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What Do We Know About Dentigerous Cysts in Children: A Review of Literature

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ABSTRACT

Dentigerous cysts (DCs) known as follicular cysts are thought to be caused by a developmental abnormality derived from the reduced enamel epithelium (REE) of the tooth forming organ. DCs considered an epithelial lined developmental odontogenic cyst which usually occurs in the second and third decade of life. DCs are the second most prevalent cystic lesions of the jaw following the radicular cyst. They are associated with partially erupted, developing, supernumerary or impacted teeth. Locations wise in the jaw, the mandibular third molars followed by maxillary canines are preferred sites. Occurrence of DCs is commonly unilateral. Bilateral presence is generally observed in syndromic cases. Non-syndromic DCs occurring bilaterally or involving both arches at the same time is very rare.

Keywords: Dentigerous cysts, Unerupted tooth, Enucleation, Odontogenic.

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INTRODUCTION

DCs are very common benign developmental cysts which generally involve impacted, unerupted permanent teeth, supernumerary teeth, odontomas, and, rarely, primary teeth [1]. They usually occur singly and are located in the mandible. DCs comprise the second most common type of odontogenic cysts, representing nearly 24% of all the true jaw cysts [1-4]. The World Health Organization classification of jaw cysts refers to the DCs as an epithelial developmental odontogenic cysts [5]. They are attached to or enclose the crown of the unerupted tooth at the cemento-enamel junction (CEJ) [1]. Though DCs may be seen in patients across a wide age range, they are most frequently discovered in patients between 10 and 30 years of age. Their frequency in the first decade of life is reported to be lower than in the second and third decades [2]. The majority of them are associated with impacted or unerupted mandibular third molars followed by maxillary canines and maxillary third molars [6]. DCs believed to develop around the crown of an unerupted or impacted tooth by accumulation of fluid between the REE and the tooth enamel or within the enamel organ. Pressure exerted by an erupting tooth on an impacted follicle obstructs the venous outflow. This leads to rapid transudation of serum across the capillary walls. Increase in the hydrostatic pressure of the pooling fluid, leads to separation of the follicle from the crown with or without REE [2]. An intra-follicular spread of periapical inflammation from a primary tooth may also result in the development of DCs [2]. DCs are frequently discovered when routine radiographs are taken to investigate a failure of tooth eruption, a missing tooth or mal-alignment. Most of the time, there is no pain or discomfort associated with the cyst unless it becomes secondarily infected. Radiographs show a

unilocular radiolucent lesion characterized by well-defined sclerotic margins and associated with the crown of an unerupted tooth. While normal follicular space is within 3–4 mm, DCs can be suspected when the space is more than 5 mm [1]. Several published case reports associated with literature of reviews did not answer some of the following questions:

- Are DCs uncommon in children?
- What could be the exact etiology of DCs in children?
- Common gender and age affected by DCs?
- Most common locations and teeth associated with DCs in children?
- Most common treatment modality for DCs in children?

The objective of this review of literature is to answer all or some of the above questions and provide some recommendations that could serve as guide line when managing DCs in children.

MATERIALS AND METHODS

Search Strategy

A detailed electronic search strategy was used for each selected data base to identify all of the articles published in relation to the stated objectives of this review. In PubMed, EBSCOhost, Ovid arms of Medline, and Google Scholar, articles were searched from 1980 to September 2016. The following search strategy was used: "Dentigerous cyst" OR "Dentigerous cysts" OR "Dentigerous cyst in children" OR "Dentigerous cysts in children". The search was complemented by checking the reference of the selected articles for additional eligible publications. Each article was reviewed manually based on the title and the abstract for primary exclusion. Then, the full texts of the remaining reports were reviewed and summarized.

Inclusion Criteria

Case reports and series that studied DCs in children were included if they fulfilled the following criteria: 1) age: below 18 years old, 2) English-language publications in the dental literature, 3) humans, and 4) Non-synrodmic cases. In contrast, studies that investigated other outcomes and papers that presented unconfirmed cases of DCs were excluded.

Selection of Studies

After the initial electronic search of titles and abstracts, the titles of all studies were scanned independently. The next step was to review all selected abstracts and apply the inclusion criteria to determine the selection of full-text articles. Full texts of all studies of possible relevance were then obtained for independent review and assessment. Disagreements between reviewers were resolved by discussion. All studies meeting the inclusion criteria underwent data extraction. Studies rejected at any subsequent stages were removed, and reasons for exclusion were recorded.

Data Extraction

The data extracted is presented in a Table.

RESULTS

Study Inclusion

The initial search yielded 6,336 titles. After screening titles and abstracts, 301 studies were selected for full text review. Further full-text reading and screening led to the exclusion of 201 studies (see flowchart figure for the study selection process), which resulted in the inclusion of 100 case reports and series reporting on DCs in children (see Table).

Aetiology:

This is broadly divided into two main categories, the developmental which included in most of the reviewed studies and the inflammatory [30, 33, 35, 36, 63, 86, 98, 101]. On few studies the patients had chief complaint of pain most of these cases had non-inflammatory aetiology [17, 18, 20, 30, 33, 35, 37, 40, 44, 46, 48, 59, 61, 76].

Sex/ Distribution

The 100 studies consist of 96 case reports and four case series [35, 59, 71, 93]. The included published data were from seventeen different countries, mainly from India. The review included 200 children, 124 males and 75 females. In one study the gender was not specified [99].

Age of Onset

The youngest age of a child diagnosed with DCs was 1 year old [7], the most common age diagnosed with DC is 10 years old.

Location: (Teeth, Jaws, Unilateral/Bilateral, Multiple)

Teeth Involvements: DCs are not associated only with permanent teeth, but also with supernumerary teeth mainly mesiodens present with upper central incisors [26, 49-53, 71, 82]. Some reports showed DCs associated with unerupted teeth with and without presentation in the maxillary sinus [27, 28, 42, 91, 102]. Few reports showed presentation with odontomes [94, 99] and primary teeth [39]. The most involved tooth with DCs is the lower left second premolar followed by the lower left first molar. Some of the study did not mention the involved teeth type.

Jaw involvements: Almost the same involvements for the mandible and the maxilla, with slight shift toward the mandibular involvement.

Unilateral Versus Bilateral Presentation: Most of the studies presented a unilateral cystic distribution, only 27 studies were bilateral of non-syndromic involvements [8, 10, 14, 21, 24, 36, 44, 47, 51, 56-59, 61, 66, 72-75, 76, 80, 83, 90, 96, 102-104].

Treatment Modality

In most of the reviewed case reports and series, the treatment modalities were enucleation and marsupialization under general or local anaesthesia. Only one case reported the use of nitrous oxide with general and local anaesthesia [96]. Some of the included clinical studies did not mention the type of anaesthetic technique used [10, 14, 16, 17, 21, 35, 36, 39, 42, 44, 46, 49, 50, 51, 54, 55, 57, 59, 66, 68, 70, 74, 78, 79, 81, 82, 85, 89, 90, 93, 98, 102, 103]. In one study the extracted tooth was re-planted after the marsupialization, however there was no enough long term follow-up [16]. In other studies, both enucleation and marsupialization techniques were used due to the presence of large cystic lesion [57, 60]. Root canal treatment of a primary central incisor and marsupialization were also reported [67]. Permanent teeth extraction was common, however some of the studies had significant amount of teeth extraction associated with enucleation and marsupialization techniques [48, 57, 59].

Table 1: CR Case Report, CS Case Series, M Male, F Female, Mx Maxillary, Md Mandibular, Uni Unilateral, Bi Bilateral, GA General Anaesthesia, LA Local Anaesthesia, Mars Marsupialization, Enuc Enucleation , ys Years.

Authors/Year/Country	Study Design	Sex	Age Years	Location	Treatment Modality	Extraction Permanent teeth	Syndromic	Pain
Suresh et al 2011, India [7]	CR	M	1	Uni, Md, 36	Enuc, GA	Yes	No	No
Devi et al 2015, India [8]	CR	M	17	Bi, Md/Mx, multiple	Enuc, GA	Yes	No	No
Bhushan et al 2014, India [9]	CR	M	14	Uni, Md 48	Enuc, LA	Yes	No	No
Tamgadge et al 2011, India [10]	CR	M	10	Bi, Mx, 14, 15, 23	Enuc,?	?	No	No
Demiriz et al 2015, India [11]	CR	F	5	Uni, Md, 34	Enuc, GA	Yes	No	No
Tilakraj et al 2011, India [12]	CR	M	4	Uni, Mx, 14	Enuc, GA	?	No	No
Jindal et al 2010, India [13]	CR	F	7	Uni, Md, 47	Enuc, GA	Yes	No	No
Arjona-Amo et al 2015, Spain [14]	CR	M	7	Bi, Md, 36/46	Mars,?	No	No	No
Pramod et al 2011, India [15]	CR	M	7	Uni, Mx, 23	Enuc, GA	Yes	No	No
Guven et al 2014, Turkey [16]	CR4	4M	7 - 9	Uni, Md, 45 - Uni, Mx, 15 Uni, Md, 33 - Uni, Md, 43 - 44	Mars, ?	No	No	No
Mishra et al 2014, India [17]	CR	F	12	Uni, Md, 35	Enuc, ?	Yes	No	Yes
Gay-Escoda et al 2015, Spain [18]	CR	M	18	Uni, Mx, 13	Enuc, LA	Yes	No	Yes
Kumar et al 2012, India [19]	CR	F	10	Uni, Md, 33, 34, 35	Mars, LA	No	No	No
Deboni et al 2011, Brazil [20]	CR	M	8	Uni, Md, 32, 33, 34, 35	Mars, LA,	Yes, 34	No	Yes
Shirazian et al 2011, Iran [21]	CR	M	10	Bi, Md, 45, 35	Mars, ?	No	No	No
Ramakrishna et al 2013, India [22]	CR	M	10	Uni, Mx, 23	Enuc, GA	Yes	No	No
Mohapatra et al 2009, Qatar [23]	CR	F	11	Uni, Md, 35	Mars, LA	No	No	No
Prasad et al 2010, India [24]	CR	F	12	Bi, Max, 13/23 - Uni, Md, 42, 43, 44, 45	Enuc, GA	Yes	No – rare case	No
Rohilla et al 2009, India [25]	CR	M	9	Uni, Mx, 22	Enuc, LA	Yes	No	No
Kalaskar et al, 2011, India [26]	CR2	M	12	Uni, Mx, 11	Enuc, LA	No	No	No
		M	12	Uni, Mx, 11				
Isser et al 2002, India [27]	CR	M	10	Uni, Mx, 23	Enuc, GA	Yes	No	No
Mamatha et al 2014, India [28]	CR	M	17	Uni, Mx, 23	Enuc, GA	Yes	No	No
Contar et al, 2011, Brazil [29]	CR	F	13	Uni, Md, 47	Mars, LA	No	No	No
Shetty et al 2010, India [30]	CR	M	11	Uni, Md, 35	Enuc, LA	Yes	No	Yes
Carvalho et al 2016 Brazil [31]	CR	M	10	Uni, Md, 35	Mars, LA	No	No	No
Bozdogan et al 2010, Turkey [32]	CR	F	6	Uni, Md, 34/35	Mars, LA	No	No	No
Santos et al 2014, Brazil [33]	CR	M	9	Uni, Mx, 21	Mars, LA	No	No	Yes
Singh et al 2014, India [34]	CR	M	13	Uni, Md, 35	Mars, LA	No	No	No
Benn et al 1996, South Africa [35]	CS15	11M/4 F	5-12	Uni, Mx, Md	Mars, ?	?	No	Yes
Narang et al 2012, India [36]	CR4	F/M	17/16	Uni, Md 35 - Md 43	Mars, ?	?	No	?

		M/M	16/9	Mx 13 - Mx 23				
Shekhar et al 2006, India [37]	CR	F	18	Uni, Max, ?	Enuc, GA	Yes	No	Yes
Hayasaki et al 2008, Japan [38]	CR	F	5	Uni, Md, 45	Mars, GA	No	No	No
Motokawa et al 1990, Japan [39]	CR	F	3	Uni, Mx	Mars, ?	No	No	No
Kara et al 2015, Turkey [40]	CR	F	16	Uni, Mx, 18	Enuc, GA	Yes	No	Yes
Silva et al 2007, Brazil [41]	CR	M	10	Uni, Md, 33, 34, 35	Mars, LA	No	No	No
Haber 2008, Canada [42]	CR	F	4	Uni, Mx	Enuc, ?	Yes	No	No
Kalaskar et al 2007, India [43]	CR	M	7	Uni, Mx, 11	Enuc, LA	Yes, 13	No	No
Hansford et al 2015, USA [44]	CR	F	6	Bi, multiple, Mx, Md, canines/PMs	Mars, ?	No	No	Yes
Kirtaniya et al 2010, India [45]	CR2	M	7	Uni, Md, 34	Mars, LA	No	No	No
		F	10	Uni, Md, 43	Mars+Enuc	No	No	No
Hegde et al 2013, India [46]	CR	M	9	Uni, Md, 35	Enuc, ?	Yes	No	Yes
Shivaprakash et al 2009, India [47]	CR2	F	10	Bi, Md, 35/45	Enuc, LA	No	No	No
		M	10	Uni, Md, 35	Mars, LA	No	No	No
Shashikiran et al 2006, India [48]	CR	M	12	Uni, Mx, 12, 14, 15	Enuc, GA	Yes all teeth	No	Yes
Ikarashi et al 2003, Japan [49]	CR	M	7	Uni, Md, 46/47	Mars, ?	No	No	No
Lustmann et al 1988, India [50]	CR2	F	9	Uni, Mx, 11	Enuc, ?	?	No	?
		M	12	Uni, Mx, 21/22	Mars, ?	?		?
Dinkar et al 2007, India [51]	CR	F	14	Bi, Mx	Enuc, ?	No	No	No
Gulses et al 2009, Turkey [52]	CR	M	10	Uni, Mx, 11	Enuc, LA	No	No	No
Kumar et al 2010, India [53]	CR	M	14	Uni, Mx, 11	Enuc, GA	No	No	No
Shetty et al 2012, Libya [54]	CR	M	10	Uni, Mx, 23, 24	Enuc, ?	Yes	No	No
Das et al 1994, India [55]	CR	F	14	Uni, Mx,23	Enuc, ?	Yes	No	No
Freitas et al 2006, Brazil [56]	CR	M	14	Bi, Mx 18, Md, 36/37	Enuc, GA	Yes	No	No
De Morais et al 2014, Brazil [57]	CR	M	15	Bi, Md, 37/47	47=Mars+Enuc 37= Enuc,?	Yes for 8's	No	No
Cury et al 2009, Brazil [58]	CR	M	5	Bi, Md, 46/36	Enuc, GA	No	No	No
Motamedi et al 2005, Iran [59]	CS26	F/F	12/11	Bi, Md, 42-34/ Uni,	Enuc/Mars, ?	No	No	Yes
		M/M	10/18	Md, 48-45	Enuc/Enuc, ?	No/Yes 38		
		F/F	8/11	Uni, Mx, 15-12/ Uni,	Enuc/Enuc, ?	Yes 13/No		
		M/M	18/5	Md, 36-38	Enuc/Enuc, ?	Yes 23/23		
		M/F	8/8	Uni, Mx, 16-12/ Uni,	Enuc/Enuc, ?	Yes 32,33,34/ 34		
		F/F	12/12	Mx, 16-11	Enuc/Enuc, ?	Yes 47/44,45		
		F/M	18/12	Uni, Mx, 22-25/ Uni,	Enuc/Enuc, ?	Yes 34,35		
		F/M	9/18	Mx, 23-25	Enuc/Enuc, ?	Yes 33,34/48		
		F/M	16/18	Uni, Md, 32-34/ Uni,	Enuc/Enuc, ?	No/Yes 13		
		M/M	18/11	Md, 44	Enuc/Enuc, ?	Yes 43/ 33		
		F/M	17/12	Uni, Md, 48-46/ Uni,	Enuc/Enuc, ?	Yes 23/ 23-25		
		M/F	14/16	Md, 48-43	Enuc/Enuc, ?	Yes 38/Yes 33		
		M/M	15/18	Uni, Md, 33-38/ Uni, Mx, 15-13 Uni, Md, 33-38/ Uni, Md, 48-46 Uni, Mx, 14-12/ Uni, Mx, 12-14 Uni, Md, 44-42/ Uni, Md, 32-34 Uni, Mx, 22-24/ Uni, Mx, 23-25 Uni, Md, 36-38/ Bi, Md, 43-33 Uni, Md, 36-38/ Uni, Md 36-38	Enuc/Enuc, ?	Yes 36, 37/37, 38		
Scariot et al 2011, Brazil [60]	CR	M	11	Uni, Mx, 11, 12, 13	Mars, LA + Enuc, GA	Yes	No	No
Reddy et al 2011, India [61]	CR	F	11	Bi, Md, 44/45 & 34/35	Enuc, GA	Yes, 44/34	No	Yes
Bharath et al 2011, India [62]	CR	M	9	Uni, Mx, 21, 22, 23	Enuc, GA	Yes	No	No
Kozelj et al 1999, Slovenia [63]	CR4	M	12	Uni, Md, 34	Enuc, LA	Yes	No	No
		F	10	Uni, Md, 35	Mars, LA Mars,	No		
		M	9	Uni, Md, 33/34	LA	No		
		F	8	Uni, Md, 32 - 36	Mars, GA	No		
Passi et al 2008, India [64]	CR	M	10	Uni, Md, 43, 44, 45	Mars, LA	No	No	No
Desai et al 2005, India [65]	CR	M	8	Uni, Mx, 21	Enuc, GA	Yes	No	No
Subash et al 2014, India [66]	CR	M	14	Bi, Mx, Md, 17, 27, 37, 47	Enuc, ?	Yes	No	No
Gondim et al 2008, Brazil [67]	CR	M	4	Uni, Mx, 11	RCT A + Mars,?	No	No	No
Carlos et al 2006, Brazil [68]	CR2	M	10	Uni, Md, 35, 36	Mars, ?	No	No	No
		M	8	Uni, Md, 42	Enuc, ?			
Asián-González et al 2007, Spain, [69]	CR	F	9	Uni, Md, 35	Enuc, LA	Yes	No	No
Kawamura et al 2004, Brazil [70]	CR	F	7	Uni, Md, 43, 44, 45	Mars, ?	No	No	No
Berden et al 2010, Sweden [71]	CS12	F/M	5/7	11/36	Mars, GA	Yes/No	No	No
		M/M	8/8	46/36		No/No		
		M/F	9/10	47/47		No/No		
		M/M	10/10	34/22		No/No		
		F/M	10/10	15/42		No/No		
		M/F	10/11	47/23		Yes 48/Yes 23		
Sumita et al 2006, India [72]	CR	M	13	Bi, Md, 35, 43, 44, 45	Enuc, GA	Yes, 35/44	No	No
Fregnani et al 2008, Brazil [73]	CR	M	5	Bi, Md, 36/46	Enuc, GA	No	No	No
Maurette et al 2008, Venezuela [74]	CR	M	7	Bi, Md, 36/46	Mars,?	No	No	No
Norris et al 1987, USA [75]	CR	F	7	Bi, Mx/Md, multiple	Mars, GA	No	No	No
O'Neil et al 1989, USA [76]	CR	M	5	Bi, Md, 46/36	Enuc, GA	No	No	Yes
Murakami et al 1995, Japan [77]	CR	M	12	Uni, Md, 35	Mars, LA	No	No	No
Berti et al 2010, Brazil [78]	CR	M	9	Uni, Md, 33/34	Mars,?	No	No	No

Martinez-Pérez et al 2001, Spain [79]	CR4	F/M F/M	9/11 8/10	Uni, Md, 45 - Uni, Md, 34/35 Uni, Md, 32/33 - Uni, Md, 34	Mars, LA/Mars,? Mars,?/Mars,?	No	No	No
Swerdloff et al 1980, USA [80]	CR	F	7	Bi, Md, 46/36	Enuc, LA	No	No	No
Aguilo et al 1998, Spain [81]	CR	F	4	Uni, Md, 45	Mars,?	No	No	No
Lustmann et al 1988, Israel [82]	CR2	F M	9 12	Uni, Mx, 11 Uni, Mx, 21/22	Enuc, LA Mars,?	No	No	No
Goyal et al 2009, India [83]	CR	F	7	Bi, Mx, 16/15/14/13/26	Enuc, GA	Yes	No	No
Jones et al 2003, UK [84]	CR	M	6	Uni, Md, 36	Mars, GA	No	No	No
Bhatia et al 2009, UK [85]	CR	F	12	Uni, Md, inverted 45	Enuc?	Yes	No	No
Yao et al 2015, China [86]	CR	M	6	Uni, Md, 45	Enuc, LA	Yes	No	No
Shah et al 1994, India [87]	CR	F	13	Uni, Mx, ?	Enuc, GA	Yes?	No	No
Counts et al 2001, USA [88]	CR	F	7	Uni, Md, 35/36/37/38	Enuc, GA	Yes	No	Yes
Das et al 1999, India [89]	CR	F	14	Uni, Mx, 23	Enuc, ?	Yes	No	No
Ziccardi et al 1997, USA [2]	CR	M	8	Uni, Md, 43/44/45	Mars, GA	No	No	No
Batra et al 2004, India [90]	CR	F	15	Bi, Md, 35/38/48	Enuc,?	Yes + 37/47	No	No
Takagi et al 1998, Japan [91]	CR	F	6	Uni, Md, 25	Mars, GA	No	No	No
Ertas et al 2003, Turkey [92]	CR	F	9	Uni, Md, 35/34/33/32	Mars, GA	No	No	No
Huseyin et al 2009, Turkey [93]	CS33	21M/1 2F	8-13	?	Mars, ?	?	?	?
Bansal et al 2014, India [94]	CR	F	13	Uni, Mx, 11	Enuc, LA	No	No	No
Yucel et al 2013, Turkey [95]	CR	F	9	Uni, Md, 43/44/45	Mars, LA	No	No	No
Ashkenazi et al 2014, USA [96]	CR	F	6	Bi, Md, 42/41/31/32	Enuc GA/LA	No	No	No
Jena 2015, India [97]	CR	F	12	Uni, Mx, 13/12/11	Mars, LA	Yes, 12	No	No
Thambi et al 2016, India [98]	CR	M	8	Uni, Mx, 26/25/24	Enuc, ?	Yes, 24	No	No
Biocic et al 2010, Croatia [99]	CR	?	10	Uni, Md	Enuc, GA	No	No	Yes
Reyes et al 2016, Cuba [100]	CR	M	9	Uni, Mx, 13	Enuc, GA	Yes	No	No
Jain et al 2016, India [101]	CR	M	6	Uni, Mx, 24	Mars, LA	No	No	No
Ustuner et al 2003, Turkey [102]	CR	M	6	Bi, Mx, 23/13	Enuc, ?	Yes	No	No
Crinzi et al 1982, USA [103]	CR	M	15	Bi, Md, 48/38	Enuc, ?	Yes	No	No
McDonnell et al 1980, Ireland [104]	CR	M	15	Bi, Md, 45/37	Enuc, GA	Yes 37	No	No
Gupta et al 2015, India [105]	CR	M	9	Uni, Md, 43/44/45	Enuc, GA	Yes	No	No

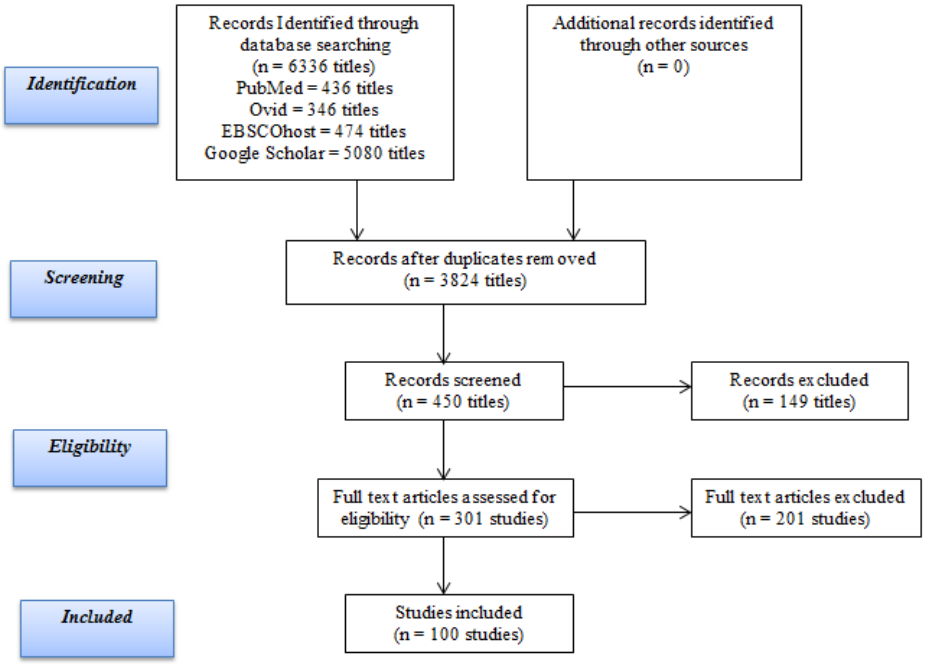

Figure: Flowchart of study selection process

DISCUSSION

DCs are benign odontogenic lesions of unknown direct causative factor. Different theories have been proposed to explain the etiology of DCs. The first theory suggests that DCs are developmental in origin and occur in mature teeth usually as a result of impaction. These cysts usually occur in the late second and third decades, predominantly involving mandibular third molars. By an alteration of REE which results in fluid accumulation either between the REE and the

crown of an un-erupted/impacted tooth or within the enamel organ itself [106].

The second theory advocates that DCs are inflammatory and occur in immature teeth as a result of inflammation from non-vital primary tooth or another source spreading to involve the tooth follicle. These are diagnosed in the first and early part of second decade, predominantly involving mandibular premolars [107]. Inflammatory DCs are found in mixed dentition, they develop when the inflammation present at the root apex of a non-vital primary tooth spreads to involve the follicle of the un-erupted immature permanent successor. In procedure like pulpotomy, possible complications like malposition, delay in eruption, enamel defects, discoloration, cyst formation, and damage to the permanent dentition have been reported [108-110]. DCs can form in the peri-radicular region after pulpotomy [111, 112] caused by an alteration of the REE (after completion of amelogenesis), which results in fluid accumulation between the epithelium and the tooth crown [113]. The persistent and prolonged inflammation may cause chronic irritation to the dental sac of the un-erupted tooth, which in turn leads to the development of DCs. On the other hand, DCs can occur in conjunction with necrotic or pulp treated teeth in the primary dentition. The time lapse between the pulp therapy and the detection of buccal bone expansion ranged from 5 months to 3 years. Taking into account that ameloblastic transformation can occur, practitioners should investigate cases of molars, particularly if they are pulpotomized [114].

DCs are the second most common cystic lesion to affect the mandible. They account for 14- 20% of mandibular cysts and between 15.2% and 33.7% of all odontogenic cysts [107]. The frequency with which DCs develop has been calculated at 1.44 in every 100 unerupted teeth [107]. They are more frequent in men than in women and are more frequent in caucasians than in individuals with darker skin [107]. The proportion of 6 to 7 year-old children affected with DCs is only 9.1% [115]. In this review, 200 DCs in children were included; it affected 124 males and 75 females.

A review of related literature shows that though the age range for the reported cases of DCs varies widely from 5 to 57 years, they most commonly manifest in the second and third decades of life [1]. Their frequency in the first decade is reportedly lower than in the second and third decades [116]. Tamgadge have documented a case of DCs in a 10 year old boy, large number of the included reports in this paper is around this age [10]. In one case report the age of the child at the time of manifestation of the cyst was 1 year, which makes this the youngest case of DCs to be documented [7]. DCs are rare in the first decade of life, and when they occur, they manifest with minimal symptoms but they can attain considerable size over a period of time.

DCs are one of the most common cysts of the jaws. A vast majority of the DCs are associated with impacted mandibular third molars, followed by the permanent maxillary canines and the maxillary third molars [107, 111]. A few cases have also been reported to occur in association with permanent maxillary central incisors, permanent mandibular first and second molars [43, 104]. DCs have also been reported to occur in association with primary teeth [39]. In this review the substantial majority of DCs involve the mandibular second premolar and the mandibular second molar, followed by the maxillary permanent canines and rarely the mandibular incisors. Bilateral DCs generally occur in association with a developmental syndrome or systemic diseases. Bilateral DCs in absence of a syndrome is rare and to date only few cases have been reported. Although this finding may reflect the true rarity of the condition, it is conceivable that bilateral DCs are either under recognized or under-reported as sometimes they are known to regress spontaneously. The earliest case of multiple DCs was recorded by Glaswald in 1844 [117]. Bilateral and multiple cysts have been reported in Basal cell nevus syndrome, Mucopolysaccharidosis, Cleidocranial dysplasia and after prolonged concurrent use of cyclosporine A and calcium channel blockers [102]. In the absence of these syndromes, bilateral DCs are rare.

DCs are usually asymptomatic, and they may be detected incidentally via routine radiographic examination or when they are large enough (>2 cm in diameter) to cause facial asymmetry [68]. Clinical signs suggestive of DCs include a retained primary tooth, delayed eruption of a permanent tooth, and painless swelling of the involved area. In radiographs, DCs appears as a well-defined unilocular radiolucency associated with the crown

of an unerupted tooth. Often the radiolucent area surrounds the crown, but sometimes it lies mainly or entirely to one side [71]. It is important to emphasize that DCs in children might show a rapid and painless expansion and may result in fractures and deformation of facial bone structures. Early diagnosis and treatment are therefore important. Failure to identify and treat such cysts may allow their expansion to produce impingement on surrounding structures like inferior alveolar nerve and roots of adjacent teeth, resulting in paresthesia and root resorption. DCs may cause displacement of adjacent teeth and resorption of teeth roots. It may displace or obliterate the maxillary sinus, nasal cavity and orbital cavity leading to diplopia [40]. A rare consequence of traumatic injury to a primary tooth is development of DCs in the corresponding permanent tooth [62].

Conservative approach with non-vital primary teeth includes the removal of the cystic lesion and the extraction of the un-erupted tooth as the main treatment to prevent the recurrence of the cystic lesion [63]. Treatment of DCs depends on its size and location. The options available for their treatment in children include enucleation of the cyst with primary closure and marsupialization and healing by secondary union [3]. Complete removal of the cyst is extremely important given that, recurrent cysts, ameloblastoma, squamous cell carcinoma and mucoepidermoid carcinoma have been reported to result from long-standing DCs or its remnants [4]. It highlights the importance of enucleation as the first choice of treatment for large cystic lesions, instead of conservative procedures like decompression and marsupialization which are used for children. Though marsupialization might help in the preservation of vital structures, keeping in view, the potential of more aggressive transformation of the cystic lining, complete removal of the lining by enucleation, with emphasis on possible preservation of vital structures combined with a thorough follow-up, is more appropriate [9].

Marsupialization of the cyst is the treatment of choice when it gives a chance to the un-erupted tooth to erupt in large cysts; however, this technique creates notable disadvantages. To illustrate, the two-stage surgical procedure may result an intolerable procedure for a child and lifting behind a pathological tissue and in a large cystic cavity, it takes long period of time for the bone to regenerate [7]. However, the recurrence of DCs is rare, especially after complete removal of the cyst or tooth eruption [16]. Marsupialization, a comparatively simple procedure, consists of surgically producing a window in the cystic wall to relieve intra-cystic tension. The possibility of bone fracture and the safe change in the patient's mandibular growth should also be assessed. For this reason, more conservative and less traditional options should be explored. The success of these techniques is put down to maintaining eruptive potential of permanent teeth that are still yet to conclude their development.

Two research papers have found that between 71.4 and 72.4% of the individuals who participated in their study presented with natural eruption of teeth enclosed in the cyst after having carried out marsupialization [118, 119]. In a case study about DCs belonging to a boy of twelve years old, which was located on the same level as the lower left second premolar and which was treated through marsupialization [77]. In another case study about DCs with repeated episodes of infection related to the mandibular canine, a primary molar, a mandibular canine and the first permanent premolar, which triggered the impaction of these teeth. In this case, after administering antibiotics and carrying out extractions of the canine and the primary molar, marsupialization was conducted to try and save the implicated permanent teeth and after 12 months, a complete reduction of the lesion and the eruption of the canine and first premolar was noted [78].

The key factors in the eruption of the tooth in the arch are bone development, the angle and the depth of the tooth in the jaw. The impacted teeth together with incomplete root development show potential for eruption, which takes place when they have two thirds of the root formation emerging in the oral cavity when they have approximately three quarters of root formation. It has also been proven that those teeth that present an axial angle less than 80º and a depth in the maxillary less than 9 mm have significantly higher chances of erupting. The size of the cyst and the existing space between adjacent teeth appear to have no influence on the eruption of these teeth [119]. In accordance with the above studies, the probability of eruption of a permanent tooth enclosed in DCs where their tooth has a depth less than 5 mm and where the angle of the tooth is less

than 25º and the space between adjacent teeth was greater than the size of the teeth [120]. It's very well established that the average time it takes teeth to erupt without carrying out orthodontic traction is approximately three months in comparison to a period of some hundred days after the more conservative approach has been used to decide whether to extract or carry out orthodontic traction. In this case study, the teeth erupted within the three months period [118, 119].

Conservative treatment is a favorable treatment modality for DCs in growing children and adolescents. Considering that the regeneration capacity of the bony structures in children is greater than in adults and the teeth with open apices have greater eruptive potential, efforts should be made to allow the involved tooth to erupt spontaneously. Intentional replantation could also be employed in teeth which have a great strategic value for maintaining the balanced occlusion. It is important to follow up with patients regularly for the possibility of recurrence of DCs. Root canal treatment of the involved teeth is another option to avoid early tooth loss. DCs associated with anterior teeth will result in failure of eruption and leads to esthetic and orthodontic problems. Absence of a central incisor can have an impact of social stigma affecting the psychology of child.

CONCLUSION

To prevent the development of DCs and to avoid unwanted effects on adjacent teeth, early detection consisting of a thorough clinical and radiographical examination is necessary for accurate diagnosis and proper treatment planning. As per the guidelines of the American Academy of Pediatric Dentistry, the first panoramic radiographic examination should be performed following the eruption of the first permanent tooth. Radiographic examination of all unerupted teeth must be done to detect any associated pathology at an early stage. Odontogenic keratocyst, unicystic ameloblastoma, central giant cell granuloma, and a large radicular cyst must be considered in the differential diagnosis of DCs. Radiographic examination alone cannot differentiate the above-mentioned lesions, so a histopathological examination should be performed. Additionally, the epithelial cells lining the lumen of DCs possess an unusual ability to undergo metaplastic transition. Untreated DCs rarely develops into an odontogenic tumor or a malignancy like squamous cell carcinoma. Therefore, early diagnosis and treatment of DCs lesion creates an importance for the prevention of the occurrence of more destructive lesions.

Conflict of Interest

The authors declare that there is no conflict of interest regarding the publication of this article.

Author's Contributions

Amani Al Tuwirqi: Wrote the initial version of the manuscript and contributed significantly to the discussion, drafting and revision of the manuscript.

Nabil Khzam: Conducted a literature review of similar articles, case reports and contributed significantly to the discussion.

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