

Prevalence of dental abscess in a population of children with vitamin D-resistant rickets

Alton G. McWhorter, DDS, MS N. Sue Seale, DDS, MSD

Abstract

A population of patients with vitamin D-resistant rickets whose present ages range from 4 to 22 years was examined to determine the prevalence of dental abscess associated with that condition. For those patients affected with abscesses in the primary dentition, four associated factors were examined: 1) the age at onset of the first abscess; 2) the tooth/teeth most commonly involved; 3) whether or not other teeth were involved over time; and 4) the order of occurrence of multiple abscesses. Six of 24 patients (25%) were affected with abscesses of the primary dentition; all had multiple abscesses. Males were more commonly involved than females. The sequence of involvement generally followed the pattern of tooth eruption; however, dental treatment in susceptible individuals shortened the time between eruption and abscess. Medication dose and measurement of radiographs were not reliable predictors of the occurrence of abscesses in this population; however, the results indicate that one abscess is a predictor of future abscesses for that patient.

Introduction

Vitamin D-resistant rickets (VDRR) is a metabolic disturbance which causes defective calcification of mineralized structures. This condition was first described in 1937 by Albright et al., who reported a case of a child who had received treatment for rickets for 10 years yet continued to have the active disease. The underlying cause of vitamin D resistance is a defect in inorganic phosphate transport that is parathyroid hormone (PTH) dependent (Arnaud et al. 1971; Glorieux and Scriver 1972). A primary disturbance in intestinal absorption of inorganic phosphate, leading to the formation of insoluble calcium-phosphate complexes in the intestinal lumen was proposed by Short et al. (1973) as a possible mechanism of calcium malabsorption. Blood studies of patients with VDRR generally exhibit normal or low serum calcium, marked lowering of serum inorganic phosphate, and elevation of serum alkaline phosphatase.

This condition has long been known to be associated with abnormalities of both the primary and the permanent dentitions (Hall 1959; Gardner et al. 1969; Vasilakis et al. 1980). Commonly reported dental abnormalities include normal but thin enamel (Gallo and Merle 1979; Seow and Latham 1986), interglobular dentin (Archard and Witkop 1966; Breen 1986) and enlarged pulp chambers with the pulp horns reaching to or beyond the dentoenamel junction (DEJ) (Tracy and Campbell 1968; Özkan et al. 1984; Bender and Naidorf 1985). The combination of these factors can lead to pulpal abscess once the integrity of the enamel is compromised. Minor caries or even physiologic attrition can remove the enamel and allow microorganisms to gain access to the pulp through the poorly formed dentin. The close proximity of the pulp to the tooth surface compounds the problem by speeding up the infective process. Teeth that appear healthy can "spontaneously" abscess.

In the literature, a primary source of information concerning dental problems associated with VDRR is case reports involving a single patient or small populations (Albright et al. 1937; Daeschner 1957; Harris and Sullivan 1960; Marks et al. 1965; Tracy and Campbell 1968; Cohen and Becker 1976; Rakocz et al. 1982; Seow and Latham 1986) and reports of VDRR patients with multiple abscesses are common. Review of these case reports gives the reader the impression that every patient diagnosed with VDRR is destined to have abscessed teeth. There are no studies with large enough patient samples to allow accurate analysis of the actual incidence of dental abscesses in individuals diagnosed with VDRR. Examination of a group of patients to determine incidence and potential predictors of abscess can lead to a more rational treatment approach aimed at prevention of abscesses and preservation of the primary dentition, rather than emergency care for abscesses after they occur.

Therefore, the present study was undertaken to answer the following questions.

1. What is the prevalence of dental abscesses in the primary dentition of a population of patients diagnosed with VDRR?
2. Does the abscess of one tooth in these patients indicate that others will abscess?
3. Which tooth/teeth are more commonly involved?
4. Is there an order of occurrence?
5. Can predictions be made of who will demonstrate abscesses or which teeth in an affected patient will abscess?

Materials and Methods

Twenty-five individuals diagnosed with VDRR treated at Texas Scottish Rite Hospital for Crippled Children (TSR) in Dallas, TX, over the past 20 years served as the sample for this study. This number represents virtually all of the children being followed for VDRR at the Hospital and is composed of members from 18 different families. Medical information pertaining to the course of the disease condition itself was obtained by examining the patient's hospital record. This information included the patient's age at the time of medical diagnosis and the type, onset, and duration of treatment.

Dental history was gathered in two ways: by examining the patients and the dental records in the dental clinic at TSR; or by taking an oral history from the parent, guardian, or family dentist. Many of the patients had received at least part of their dental treatment in the dental clinic at TSR; therefore, the information was available in the hospital chart. If the chart did not provide the desired information, the parents or guardian were informed by mail of the purpose of the study and their consent to participate was obtained. The investigator then contacted them by phone to obtain a brief dental history of their child. For those children who had abscesses in the primary dentition, the investigator obtained permission to contact the dentist who had treated the child (if treatment had not been rendered in the Hospital's dental clinic).

Available diagnostic dental radiographs, both bite-

wings and periapicals exposed at the initial or recall appointments using accepted standard techniques of both exposure and processing, were examined for findings commonly reported with VDRR (pulp horns extending to or beyond the DEJ and enlarged pulp chamber). These characteristics have been implicated in tooth abscesses in VDRR patients.

Drug dose, serving as an indication of disease severity, was compared to the occurrence of abscesses to test its reliability in predicting susceptible individuals.

For patients who demonstrated a dental abscess in the primary dentition, four associated factors were examined:

1. The age at onset of the first abscess
2. The tooth/teeth most commonly involved
3. Whether or not other teeth were involved over time
4. The order of occurrence of multiple abscesses.

The prevalence of abscesses in this population was represented as the percentage of occurrence.

Using the "Chronology of the Human Dentition" chart modified by Lunt and Law (1974), the age at the time of each abscess was used to determine the approximate time that the tooth had been present in the oral cavity. Teeth that abscessed following operative treatment also were included in the study, but teeth with large carious lesions prior to treatment were not.

Results

The final population of patients consisted of 14 females and 10 males whose current ages ranged from 4

Table 1. Individuals demonstrating dental abscesses in the primary dentition

Patient Number	Age at Onset	Age at Last Abscess	Current Age	Maximum Dosage*	Sequence of Involvement
111 [†]	3y 0m	7y 2m	15y 4m	24	E, F, S, K, G, H, L, T
114 [‡]	3y 1m	7y 7m	13y 9m	480	N, F, C, M, H
122 [§]	3y 9m	4y 5m	5y 8m	80	F, E
115	4y 1m	8y 3m	12y 4m	440	E, G, P, Q, C, S, H, K
120 [§]	4y 6m	6y 3m	7y 2m	100	F, Q, G
118	5y 5m	9y 0m	10y 0m	300	L, N, O, P, Q, B, S, I

* Dosages are in thousands of units of vitamin D per day.

[†] Teeth E, F, S, and K were lost when the patient was very young. The mother states that E and F were lost about the time the child was three, but she had no idea when S and K were lost. Dental records were unavailable.

[‡] Patient also had 6 additional teeth abscess but all had received prior operative treatment. The teeth involved were K, B, I, A, J, and L. If these teeth were inserted in the sequence of involvement, it would be as follows: N, F, K, B, I, A, J, C, L, M, H.

[§] Siblings.

^{||} Patient also had 2 additional teeth abscess but both had received prior operative treatment. The teeth involved were J and K. If these teeth were inserted in the sequence of involvement, it would be as follows: L, N, O, P, Q, B, S, I, J, K.

to 22 years. One of the original 25 patients was dropped from the study because she had been discharged from the hospital's care and could not be reached by mail or phone. Of the remaining 24, six (25%) had abscesses of primary teeth. One of 14 females and five of 10 males were affected.

The range of onset of dental abscesses in these six patients was 3 years 0 months to 5 years 5 months (mean, 4 years 0 months). No primary teeth were involved beyond 9 years 0 months of patient age. The patients in the present study who had teeth "spontaneously" abscess had a minimum of two and a maximum of eight. The only affected female, a sibling of one of the affected males, had abscesses of three teeth. Two male patients had eight posterior teeth (two in one patient and six in the other) abscess following operative treatment. The forms of treatment included: one sealant, five occlusal amalgams, one stainless steel crown, and one pulpotomy followed by a stainless steel crown. Whether or not these teeth would have abscessed without operative intervention cannot be determined. These findings are summarized in Table 1 (see previous page).

One affected male had a sister in the study group who was not affected by dental abscesses, and another had two female cousins diagnosed with VDRR who experienced no problems with their primary dentition.

In the six patients, a total of 42 teeth abscessed. Of this number, 34 abscessed without gross caries or previous treatment. The remaining eight teeth abscessed after receiving treatment and were all posterior teeth. Fifty-seven per cent of the total were anterior teeth and 43% posterior. Mandibular and maxillary teeth were involved equally. Using the estimated times of eruption described by Lunt and Law (1974), the average time that each tooth had been present in the oral cavity was determined. Incisors had been

present for a mean of 48 months following eruption, cuspids for a mean of 65 months, and molars for a mean of 68 months. When the posterior teeth that had been treated previously are examined separately, these teeth abscessed after a mean of 51.5 months. These findings are summarized in Table 2.

Medication doses were examined for ages 3 through 10 to determine if those patients demonstrating abscesses were taking larger daily doses than patients who did not. The time frame of ages 3 through 10 was chosen because all of the abscesses occurring in this population fell into this age range. The dose information for one patient who had abscesses of primary teeth could not be included in the calculation of the average doses per age. This patient had been treated with a different form of the vitamin and no comparable conversion to thousands of units per day could be made.

Table 2. Comparison of average times since eruption (incisors vs. cuspids vs. molars)

Tooth	Times Involved	Months from Eruption to Abscess				Total	
Incisors							
D	0	-	-	-	-	0	
E	3	39	43	26	-	108	
F	4	28	44	35	26	133	
G	3	52	64	75	-	191	
N	2	57	22	-	-	79	
O	1	62	-	-	-	62	
P	2	62	58	-	-	120	
Q	3	57	53	58	-	168	
	18					861	Avg 48
Cuspids							
C	2	52	64	-	-	116	
H	3	72	79	67	-	218	
M	1	54	-	-	-	54	
R	0	-	-	-	-	0	
	6					388	Avg 65
Molars							
A	1	*36	-	-	-	36	
B	2	56	†44	-	-	100	
I	2	92	†44	-	-	136	
J	2	†79	†41	-	-	120	
K*	3	†81	†32	72	-	185	
L	3	49	†55	70	-	174	
S*	2	72	74	-	-	146	
T	1	59	-	-	-	59	
	16					956	Avg 68
							Avg 60[‡]

* Additional tooth lost due to abscess; dental records were unavailable to determine the time lapsed since eruption.

† Tooth operatively treated prior to abscess.

‡ Includes teeth which abscessed following operative treatment. The average time in the mouth before abscess for those teeth that had been treated prior to the abscess was 51.5 months.

Comparison of medication dose with the occurrence of abscess indicated that, with the exception of the 3 year age group, the patients demonstrating abscesses were taking larger average doses of vitamin D than those patients not affected by abscesses. Table 3 shows the size of the groups used to calculate the mean dose at each age.

Examination of dental radiographs revealed the presence of elongated pulp horns and enlarged pulp chambers in some of the affected patients, but the finding was not consistent. These characteristics also were present in some of the VDRR patients who did not have abscesses in the primary dentition.

Discussion

The results of this study indicated that abscesses in the primary dentition associated with VDRR are not as prevalent as commonly believed. Six of 24 patients (25%) diagnosed with the condition manifested abscesses of primary teeth. This population represents virtually all of the children presenting to Texas Scottish Rite Hospital for Crippled Children with VDRR over the past 20 years and therefore, though small, is believed to constitute a valid sample which is representative of the disease.

Each patient's medication history for those being treated between the ages of 3 and 10 was examined to determine if medication dose, serving as an indication of the severity of the disease, could be used to predict the occurrence of abscesses. Overall, patients who manifested abscesses in the primary dentition were taking larger doses of vitamin D than those patients who did not. However, in this population, it was determined that vitamin D dose could not be shown to be reliable for predicting the occurrence of abscesses due to the small numbers that could be included in the calculation of each mean. In the group demonstrating abscesses, information from only five patients could be used (one could not be included due to the type of medication prescribed).

Enamel and dentin formation of the primary dentition occurs between approximately 4 months in utero and 11 months of age, and it is during this time that the defects in primary tooth development which may predispose the patient to abscesses occur. For this reason, it seems unlikely that even the early onset of medical treatment in these patients will have a profound effect on the development of the primary dentition, therefore preventing future dental problems associated with the primary dentition. However, the permanent teeth form after birth, and their development could possibly be affected by medication started soon after birth.

VDRR is transmitted as an X-linked dominant disorder and is passed on directly from an affected female

to half of her sons and half of her daughters. An affected male will give it to all of his daughters and to none of his sons. Previous investigators have reported that VDRR males demonstrate a more severe form of the disease than do females (Winters et al. 1958). Females may be spared the full impact of the disease due to the inactivation of one of the X-chromosomes in each cell as described in the single-active-X hypothesis (Lyon hypothesis). This hypothesis explains how a female having 2 X chromosomes does not make twice as much product of X-linked genes as the male, who has only one X: there is only one active X chromosome in any given cell (Nora and Fraser 1989). The gene carrying the trait may be inactivated in approximately 50% of the cells, thereby diluting the trait. Males who have received the X-chromosome carrying the trait from their mothers will have the condition expressed. Dental abnormalities have also been reported to be more common in males than females in a group of patients diagnosed with VDRR (Tracy and Campbell 1968). The findings of the present study are in agreement with Winters et al. (1958) and Tracy and Campbell (1968) in that males were more severely affected with both the disease and dental abscesses than females. Five of 10 males (50%) included in the study required dental treatment for abscesses in their primary dentition, while treatment was required for only one of 14 females (7%).

In these six affected patients, a total of 42 primary teeth abscessed. Anterior teeth were more commonly involved than posteriors (24 of 42 — 57%), with five of the six patients (83%) having an anterior tooth abscess first. Examination of the sequence of abscesses in each

Table 3. Average dosage per age patients with vs. patients without dental abscesses

Age	Average Dosage* for Patients Demonstrating Dental Abscesses	N	Average Dosage* for Patients Not Demonstrating Dental Abscesses	N
3	41	5	49	9
4	76	5	71	12
5	115	5	73	12
6	106	4	101	12
7	170	4	115	11
8	248	3	140	11
9	279	3	178	10
10	250	2	174	9

* Dosages are in thousands of units of vitamin D per day.

N = Number of patient dosages totaled to determine average dosage.

patient reveals that the occurrence loosely follows the sequence of eruption. Removal of the enamel by attrition exposes the poorly mineralized dentin to microorganisms that can invade the pulp; therefore, smaller anterior teeth that have less enamel for protection of the pulp will be more readily involved in susceptible individuals. Generally, posterior teeth will become involved later than anterior teeth unless operative treatment has been rendered. In that event, the time between eruption and abscess is reduced and the sequence no longer follows the order of eruption.

In those VDRR patients who have extensions of the pulp to the DEJ as reported by Cohen and Becker (1976) and Özkan et al. (1984), invasive operative procedures, such as cavity or crown preparation, would expose the pulp. An obvious exposure might not occur; however, an undetected microexposure could lead to abscess at a later time.

In this study population, one tooth was treated with a pulpotomy and later removed due to failure of the procedure. This tooth may have had pulpal involvement of a more severe nature than believed at the time of diagnosis and treatment, therefore resulting in eventual failure.

Patients in the study group who did not demonstrate dental abscesses received routine operative treatments including sealants, amalgams, pulpotomies, and stainless steel crowns. These patients did not later develop abscesses of these teeth as a result of the treatment. The defect which predisposes the patient to abscesses apparently is not present in all individuals diagnosed with VDRR; therefore, providing operative dentistry for them will not result in abscess of the treated teeth. Obviously, the restoration of teeth in a predisposed patient hastens the occurrence of abscesses, indicating that it is the defect present in the tooth and not the operative procedure itself that leads to abscesses in these patients. One must conclude that the occurrence of a "spontaneous" abscess or an abscess following a shallow cavity preparation determines the need for aggressive preventive dental treatment.

The most interesting finding in this study population was the fact that none of the children had only one abscess. Numbers of abscesses per individual ranged from a low of two to a high of 11. The results of this study would indicate that one abscess is a predictor of future abscesses. The child who had only two abscesses was the youngest in the study and if followed over a period of years will most probably demonstrate abscesses of other primary teeth. Therefore, in a patient who is susceptible to abscesses, i.e. one who has had an abscess of one or more primary teeth uninvolved with caries or trauma, it is recommended that consideration be given to prophylactic treatment of all posterior primary teeth with pulpotomies and stainless steel crowns.

Currently there are no data to support this recommendation, but if the treatment can be performed early, before actual invasion of the pulp by bacteria, the more conservative choice of pulpotomy would be expected to be successful. However, if attrition has already allowed bacterial ingress and the pulp is inflamed irreversibly, pulpectomy would be the treatment of choice. For VDRR patients, extraction should only be considered if pulp therapy fails.

Two commonly reported dental anomalies associated with VDRR are enlarged pulp chambers (Daeschner 1957; Rakocz et al. 1982; Seow and Latham 1986) and elongated pulp horns extending to the DEJ (Gardner et al. 1969; Özkan et al. 1984; Bender and Naidorf 1985). Findings reported in the literature concerning elongated pulp horns are confirmed by histological study of extracted teeth (Harris and Sullivan 1960; Witkop 1971; Breen 1986). In ground sections, the defect which allows the pulp to extend to the DEJ can be visualized.

Bender and Naidorf (1985) report that pulp horns extending to the DEJ seen radiographically can be considered pathognomonic for VDRR. Examination of radiographs in this study did not consistently reveal extensions of the pulp toward the tooth surface.

However, to explore the possibility that predictions of the severity of dental manifestations, i.e. occurrence of abscesses, could be made by examination of dental radiographs, two measurements of the films were made: the distance from the pulp horn to the DEJ, and the width of the pulp chamber. It was determined that measurement of dental radiographs exposed in the course of treatment was not a reliable method for predicting either which patients would be affected with dental abscesses or determining which teeth in an affected patient would abscess. A complicating factor in examining dental radiographs is introduced by the radiographic technique itself. Distortion of the image caused by the angulation of the X-ray beam can obscure findings that would indicate potential problems for these patients. While the cause of abscess may be determined histologically, it is not apparent radiographically.

Conclusions

1. Twenty-five per cent of the patients diagnosed with VDRR were affected with abscesses of their primary dentition.
2. In patients demonstrating abscesses in the primary dentition, the sequence of involvement generally followed the pattern of tooth eruption with anterior teeth more commonly involved. Operative treatment shortened the time between eruption and the occurrence of the abscess in the predisposed patient.

3. Males were more commonly affected with the abscesses associated with VDRR than females. Fifty per cent of the males in the population were affected while only 7% of females were affected.
4. Teeth in either arch are equally likely to abscess in an affected patient.
5. The abscess of one tooth indicates that at least one other tooth will be affected.
6. Predictions of the occurrence of abscesses in the population cannot be made either from medication dose (serving as an indication of disease severity), or from measurement of radiographs.
7. Further study of a population diagnosed with VDRR is needed to determine if the incidence of dental abscesses in a larger population is similar to the 25% reported here.

Dr. McWhorter is assistant professor, and Dr. Seale is professor and chairman, Department of Pediatric Dentistry, Baylor College of Dentistry, Dallas, TX. Reprint requests should be sent to: Dr. Alton G. McWhorter, Department of Pediatric Dentistry, Baylor College of Dentistry, 3302 Gaston Avenue, Dallas, TX 75246.

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