more than two teeth other than first molars and incisors<sup>[1]</sup>.

Root resorption in permanent teeth may be broadly classified into internal and external resorption. Internal eruption has its origins in the dental pulp whereas external resorption begins in the periodontal ligament (PDL). External root resorption (ERR) has various causes<sup>[2]</sup> including periodontitis<sup>[3]</sup> and systemic disorders (Table-I).

There are very few evidences of simultaneous occurrences of AgP with root resorption in absence of any systemic/endocrine

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disorders and/or syndromes. Systemic disorders showing both aggressive periodontitis & root resorption include Hypophosphatasia, PapillonLefevre Syndrome, Leukocyte Adhesion Deficiency Syndrome, Chediak Higashi Syndrome, Neutropenia, Down's syndrome, EhlerDanlos Syndrome.

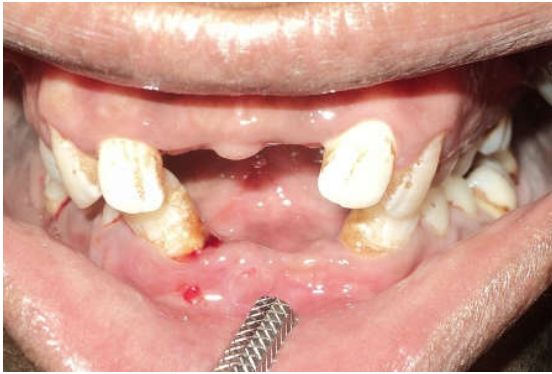
The purpose of presenting the following case was to highlight the severe amount of ERR that occurred along with LAgP in absence of any present systemic disorder or syndrome suggestive of childhood Hypophosphatasia could be the cause of ERR.

**Case Report**

An 18 year old male patient reported with the complaint of poor aesthetics due to missing upper and lower front teeth (Figure-1). History revealed that the anterior teeth were lost due to extreme mobility. Patient did not give any significant past medical history. Patient's parents and siblings too did not report about any similar complaints.

Clinical examination revealed; a fully developed permanent dentition in which the following teeth were missing: 11, 21,

31, 32, 41, and 42. All the teeth present exhibited normal crown morphology, with symmetrical alignment. A generalized probing pocket depth of  $\leq 3$ mm was noted. Tooth 16 exhibited grade-III mobility while all remaining permanent first molars (26, 36, and 46) exhibited grade-II mobility. The gingival soft tissues exhibited minimal inflammation (Figure - 1). The amount of plaque and calculus was moderate. There was no occlusal discrepancy detected (Figure-2). Caries and periapical lesions were absent. Maxillary right first molar was extracted due to excessive mobility.



**Fig 1** Intra-oral Clinical Examination



**Fig 2** Study Model Showing Symmetrical Maxillary and Mandibular Arches



**Fig 3** OPG of the patient showing arc shaped bone loss and root resorption with first molars



**Fig 4** IOPA with 46 showing arc shaped bone loss and severe root resorption.

Radiographs revealed an arc shaped vertical bone loss with all the permanent first molars extending from the second premolar to the second molar. There was extensive root resorption in the permanent first molars till the cervical region (Figure-3, 4).

Teeth 12, 22 exhibited root resorption in cervical region. The alveolar sockets in the first molar region showed hyperostotic bone formation suggestive of reparative activity and chronic low grade infection. The pulp chambers of mandibular canines and all first and second molars were enlarged and elongated (Figure-3). There was periapical bone loss with tooth 43. Dilacerations were seen with 33 and 43. The interdental bone levels were normal in the premolar region (Figure-4). The clinical & radiographic features were suggestive of localized aggressive periodontitis.

In order to detect for the presence of any systemic disorder causing bone destruction and root resorption; biochemical investigations were advised (Table-II).

**Table I** Systemic/Endocrine causes of Root Resorption

Sr. No.	Author(s)	Systemic or Endocrine Disturbance
1.	Sunde and Hals (1961)	Hypoparathyroidism
2.	Goultshin <i>et al</i> (1982)	Hyperparathyroidism
3.	Tangney <i>et al</i> (1979) <sup>[4]</sup>	Hypophosphatasia
4.	Eyring and Eisenberg (1968)	Hyperphosphataemia
5.	Rudiger and Berglundh (1999) <sup>[5]</sup>	PapillonLefevre Syndrome

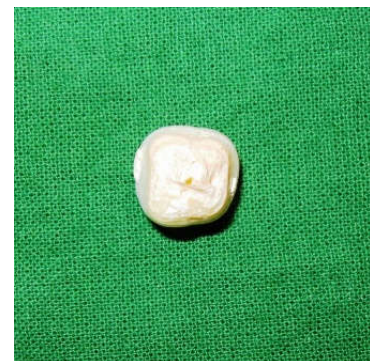
**Table II** Biochemical Investigations

Parameter	Normal Values	Results in patient
1. Serum Alkaline Phosphatase	30-115 U/lit	32 U/lit
2. Serum Calcium	2.1-2.6 mmol/lit	2.0 mmol/lit
3. Parathormone	10-60 pg/ml	28 pg/ml

The extracted maxillary right first molar was sent for histological examination.



**Fig 5** Extracted tooth 16 showing root resorption



**Fig 6** Concavities on root surface suggestive of external root resorption

Ground section revealed concave areas of resorption on the remaining cemental surfaces (Figure-7). The rough cemental surfaces with the evident pattern of resorption of roots were suggestive of ERR (Figure-5, 6). The predentin layer was defective in the coronal as well as radicular portion while the dentinal tubules were hypoplastic suggestive of hypophosphatasia.

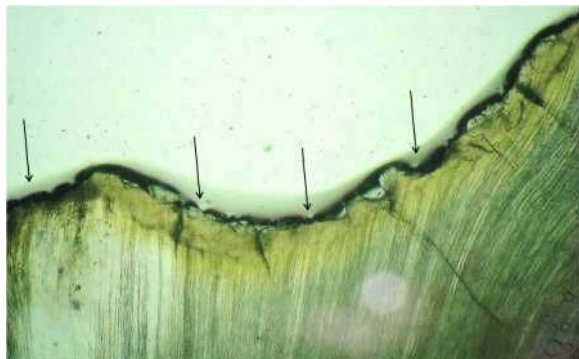


Fig 7 Ground section at 10 X magnification showing resorption lacunae.

## DISCUSSION

This article reports a case of LAGP with ERR at a young age in absence of any significant local factors or detectable systemic disorders. With a normal serum biochemical and hormonal profile and an absence of local or systemic etiological factor detectable; no absolute etiology was identified.

In the available literature, simultaneous occurrence of LAGP with ERR in absence of any systemic disorder is hardly documented. Lindsog & Blomlof (1983) [6] in a scanning electron microscopic study concluded that roots of teeth affected by aggressive periodontitis showed extensive areas of cementum hypoplasia with exposed dentinal tubules. The hypoplasias were found also on the infracrestal part of the root indicating that they had not been caused by exposure to the oral environment or by any treatment. Cementum hypoplasias were not found in any of the healthy teeth from the control group.

The rate of disease progression slows down or stops entirely in a small percentage of patients with LAGP (Lang, 1999). Antonio CA *et al* (1999) [8] quoted that the spread of root resorption is associated with inflammation, and the capacity for repair of root resorption is diminished with greater severity of periodontitis. We can thus presume that the cementum was hypoplastic as result of LAGP in the current case and was resorbed during an exacerbated phase of disease. While currently, the lack of inflammation and the reparative activity visible radiographically in the alveolar sockets of molars signifies that the disease is in a passive phase.

The significant amount of bone loss and resorption pointed towards a plausible role of hypophosphatasia. Four types of hypophosphatasia are recognized, depending on the severity and the age of onset of the symptoms [7]. Generally, the younger the age of onset the more severe is the expression of the disease. As some patients grow older, serum alkaline phosphatase levels may reach normal [7].

Childhood hypophosphatasia is usually detected at a later age and has a wide range of clinical expression. A consistent feature is the premature loss of the primary teeth without evidence of a significant inflammatory response. Features of hypophosphatasia in the current case include alveolar bone loss, varying levels of gingivitis, large pulp chambers, loosening and premature loss of permanent incisors, cementum hypoplasia and resorption. But the serum levels of alkaline phosphatase are within the normal range, although towards the lower end. We can only hypothesize that initially patient had hypophosphatasia and currently with increasing age the serum levels of alkaline phosphatase have attained a normal level.

## CONCLUSION

The diagnosis of LAGP was made upon the clinical and radiological evidences. Based on the presenting features and Histopathology reports, following differential diagnoses for the etiology of ERR were formulated:

1. Aggressive Periodontitis
2. Hypophosphatasia (Childhood Form)
3. Idiopathic Root Resorption

The etiopathogenesis of root resorption in the current case was obscure. At present there is no preventive or therapeutic regimen available for the type of root resorption in this case report. Further investigations are needed to explore the possible mechanisms of simultaneous occurrence of both the entities. We, hence, stress the fact that patients with multiple root resorptions should be screened for aggressive periodontitis as an underlying etiological factor.

## References

1. Lang N, Bartold PM, Cullinan M, *et al*: Consensus report: Aggressive periodontitis. *Ann Periodontol* 1999;53.
2. Fuss Z, Tsesis I, Lin S. Root resorption-diagnosis, classification and treatment choices based on stimulation factors. *Dent Traumatol* 2003; 19:175-182.
3. Rodriguez-Pato RB. Root resorption in chronic periodontitis: A morphometrical study. *J Periodontol* 2004; 1027-1032.
4. Tangney NJ. Hypophosphatasia: A case report and literature review. *Ir Med J* 1979; 72: 530-531.
5. S Rudiger, T Berglundh: Root resorption and signs of repair in Papillon-Lefevre syndrome. A case study. *Acta Odontol Scand* 1999; 57: 221-224.
6. Lindsog S, Blomlof L. Cementum hypoplasias in teeth affected by juvenile periodontitis. *J Clin Periodontol* 1983; 10: 443-451.
7. Neville BW; Oral Manifestations of Systemic Diseases; Oral and Maxillofacial Pathology 2<sup>nd</sup> Edition; 2002: 730 - 732.
8. Antonio Crespo Abelleira, Maria Angeles Rodriguez Cobos, Isaac Manuel Fuentes Boquete, Maria Teresa Castano Oreja, Francisco Javier Jorge Barreiro, Raquel Beatriz Rodriguez Pato. Morphological study of root surfaces in teeth with adult periodontitis. *J Periodontol* 1999; 70: 183-1291.

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