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Non-Syndromic Bilateral Dentigerous Cyst in 9-Year-Old Boy: A Case Report and Literature Review

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Case Report

History

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ABSTRACT

Dentigerous cyst is a developmental odontogenic cyst that often develops in the second and third decades of life. This is one of the most common odontogenic cyst types that involve developing, or impacted teeth, partially erupted, accounting 20% of all jaw cysts. Dentigerous cysts are usually unilateral, bilateral presentations have been associated with syndromes such as Cleidocranial Dysplasia and Maroteaux–Lamy syndrome. However, they can occur without any syndrome association, but are extremely rare. This article reports the case of bilateral dentigerous cysts that is non-syndromic in a 9-year-old boy.

Keywords: Bilateral Dentigerous cyst, Developmental cyst, Decompression, non-syndromic, Unerupted teeth.

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Introduction

Dentigerous cyst (DC), which Paget first identified in 1863, are slow-growing odontogenic tumors of developmental origin that are most frequently found on the crown of an impacted or unerupted third molar.[1] They are the most common type of jaw developmental cyst, accounting for approximately 20% of all jaw cysts.[2] It is derived from the reduced enamel epithelium and has been described as an epithelium lined cavity. It is usually asymptomatic although it can increase in volume causing pain and compression of nerve structures.[3] These cysts are usually unilateral, developing around the crowns of impacted mandibular third molars.[2] Radiographically, a unilocular radiolucency is associated with the retained or embedded tooth. Syndromes like Cleidocranial Dysplasia and Maroteaux-Lamy have been linked to bilateral dentigerous cysts. It is unusual to find bilateral dentigerous cysts that are not syndromic, especially in the mixed dentition.[2] So far, less than twenty such cases have been reported.[1] According to the study conducted by Nuraini H, DC occurred more commonly in permanent teeth than primary. He also stated that among the primary dentition, DC is more often due to the impaction of mandibular molar followed by maxillary canines, maxillary second molar, and mandibular canines.[4] We report a rare case of bilateral non-syndromic dentigerous cysts in a 9-year-old boy and Literature review to emphasize the importance of early diagnosis and management of this condition to prevent potential complications.

Case Report

A 9-year-old male patient presented to the institution complaining of swelling in the right and left lower back tooth region for one month. He was apparently well two months back, later a swelling developed that gradually attained the present size. He also complained of occasional pus discharge, associated with pain which was intermittent, dull, non-radiating, aggravating on eating hard foods relieved on taking medication. There was no history of paraesthesia, fever and salty discharge. Patient visited private dental clinic 10 days ago for the same complaint where he was given medication and referred to our hospital. On extra oral examination there was no obvious swelling bilaterally but intra orally there are bilateral ovoid swellings in the vestibular region. On palpation, a swelling of

2x3 cm is noted extending 1cm away from symphysis region, 4cm away from ear lobule lying in 74 region mesial to 36 on the left side and a 2x2 cm swelling extending from 84 tooth region to mesial aspect of 46 on right side. (Figure1) Based on these features he was provisionally diagnosed as Radicular cyst in relation to 74,75,85 and Chronic apical periodontitis irt 84.

For radiographic evaluation, a CBCT was advised. Axial section of mandibular arch showed multiple well defined hypodense areas surrounded by hyperdense tooth like structures irt 85, 46, & 74,75 regions respectively with thin expanded corticated borders. (Figure 2) Panoramic view showed multiple well defined hypo dense areas were seen and displacement of tooth were also noticed.(Figure 3) Based on these details, it was given as dentigerous cyst. Marsupialization of tissue was done irt to 75, 84,85 along with extraction of 75 and submitted for histopathological examination.

On microscopic examination, tissue bits from the right side showed, focal areas of odontogenic cell clusters resembling cell rests with inflammatory areas whereas those from left side showed odontogenic islands with acanthomatous change and odontogenic cell clusters resembling cell rests with inflammatory areas. (Figure 4) Correlating clinically and radiographically, it was diagnosed as bilateral inflamed dentigerous cyst. Five days after marsupialization the patient was given lingual arch space maintainer.

In the follow-up visit of one week, Clinical examination showed tissue overgrowth in 84 region that was surgically removed as it interrupts the eruption. Histopathological examination revealed granulation tissue with odontogenic epithelial lining.

In further follow up visit of one week, OPG showed a bilateral well-defined radiolucency involving the premolar regions of 34, 35, 45 with intact inferior border of mandible. (Figure 5) The tissue bits obtained from right and left premolar region. A decompression tubes were placed on either side attached to a denture. The histopathological examination of specimen showed features consistent with that of first biopsy and was diagnosed as bilateral dentigerous cyst. The patient is under the follow-up.

Figure 1: Clinical image

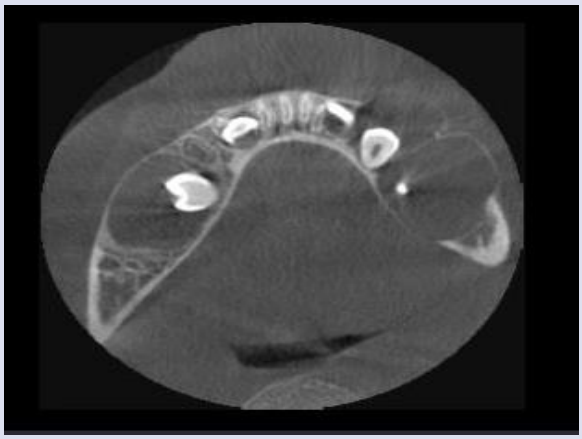

Figure 2: Axial section of CBCT showing multiple well defined hypodense areas.

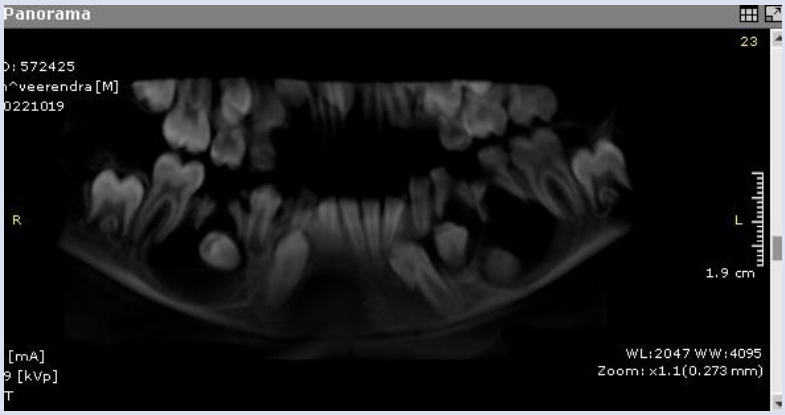

Figure 3: Panoramic image showing multiple well defined hypo dense areas

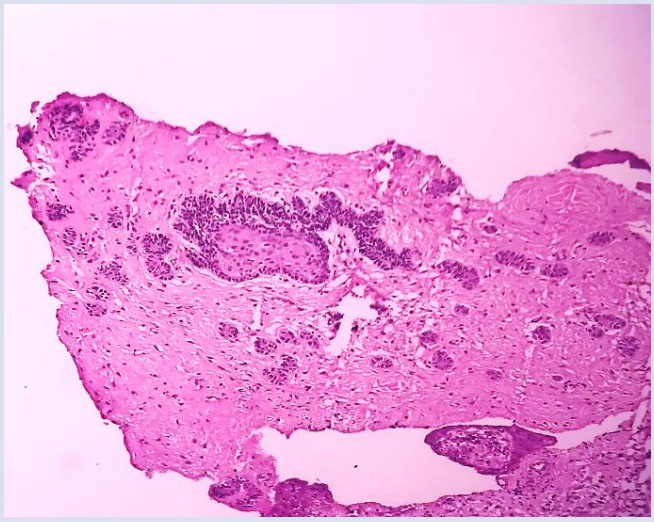

Figure 4: Histopathology showing odontogenic epithelial islands and cell clusters.

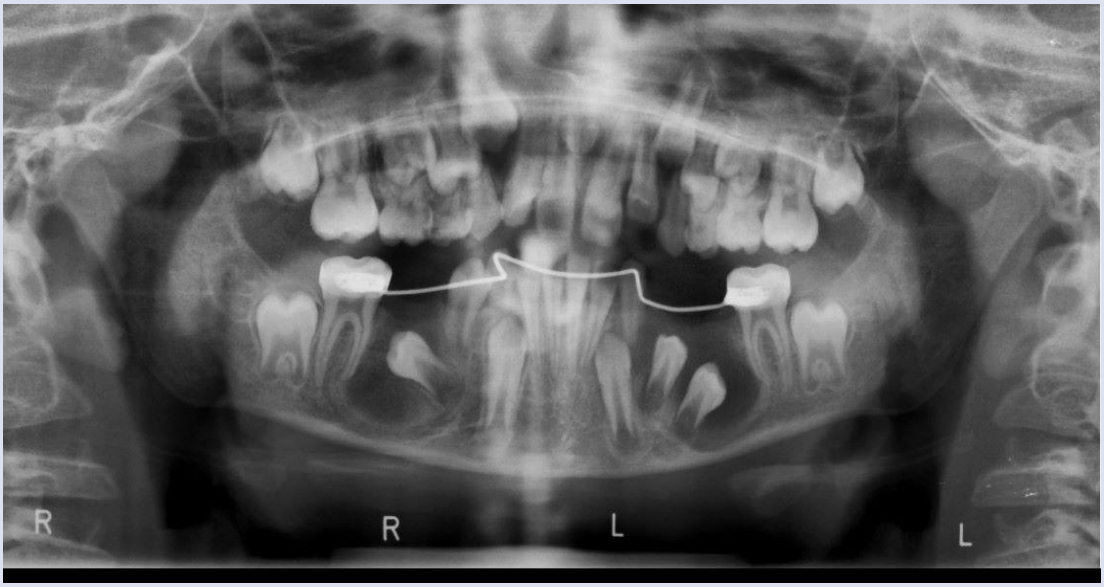

Figure 5: OPG showing bilateral well-defined radiolucencies in premolar areas

Review of cases reported in literature: [5-68]

Table 1: Review of non- syndromic bilateral DC cases reported so far

S No	Authors	Age	Sex	Location	Treatment
1.	Ivy et al (1939)	14	F	15,18,28,37,38,48	Enucleation
		18	F	48, 47, 35,38	
		12	F	43 48, 32, 33, 37	
		11	M	13,23,34	
2.	Myers (1943)	19	F	38,48	Enucleation
3.	Catania et al.(1952)	19	F	18,28,32,33,38,42,43,48	Enucleation
4.	Henefer (1964)	52	F	18,28	Enucleation
5.	Stanback (1970)	9	M	36,46	Enucleation
6.	Callaghan (1973)	38	M	38,48	Enucleation
7.	Burton (1980)	57	F	38,48	Enucleation
8.	Swerdloff (1980)	7	F	38,48	Enucleation
9.	Crinzi RA (1982)	15	F	38,48	Enucleation
10.	Norris et al (1987)	14	F	13,14,15, 17,23,24,25,27	Marsupialisation
11.	McDonnell DG (1988)	15	M	35,37,45,47	Enucleation
12.	Eidinger GB (1989)	15	M	36,46	Enucleation
13.	O Neil DW et al (1989)	5	M	36,46	Enucleation
14.	Banderas JA et al (1996)	38	M	38,48	Enucleation
15.	Sands and Tocchio (1998)	3	F	31,36,41,46	Enucleation
16.	Ko kS et al (1999)	42	M	38,48	Enucleation
17.	De Biase et al & others (2001)	8	M	36,46	Enucleation
18.	Shah N et al (2002)	39	M	38,48	No treatment
19.	Ustuner et al (2003)	6	M	13,23	Enucleation
20.	Batra et al (2004)	15	F	35,38,45,48	Enucleation
21.	Freitas et al (2006)	14	M	18,28, 35,45	Enucleation
22.	Lung and Ganatra (2006)	77	M	38,48	Enucleation
23.	Sumita M et al (2006)	13	M	35,45	Enucleation
24.	Mahajan et al (2006)	13	M	35, 44	Enucleation
25.	Yamalik et al (2007)	51	M	NA	Enucleation
26.	Farahani &Lotfalian (2007)	37	M	13,23,33,43,32	Enucleation
27.	Fregnani et al (2008)	5	M	36,46	Enucleation
28.	Carter et al (2008)	81	M	38,48	Enucleation
29.	Chew YS et al (2008)	30	F	38,48	No treatment
30.	Cury et al (2009)	5	M	38,48	Enucleation
31.	Grewal HK et al (2010)	11	M	13,43	Enucleation
32.	Saluja et al (2010)	22	M	14,15,24,25,34,35,44,45 11,12,21,22,33,43	Enucleation
33.	Tikekar S et al (2010)	11	M	34,35,44,45	Marsupialization
34.	Gaur G et al (2010)	7	F	16,26	Enucleation
35.	Kannan et al (2010)	32	M	13,23	Enucleation
36.	Prasad LK (2010)	12	F	13,23	Enucleation
37.	Shiva S et al (2011)	10	M	35,45	Marsupialization
38.	Reddy et al (2011)	11	F	34,35,44,45	Enucleation
39.	Prabhakar v et al (2011)	10	F	13,23, 11,21	Enucleation
40.	Tamgadge et al (2011)	10	M	14,15,24,25,13,23	Enucleation
41.	Kanth et al (2011)	9	F	34,35,44,45	Enucleation
42.	Ozkan et al (2011)	NA	M	38,48	Enucleation
43.	Gonzalez SM (2011)	10	F	33,43	NA
44.	Shirazian and Agha-Hosseini (2011)	10	M	34,35,44,45	Marsupialization
45.	Akay et al (2011)	7	F	35,45.	Marsupialization
		8	F	35,45	
		7	M	35,45	
		8	M	15,25,35,45.	
		9	F	13,23	
		8	F	13,23, 14,15,24,25	
		9	M	35,45	
46.	Ishihara et al (2012)	NA	NA	14,15,24,25,34,35,44,45	Enucleation and auto-transplantation
47.	Aher V et al. (2013)	24	M	18,28,38,48	Enucleation
48.	Jia et al (2013)	61	F	38,48	Enucleation
49.	Deshpande et al (2013)	9	M	34,35,44,45	Marsupialization
50.	Byatnal AA, (2013)	13	M	NA	NA

	NA	NA	38,48	
51. Lin et al (2013)	NA	NA	13,23	Enucleation
	NA	NA	15,23	
52. Imada et al (2014)	42	F	NA	Marsupialization & Enucleation
53. Cura et al (2014)	45	M	44, 43,33 14,15,24,25	Enucleation
	34	M	33,43	Enucleation
54. Kaushik A, et al. (2014)	17	F	13,23	Marsupialisation
55. Vassiliou et al (2014)	38	F	36,37,46,47	Enucleation
56. Morais et al (2014)	15	M	37,47	Marsupialisation (rt) and enucleation of (lt) cyst
57. Naik et al (2014)	25	M	38,48	Enucleation
58. Rocha et al (2014)	43	F	13,23	Enucleation
59. Shetty et al (2014)	12	M	33,43	Enucleation
60. Swain et al (2014)	9	F	13,23	Enucleation
61. Devi P et al (2015)	17	M	33,43,34,35,44,45	Enucleation
62. Hansford JT (2015)	6	F	NA	NA
63. Sanjay CJ (2015)	24	F	33,43	NA
64. Singh et al (2015)	8	M	33,43	Enucleation
65. Bansal et al (2015)	34	M	33,43	Enucleation
66. Jae-Yun jeon (2016)	15	M	18,28,38,48	Enucleation
67. Kashyap et al (2016)	23	M	Mx right and left supernumeraries and 23	Enucleation
68. Gnanaselvi et al (2016)	14	M	13,14,23,24,33,34,43,44	Enucleation
69. Dhupar A (2017)	8	M	13,14,15,23,24,25,34,35,44,45	Enucleation
70. Khandeparker RV et al (2018)	10	M	15, 23	Enucleation
71. Moturi K et al (2018)	19	F	Multiple teeth in Mx and Md arches	Enucleation
72. Vasiapphan H (2018)	27	M	38,48	Enucleation
73. Pant B et al (2019)	10	M	13,23	Enucleation
74. AlKhudair B (2019)	19	M	Mx sinus	Enucleation
75. Sindi AM (2019)	44	M	37,47	Enucleation
76. Sharma S, (2019)	27	F	18,28	Enucleation
77. Sethi A (2020)	12	M	35,45	NA
78. Bang KO et al (2021)	55	F	38,48	NA
79. Arici M (2022)	32	F	18,28	Enucleation

*NA- not available, F- Female, M- Male, Md- mandibular, Mx- maxillary, Rt-right, lt-left, Location - Tooth Numbering system according FDI (Federation Dentaire Internationale) System.

Discussion

A dentigerous cyst can be defined as “a cyst that encloses the crown of an unerupted tooth, expands the follicle and is attached to the cemento-enamel junction of the unerupted tooth”.[68] They tend to occur across a wide age range, peaking between the second and fourth decade. Dentigerous cysts' precise histogenesis is unknown; however the majority of publications favour a developmental origin from the tooth follicle.[69] Two different forms of dentigerous cysts were put forward by Al Talabani and Smith. One results from early stellate reticulum degeneration, which is linked to hypoplastic teeth, and the other develops after the crown completion through fluid buildup between the layers of decreased enamel epithelium.[70]

Racial, ethnic, sexual, and individual variables can affect how normally teeth erupt into the oral cavity, and these are typically taken into account when establishing standards for proper eruption.[71] There are numerous eruption regulating molecules like EGF, EGF-R, CSF-1R, CSF-1, IL-1R, IL-1, c- NFB, Fos, TGF-b1, MCP1, PTHrP, OPG, Cbfa-1, and RANK/RANK L that reside in dental follicle with a few in Stellate reticulum.[72] The anomalies in the eruption, however, might be caused by a gene abnormality.[73]

Syndromes such as Cleidocranial Dysplasia, Maroteaux-Lamy Syndrome, and Mucopolysaccharidosis, are frequently observed in conjunction with the bilateral and multiple cysts. Without these characteristics, bilateral dentigerous cysts are uncommon.[74] All of these circumstances disrupt tooth development or eruption in some way, which accounts for the presence of many cysts. The age and sex predilections for non-syndromic cases according to literature are given in the Graph (1). In terms of etiology, Batra P reported Chromosomal polymorphism in bilateral dentigerous cyst whose karyotyping revealed a large secondary constriction in chromosome 1qh+ 2.[75] The combined effect of

cyclosporin and a calcium blocker was highlighted by Yamalik K et al.[76] in the development of bilateral dentigerous cysts. But there is no proof linking them to the bilateral prevalence of dentigerous cysts, and they could simply be coincidental findings. In 2001, De Biase A et al. observed that long-term, concomitant usage of cyclosporine A and calcium channel blockers can result in bilateral mandibular dentigerous cysts.[77]

The most common site of occurrence is Mandibular, Maxillary third molars and maxillary cuspids but the literature search showed other combinations as well.[78] Aher V et al and jae Yun jeon reported occurrence of bilateral DC in both maxillary and mandibular third molars.[79,80] According to the cases reported thus far, Mandibular third molars (20.65%) are the most frequently affected site for Dentigerous Cysts, followed by mandibular premolars (11.95%) and maxillary canines (10.86%). Sizable fractions (44.56%) of cysts develop in other combinations at different jaw regions.[5-68]

Only 91 cases of multiple dentigerous cysts that are not syndromic have been documented thus far. (Table 1) In non-syndromic patients, multiple dentigerous cysts are regarded to be incredibly unusual. In a study of 2944 cases of odontogenic cysts, Ochsenius et al. found 546 patients with dentigerous cysts, 61 (11%) of whom had synchronous dentigerous cysts. None of these patients had any metabolic or syndromic conditions, but it is unclear how many of these cysts were bilateral or led to root resorption.[81]

A complicated interdisciplinary strategy is required for the management of many impacted teeth linked to dentigerous cysts. When planning the course of treatment, the patient's age, the location of their teeth, the number of impacted teeth associated with dentigerous cysts, and any accompanying metabolic, genetic, or syndromic disorders must all be taken into account. Patients with many impactions require guided eruption of numerous teeth with the use of coordinated multidisciplinary care.[82] Enucleation of minor lesions can prevent harm to the implicated permanent teeth. It is possible to decompress and marsupialize larger lesions to release the pressure inside the cysts, followed by enucleation—also known as Waldron's procedure—when the size has shrunk.[83,84] The management procedures followed so for the cases reported in literature was given in the Table 2.

Dentigerous cysts does not pose any challenges after complete removal as they do not reappear.[85] This is due to the diminished enamel epithelium's fatigued state after differentiating and forming tooth crown enamel before developing into a cyst.[86] Shah et al suspected spontaneous regression of these cysts after 3 years, without any intervention.[87]

Graph 1: Percentages of occurrence of Bilateral DC according to Literature: Age and Gender

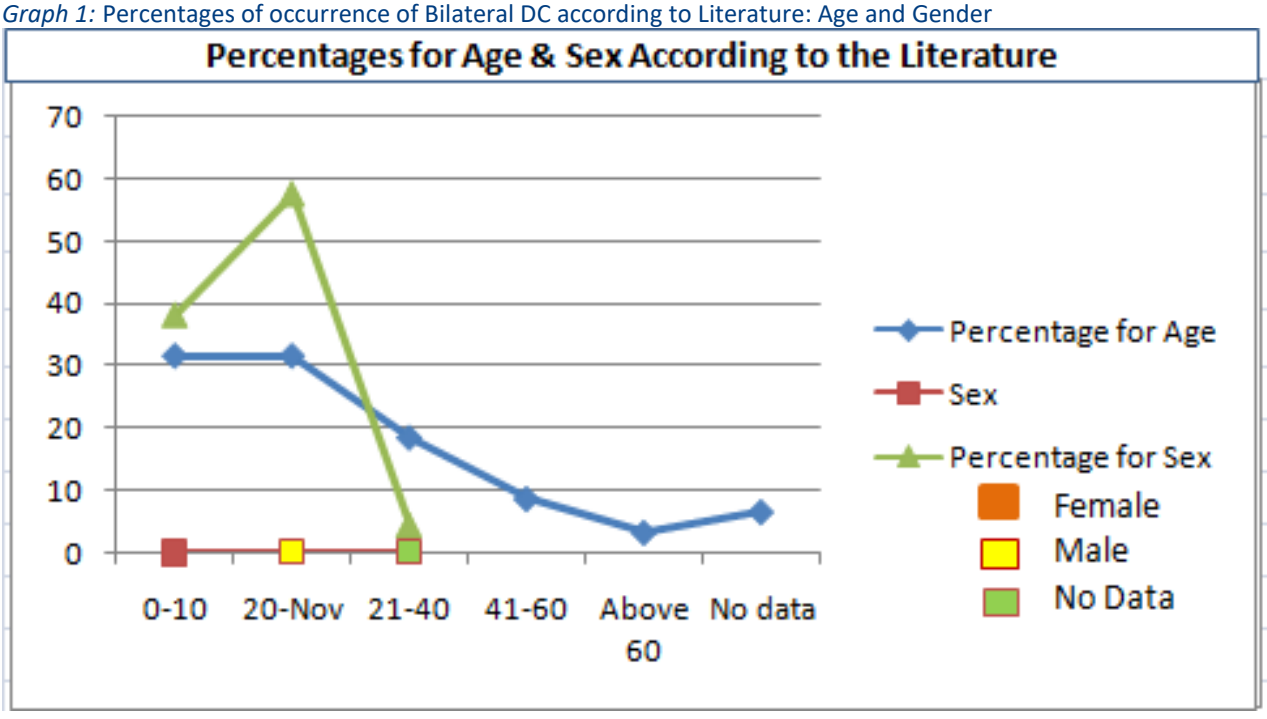

Table 2: Treatment modalities followed for bilateral DC according literature

S no.	Treatment	Percentage (%)
1.	Enucleation	75
2.	No treatment	2.17
3.	Marsupialization	15.21
4.	No data available	6.52
5.	Marsupialization + Enucleation	1.08

Conclusions

The occurrences of non syndromic dentigerous cysts are very rare and are challenging to draw conclusions on their etiopathogenesis due to their rarity and the dearth of evidence available in the literature.

Declaration by Patient

The authors attest to obtaining all necessary patient consent papers. The patient has agreed on the form to the publication of his photos and other clinical data in the journal. The patient is aware that while every attempt will be made to keep their identity a secret and that their name and initials won't be published, anonymity cannot be guaranteed.

Authorship Contribution

RTV, SP conceived and designed the study, RTV acquired the data; and SP, RM conducted data analysis and interpreted the results. RTV, SP wrote the initial and final drafts of the manuscript, and RM, RT, SRP, provided logistic support. All the authors drafted the manuscript and designed the tables with critical revisions. All authors discussed the results and commented on the manuscript. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

Conflict of Interest

None

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