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Invasive Cervical Resorption and the Oro-Facial Cleft Patient: A Review and Case Series

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ABSTRACT

Introduction: Invasive Cervical Resorption (ICR) has an unknown aetiology, yet it exhibits very aggressive behaviour compared with typical external root resorption, posing a high risk of tooth loss.

Aim: To investigate the number of patients at the Dublin Cleft Centre with an oro-facial cleft who experienced ICR and to identify any possible aetiological factors.

Materials and Method: A retrospective investigation of all oro-facial cleft patients treated at the Cleft Centre, St James's Hospital, Dublin. All patients' clinical and radiological records were reviewed. Patients where tooth loss became inevitable due to Class 4 ICR were analysed.

Results: From 588 oro-facial cleft patients, 14 (2.38%) patients with ICR were identified. Of these 8 (57%) were female and 6 (43%) were male. Mean age at diagnosis was 28 years (range = 16-49 years). Cleft type: 6 (42.1%) unilateral cleft lip and palate, 8 (57.9%) bilateral cleft lip and palate. SeventeenICR affected teeth in total, with eleven (65%) maxillary central incisors, two (12%) maxillary lateral incisors, four (23%) maxillary canines, and one (7%) central, lateral and canine affected. Some, (n=10, 71.4%) presented with ICR resulting in immediate tooth loss. Other patients (n=4, 28.6%) developed ICR during or following prosthodontic treatment at the Cleft Centre. Tooth loss for this cohort, though not immediate, was inevitable. All had undergone fixed orthodontic appliance treatment and twelve had received dento-alveolar bone grafts. A number (n=7, 50%) had undergone osteotomy, two (14%) had received night guard vital dental whitening and one had a history of trauma.

Conclusions: ICR, given its aggressive nature and ill-understood aetiology, poses significant treatment challenges. The most severe form of ICR (Class 4) leads inevitably to tooth loss. The slow–moderate progression of ICR may explain the late presentation found in this study, reinforcing the importance of long-term follow-up of this special dental care group.

INTRODUCTION

Oro-facial clefting is the most common congenital malformation of the head and neck.[1] Dental anomalies in the anterior and pre-maxillary area are long-recognised in oro-facial clefting, and these include abnormalities in tooth number, size, form, development, position and eruption.[2-6] Dental crown anomalies include enamel hypoplasia, microdontia, macrodontia and dilacerations, whilst root malformations may range from delayed development to dilaceration, interrupted formation or microform giving rise to unfavourable crown root ratios.[3,8] These defects may be attributed to the cleft itself, to genetic factors or to traumatic factors relating to surgery and resultant scar tissue formation.[2,7]

Invasive cervical resorption (ICR) is a relatively uncommon and often aggressive form of external root resorption that can affect any tooth in the permanent dentition.[9] ICR is characterised by aggressive invasion of the cervical region of the root by fibro-vascular tissue, which progressively resorbs dentine enamel and cementum leading to progressive destruction of the tooth structure at the zone of connective tissue attachment.[10-12] The clinical appearance of teeth affected by ICR varies considerably depending upon the extent of the resorptive process. Some teeth may display an obvious 'pinkish' colour in the tooth crown as the highly vascular resorptive tissue becomes visible through the thin residual enamel,[9] although more frequently, the condition is only detected radiographically,[13] often as an incidental finding. Histologically, the pulp remains protected by a thin layer of pre-dentine until late in the process. More advanced lesions may display fibro-osseous characteristics with deposition of ectopic bone-like calcifications, both within the resorbing tissues and directly on the dentine surface.[14]

The aetiology of ICR is unknown.[9,14] Physical or chemical trauma to the cemento-enamel junction region is considered a significant predisposing factor.[9,14,15] Trauma may be direct or associated with orthodontic tooth movement, periodontal or dentoalveolar surgery, tooth transplantation, bone grafting, segmental maxillary orthognathic surgery, intra-coronal bleaching and tetracycline conditioning of roots.[10,11,15] Additionally, ICR has been found in association with heat damage to bone or where there has been impairment of blood supply at the cemento-enamel junction.[15] Systemic factors such as viral infections, hormonal abnormalities and renal pathology have also been reported in association with ICR,[15] and there is familial predisposition.[16]

It is important to differentiate ICR from internal resorption, and a diagnosis is normally achieved by both clinical and radiographic examination. Long cone periapical views using a parallax technique can be taken in order to follow the outline and continuity of the pulp chamber. The lesion of ICR will appear to move on the films with the X-ray tube angulation.[17] The use of cone beam computed tomography has been advocated[18] as in addition to detecting the presence of a resorptive defect, it will show its extension in three dimensions and whether or not it has invaded the pulp. Treatment of ICR is difficult, and the outcome is often uncertain.[9, 19-21] The main aim of treatment is to completely remove the resorptive tissue with an excavator or slow-speed bur, followed by conditioning of the dentinal walls to remove any remnants of resorptive tissue before restoration. There appears to be no consensus as to the most appropriate restorative material, with glass-ionomer cement, composite resin, amalgam, Mineral Trioxide Aggregate and calcium-enriched mixture cement all having been advocated.[22-25] To facilitate treatment planning, Heirthersay[9] divided ICR into four distinct clinical types based upon the severity of the lesion (Table 1). The poor survival rate of Class 4 lesions indicate that while the affected tooth may be left to progressively resorb, its extraction and prosthodontic replacement is inevitable.

Approximately 100 oro-facial cleft affected children are born in Ireland annually. These children require comprehensive and complex multidisciplinary care from early infancy through to early adulthood. In addition to a range of essential medical specialties and therapies, dental specialties include paediatric dentistry, orthodontics, maxillofacial surgery and advanced restorative-prosthodontic care. These have significant time and cost implications for the patient and the health system provider. In 2004, an Advanced Restorative Clinic was established at the Cleft Centre, St James's Hospital, Dublin, and since then, care has been provided for over 500 patients.

AIM

The aim of this study was to investigate the number of oro-facial patients at the Dublin Cleft Centre who experienced ICR and to identify any possible aetiological factors.

METHOD

A retrospective investigation was carried out on all patients treated at the oro-facial Cleft Clinic of the Maxillo-facial Department, St James's Hospital, Dublin. All patients treated since the clinic opened in 2004 had their clinical and radiographic records reviewed. The records of ICR patients where tooth loss became inevitable underwent a detailed analysis, and the following parameters were recorded:

- Condition of crown (including enamel hypoplasia and size and form of crown)
- Condition of root (including size, dilaceration, pre-existing external root resorption, vitality and apical form as determined radiologically
- Gender and ethnicity
- Cleft type, cleft side and if syndromic or non-syndromic
- Surgical history (including bone grafting, implant insertion, fistula repair, osteotomies, repeat or additional surgery (such as late lip or nasal reconstruction))
- Dental history (including previous orthodontic treatment, crown whitening, restorative or prosthodontic procedures).
- Trauma history (independent of any dental intervention)
- Age at identification and treatment duration before tooth loss became inevitable where ICR developed during prosthodontic operative care

RESULTS

The records of 588 cleft lip and palate (CLP) patients were reviewed and 14 (2.38%) patients were identified with ICR, of which 8 (53%) were female and 6 (47%) were male. A summary of results is shown in Table 2. All patients were Caucasian and all were non-syndromic CLP. The age range of patients was 16-49 years (mean = 28 years). Cleft types of these patients were identified as 6 unilateral CLP (3 right-sided and 3 left-sided) and 8 bilateral CLP. There were 17 ICR affected teeth in total, and these were all found in the upper arch, of which 11 (65%) were maxillary central incisors, 2 (12%) were maxillary lateral incisors, and 4 (23%) were maxillary canines. The majority 12 (86%) of patients had one ICR affected tooth, whilst 1 (7%) and 1 (7%) of patients had two and three ICR affected teeth respectively. Bilateral CLP was present in both of the cases with more than one ICR affected teeth. In the single patient with three ICR affected teeth, the maxillary central, lateral and canine on the same side were affected. In all of the patients with unilateral CLP, the ICR affected tooth was on the same side as the cleft. A developmental

crown anomaly was found in 12 (85%) patients, and a developmental root anomaly was found in 9 (64%) patients. Over half (n=8, 57%) of patients had a combined crown-root anomaly.

All of the patients had received orthodontic treatment, and 2 (14%) had received vital night guard dental whitening. Only one patient gave a history of dental trauma. This male patient presented with a non-vital maxillary central incisor prior to the development of ICR. The majority (n=13, 93%) of patients had undergone surgical bone grafting, 7 (50%) had received a maxillary advancement osteotomy, 6 (43%) had received a surgical fistula repair and 4 (29%) had received additional surgery for late lip and/or nasal reconstruction. Most (n=10, 71%) of the patients presented to the prosthodontic clinic with an existing ICR affected tooth, and all of these patients were symptomless (Figures 1-6). Four patients (29%) developed ICR during or following attendance to the prosthodontic clinic. These were all symptomless. One of these patients developed ICR immediately after placement of a minimally-prepared resin bonded bridge, with loss of the tooth within a 15 month period (Figure 4). Another patient lost two teeth, a maxillary central incisor and maxillary canine, one year and two years respectively following temporary crown preparation. The remaining two patients, who had received prosthodontic care, developed ICR following discharge, losing the affected teeth five years (Figure 5) and six years (Figure 6) later.

DISCUSSION

Preserving the integrity of both hard and soft dental tissues is central to oro-facial cleft care. Healthy dental tissues not only benefit and assist the various necessary operative procedures, but are vital to long-term oral health, function, facial aesthetics and quality of life outcomes. It is recognised that surgery, orthodontic tooth movement, intra-coronal bleaching and prosthodontic operative procedures may have an effect upon dental root structure.[8,9] Control of these risk factors is particularly critical in oro-facial cleft-affected patients, given that the available dental tissues, both hard and soft, are frequently developmentally compromised.

ICR is an uncommon but aggressive form of external root resorption. Its aetiology is poorly understood but trauma, alone, or in association with dental or surgical operative procedures is a recognised predisposing factor. Treatment of ICR is difficult leading to an uncertain long-term prognosis.[8-10] The fundamental objectives in managing ICR are to expose the defect, remove the granulation tissue and to

seal the defect.[12,19] Heithersay's clinical classification can provide guidance to clinicians in assessing and managing the affected teeth, and careful case selection is advised in order to achieve a good prognosis.[9,10] Heithersay recommends treating Class I-3 cases. Management of Class 4 ICR is difficult and has a high risk of failure. Therefore he considers that Class 4 ICR cases may be left in-situ untreated for as long as they remain asymptomatic.[9-11] Otherwise, extraction is the only viable option, as ultimately was the case for all affected patients in this present study.

In our study, the ICR affected maxillary central, lateral and canine teeth were symptom free at presentation in all cases. Our study also supports previous findings that maxillary central incisor teeth are the most frequently affected and it is thought it is because these teeth are more prone to dental trauma.[26] Cleft patients are no more susceptible to trauma than the non-cleft population but incisors are most at risk. All of the unilateral CLP patients had just one ICR affected tooth and this was invariably associated with the cleft side. More than one affected ICR tooth was associated with bilateral CLP patients, and in this study the ICR affected teeth were confined to the cleft side. Common to all subjects was a history of fixed orthodontic treatment.

In patients who developed ICR unexpectedly during prosthodontic care, no common predisposing factor could be determined. Neither could the duration before tooth loss became inevitable be predicted.

While the severity of the cleft influenced the number of ICR affected teeth, no association with the number of ICR affected teeth and the range or extent of surgical procedures could be found. In addition no association with age and ICR was found. Interestingly nine of the patients were over 25 years of age at presentation. Given the recognised improvement and refinements in surgical cleft care techniques in recent years, the possibility of the cohort in the present study being subjected to less conservative surgical procedures could not be discounted. We recognise that we are reporting observations on a small number of cleft patients and that these were from one centre. Nevertheless the fact that ICR developed late and that this is a difficult condition to treat highlights the need for long term follow up of cleft patients.

CONCLUSIONS

This paper reports on the experience of one specialist Cleft Centre regarding patients with ICR. As with similar Cleft Centres, patients underwent extensive specialised multidisciplinary dental care over a period of many years. This treatment included plastic surgery, orthodontics, prosthodontics, orthognathic surgery, fistula repair – all procedures recognised to have inherent predisposing traumatic risks. Whilst some patients presented to the prosthodontic clinic with ICR and tooth loss was immediately recognised, for others, the onset of ICR at or after prosthodontic care had been completed was unexpected. In neither group could the onset of ICR be predicted. This paper reinforces the importance of long-term review of oro-facial cleft affected patients in order to diagnose and treat possible cases of ICR at an early stage so as to avoid further tooth loss from an already compromised dentition.

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Table 1. Heithersay's classification of ICR[9]

Class	Description
1	A small invasive resorptive lesion near the cervical area with shallow penetration into dentine

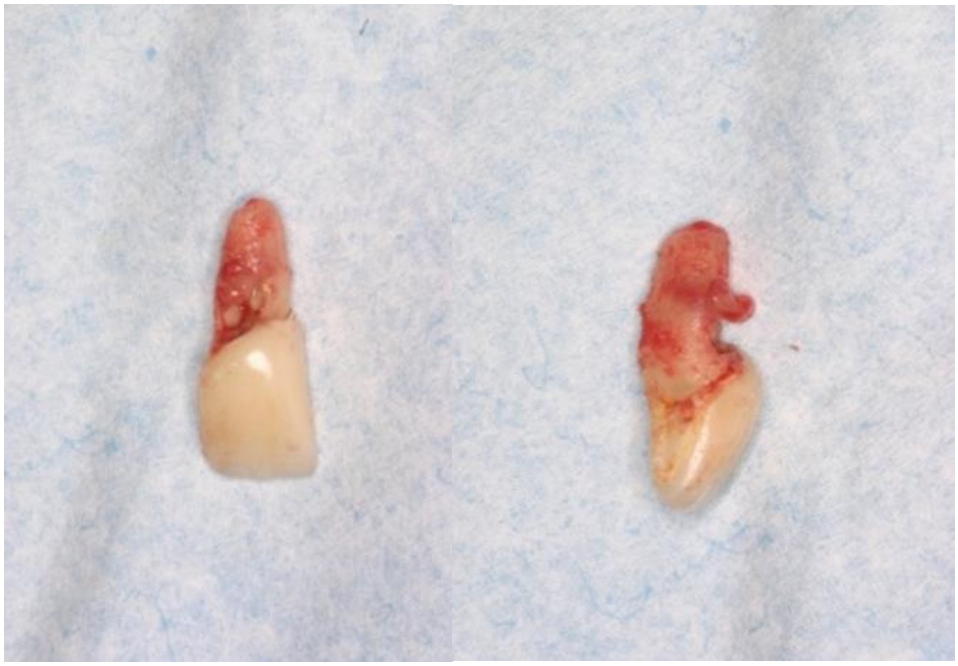
2	A well-defined invasive resorptive lesion that has penetrated close to the coronal pulp chamber but shows little or no extension into the radicular dentine
3	A deeper invasion of dentine by resorbing tissue not only involving the coronal dentine but also extending into the coronal third of the root
4	A large invasive resorptive process that has extended beyond the coronal third of the root

Table 2. A summary of the 14 patients with class 4 ICR

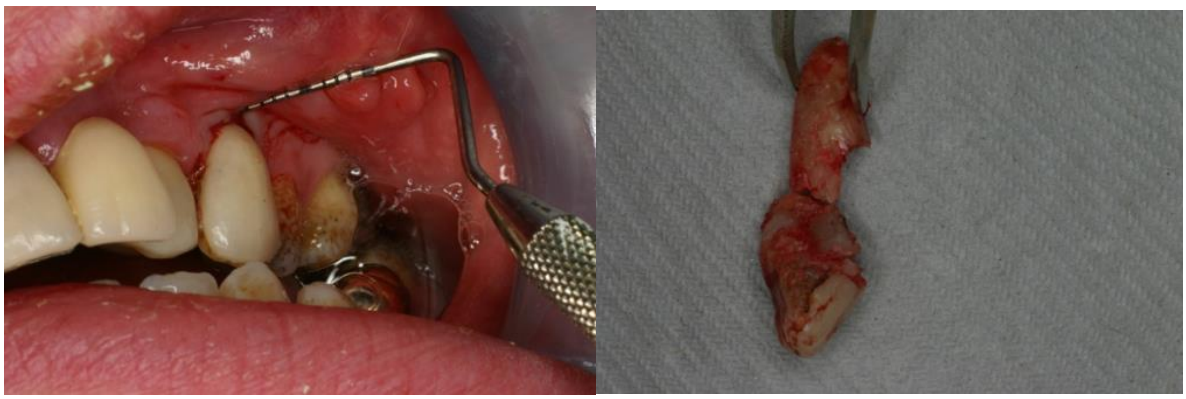
Case	1	2	3	4	5	6	7	8	9	10	11	12	13	14
Gender M:F	F	F	F	M	M	F	M	F	F	M	M	F	M	F
Caucasian	√	√	√	√	√	√	√	√	√	√	√	√	√	√
UCLP Side	√ R	–	√ R	√ L	–	–	√ L	√L			√ R			
BCLP	–	√	–	–	√	√	–	–	√	√	–	√	√	√
Non Syndromic	√	√	√	√	√	√	√	√	√	√	√	√	√	√
Developmental Crown Anomaly	√	√	√	–	√	–	√	√	√	√	√	√	√	√
Developmental Root Anomaly	√	√	–	√	√	–	√	–	√	√	√	–	–	√
Bone Graft	√	√	√	√	√	√	√	√	√	–	√	√	√	√
Osteotomy	–	√	√	√	–	–	–	–	–	√	–	√	√	√
Fistula Repair	√	√	–	–	–	–	–	–	–	–	√	√	√	√
Tooth/Teeth ICR affected	UR 1	UL 2	UR 3	UL 1	UL1,2, 3	UR 3	UL 1	UL 1	UL 1	UL 1	UR1	UL1	UL1	UL1,3
ICR: Age at identification	16	21	28	37	27	29	49	26	17	33	29	23	38	23 years
ICR presentation immediate tooth loss	√	√	√	√	√	√	√	√	√	√	–	√	–	–
ICR during/after prosthodontic treatment	–	–	–	–	–	–	–	–	–	–	√	√	√	√
Duration to tooth loss	–	–	–	–	–	–	–	–	–	–	6 years	15 months	5 years	UL1 1Year, UL 3 2 years
Type of prosthodontic treatment	–	–	–	–	–	–	–	–	–	–	*A	*B	*C	*D
Orthodontic Treatment	√	√	√	√	√	√	√	√	√	√	√	√	√	√
Tooth Whitening	–	–	–	–	–	–	–	–	–	–	–	–	√	√
Trauma	–	–	–	–	–	–	–	–	–	–	√	–	–	–
Additional Surgeries	–	–	–	–	√	–	–	–	–	–	–	√	√	√
Implant	–	–	–	–	–	–	–	–	–	–	–	–	√	–

***A=RCT & bridge preparation *B=minimal preparation resin bonded bridge *C=Veneer & implant adjacent *D=Crown preparation and temporaries**

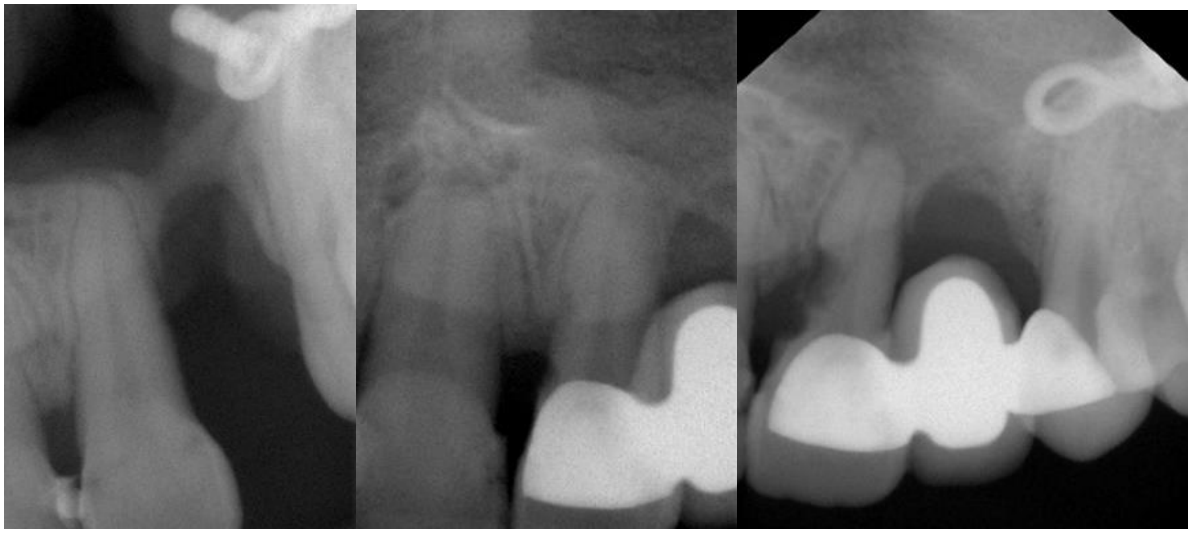
Figures 1a and 1b: An example of an ICR affected maxillary canine following extraction



Figures 1c and 1d: An example of an ICR affected maxillary central incisor following extraction



Figures 2a and 2b: Pre-extraction and post-extraction images of a class 4 ICR affected maxillary canine that suffered general enamel hypoplasia and had been restored with a composite veneer



Figures 3a-3c: Case 12 showing pre-oronasal fistula repair and post-maxillary advancement osteotomy (3a), post-fistula closure and 10 months post-placement of a minimally prepared resin bonded bridge (3b & 3c)

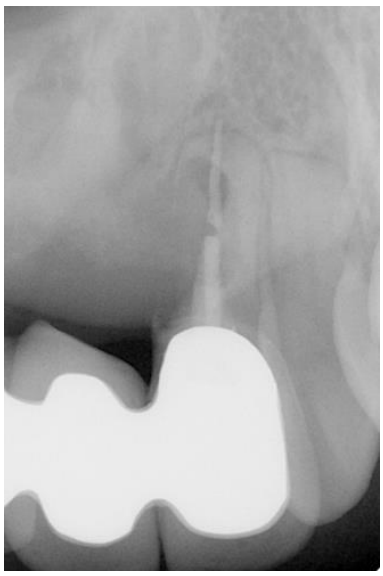


Figure 4: Case 11 presenting 6 years following prosthodontic treatment and discharge

Figures 5a-5d: Case 6 showing a class 4 ICR affected maxillary right canine. Note the 'pinkish' colour at the cervical margin

Figures 6a-6d: Case 13 showing a class 4 ICR affected maxillary left central incisor. Implant and bone graft had been completed 4 years previously