

Prevalence of pulp stones: A systematic review and meta-analysis

RezaJannati¹ | Mahdi Afshari² | Mahmood Moosazadeh³ |

Seyedeh Zahra Allahgholipour¹ | Milad Eidy¹ | Mojtaba Hajihoseini¹

¹Student Research Committee, Mazandaran University of Medical Sciences, Sari, Iran

²Department of Community Medicine, Zabol University of Medical Sciences, Zabol, Iran

³Health Sciences Research Centre, Addiction Institute, Mazandaran University of Medical Sciences, Sari, Iran

Correspondence

Mahmood Moosazadeh, Health Science Research Center, Addiction Institute, Mazandaran University of Medical Sciences, Sari, Iran
Email: mmosazadeh1351@gmail.com.

Abstract

Objective: During the last years, different prevalences of dental pulps had been reported from different parts of the world. Combining these original estimates will provide useful information for policymaking in the field of health, treatment and research. This study aims to estimate the global prevalence of pulp stones.

Methods: Electronic databanks such as PubMed, Scopus, Science direct, Cochrane, and also Google scholar motor engine were searched by 2 researchers using related keywords. The heterogeneity between the results was assessed using Cochrane and I^2 indices. The initial estimates were represented by forest plot and were combined according to random effects models. Factors responsible for heterogeneity were investigated using meta-regression models, and studies responsible for heterogeneity were assessed following sensitivity analysis.

Results: Prevalence of pulp stones had been reported in 16 studies including 14 093 subjects. The pooled prevalence (95% confidence interval) of pulp stones among the total population, men and women were estimated as of 36.53% (27.17-45.88), 32.58% (24-41.15), and 39.23% (28.73-49.73), respectively. In addition, of 193 687 teeth investigated during the primary studies, 9.57% (95% confidence interval: 7.05-12.08) were affected by pulp stones.

Conclusion: Our meta-analysis shows a considerable prevalence of pulp stones, especially among women.

KEYWORDS

dental, meta-analysis, prevalence, pulp stone

1 | INTRODUCTION

Pulp stone is a calcified mass establishing within healthy or diseased teeth.¹ It may appear in the coronal or root portion of the pulp freely, adherent or embedded into the dentin. The stones have different sizes, and those under 200 microns are not visible by routine radiographical procedures.²

Several factors have been reported for pulp stones including age, mis-perfusion of the pulp, genetic background and prolonged stimulators such as dental decay or deep dental filling. Pulp necrosis was often revealed following trauma but sometimes is observed after orthodontic therapy. In general, pulp stone was considered as a part of ageing particularly in the elderly. However, it can develop during systemic or genetic diseases.³

Most researchers believe that pulp stones were developed during ageing. In addition, no enough evidence has existed regarding the asso-

ciation between gender and pulp stone. However, some studies show higher rates of these stones among women. Moreover, according to the most evidence, the rate is the same in both maxillary and mandibular bones.³

Prevalence of pulp stone has been reported between 8% and 90%. Such variations might be according to the design of the studies as well as the radiological techniques.⁴ The small stones are painless. However, larger ones can be painful.⁵

Based on our initial search, several studies have been carried out regarding the prevalence of pulp stones reporting various estimates just from the limited areas. No comprehensive study was found to estimate the global prevalence of this problem. Such total estimates are required for establishing appropriate treatment planning. Meta-analysis is a reasonable technique combining the results of primary studies. This study aims to estimate the total prevalence of pulp stones and its related factors in the world.

2 | MATERIALS AND METHODS

This study was conducted according to a predesigned protocol based on the PRISMA checklist. The protocol was registered in the PROSPERO (CRD42017071651)(6).

3 | SEARCH STRATEGY

Electronic databanks such as PubMed, Scopus, Science direct, Cochrane, and also Google scholar motor engine were searched by 2 researchers up to July 10, 2017 using the following keywords: "Dental Pulp Stone," "Pulp Stones, Dental," "Pulp Stones," "Stone, Pulp," "Pulp Calcification, Dental," "Prevalence," "Epidemiology," "Frequency." They also reviewed the references to find any additional relevant study.

4 | INCLUSION CRITERIA

Papers written in English, descriptive studies reporting the prevalence of pulp stone among men, women or total populations without any geographical limitation were included in our review.

5 | SELECTION OF THE STUDIES

All identified studies during the electronic search were reviewed. The duplicated evidence were omitted, and irrelevant papers were excluded after investigating the titles, abstracts, and full texts respectively.

6 | QUALITY ASSESSMENT

The relevant studies remained after the screening were quality assessed using the STROBE checklist. This standard tool assessed all aspects of the methodology of the primary studies such as sampling, type and study design, data collection, the definition of the variables, diagnostic criteria and statistical methodology. Minimum and maximum quality scores for each paper based on this checklist were 0 and 44, respectively, which were classified into low quality (less than 15.5), moderate quality (15.5-29.5) and high quality.^{7,30-44}

7 | DATA EXTRACTION

Study title, the name of first author, date and place of the study, type of the study, type of imaging applied for diagnosis, total sample size as well as the sample size of each gender, total prevalence of pulp stone as well as the prevalence for each gender, number of teeth studied and prevalence of stones based on teeth count were extracted from each study.

8 | STATISTICAL ANALYSIS

Data were analysed using Stata version 14. The heterogeneity of the results of the primary studies was assessed based on Cochrane and I^2 indices considering $P < 0.1$. The degree of heterogeneity was classified based on Higgins criteria.⁸ According to the significant heterogeneity, random effect model was applied for meta-analysis assuming that the primary studies are random samples of a larger population.⁹ Meta-regression models were designed to investigate the role of heterogeneity factors. Point and pooled prevalences of pulp stones were shown by forest plots. The role of each primary study in the heterogeneity was assessed during sensitivity analysis. Publication bias was checked using the Egger's test.

9 | RESULTS

During a comprehensive search, 952 studies were identified. After a sequential screening (duplicate exclusion, review of titles, abstracts and full texts), 5 case-control studies,¹⁰⁻¹⁴ 1 review literature¹⁵ and 8 studies due to not reporting prevalence¹⁶⁻²³ were excluded. The remaining 16 papers^{1-3,24-36} were quality assessed all of which had high to moderate quality scores and were considered eligible for meta-analysis (Figure 1 and Table 1).

All of the finally selected articles were cross-sectional, one of which was conducted in Australia, 2 were conducted in India, 4 studies were carried out in Iran, 1 in Iraq, 1 in Malaysia, 1 in Nigeria, 1 in Saudi Arabia and 5 studies were conducted in Turkey. All of these studies were published during 2002-2015.

These primary studies reported the prevalence of pulp stones among 14 093 subjects varied from 12% in Gulsahi study conducted among 519 Indian people to 63.6% reported by Colak among 814 patients in Turkey. Because the significant heterogeneity between the primary results (I^2 : 99.3%, Q : 2093.16, $P < 0.001$), the random effect model was applied to estimate the combined prevalence. The total prevalence of pulp stone was estimated as of 36.53% (95% confidence interval: 27.17-45.88) (Figure 2A). Meta-regression models revealed that the radiography was not a source of heterogeneity (I^2 = -6.54, P = 0.138). Sensitivity analysis showed that Turkal study had the most influence on the heterogeneity. So that omitting this study reduced the degree of the heterogeneity from 99.3% to 98.6% and also the prevalence of pulp stone was changed to 38.12% (29.36-46.90). Also, the Egger's test showed no evidence of publication bias (I^2 = 13.54, P = 0.124).

Prevalence of pulp stones among men was reported by 16 studies including 6114 subjects varied between 10% in the study conducted by Turkal among 3052 men in Turkey and 58.5% in a Colak study carried out among 352 Turkish individuals. Using random effect model (I^2 : 98%, Q : 741.87, $P < 0.001$), the total prevalence (95% confidence interval) of pulp stones among men was estimated as of 32.58% (24-41.15) (Figure 2B).

Prevalence of pulp stone among 7979 women was estimated in 16 studies. Minimum and maximum prevalences were reported from 7.02% in Gulsahi study carried out among 313 Indian women to 69.6%



FIGURE 1 Flowchart of study selection

conducted by Sisman among 303 Turkish women. Using random effect model (I^2 : 99.1%, Q : 1616.35, $P < 0.001$), the total prevalence (95% confidence interval) of pulp stone among women was estimated as of 39.23% (28.73–49.73) (Figure 2C).

Prevalence of pulp stone based on the number of investigated teeth was reported in 15 primary studies among 193 687 teeth. These estimates varied from 2.1% in Turkal study conducted among 96 240 teeth to 27.8% reported by Colak among 12 928 teeth in Turkey. Using random effect model because of significant heterogeneity (I^2 : 99.8%, Q : 7150.45, $P < 0.001$), pulp stone was observed in 9.57% (95% confidence interval) of the samples (Figure 2D).

10 | DISCUSSION

Our study estimated the global prevalence of pulp stones based on genders. Combining the results of studies carried out among 14 093 subjects, the total prevalence of pulp stone was estimated as of 36.5%. Minimum and maximum rates were observed in India (12%) and Turkey (63.6%), respectively. Different prevalences have been reported in

other countries as well as in different regions in each country. The type of diagnostic methods and study samples can be responsible for these variations. For example, such studies did not recruit eroded or repaired teeth samples.

Our study also estimated the prevalence of pulp stones among men and women. Just limited information was extracted from the primary studies in term of other factors such as age, background diseases, type of tooth and jaw. Therefore, subgroup analysis was not performed based on such variables. However, these issues were briefly mentioned in the results.

More than half of the primary studies reported higher prevalences of pulp stones among women.^{21,22,27–29,32} That was the case for the total estimate. Sener reported that higher frequency of bruxism among women is responsible for the higher rate of pulp stones.²⁷

Studies conducted by Hekmatian,³⁴ Kazemizadeh,³⁵ Javadzadeh,³⁶ and Al-Nathan²⁵ reported significantly higher rates of pulp stones among subjects aged over 50. In general, senile changes in the dental structure is inevitable which occurs following second dentin deposition, hypoperfusion of the pulp, atherosclerotic changes, and pulp destruction during ageing.¹

TABLE 1 Characteristics of the primary studies entered into the meta-analysis

Study number	First author	Publication year	County	Radiographic choice	Total		Male		Female		Teeth	
					<i>n</i>	Prevalence	<i>n</i>	Prevalence	<i>n</i>	Prevalence	<i>N</i>	Prevalence
1	Sreelakshmi	2014	India	OPG	150	53	75	52	75	54.7	4399	6
2	Gulsahi	2009	Turkey	Periapical radiograph	519	12	206	18.4	313	7.02	13 474	5
3	Al-Nazhan	2011	Saudi Arabia	Bitewing radiographs	600	46.8	319	36.7	281	58.4	8456	10.2
4	Ranjitkar	2002	Australian	Bitewing radiographs	217	46	123	44.7	94	47.9	3296	10.1
5	Colak	2012	Turkey	Bitewing radiographs	814	63.6	352	58.5	462	67.5	12 928	27.8
6	Şener	2009	Turkey	Periapical+bitewing radiographs	536	38	270	30.7	266	45.5	15 326	4.8
7	Turkal	2013	Turkey	Periapical radiograph	6912	12.7	3052	10	3860	14.2	96 240	2.1
8	Bains	2014	India	Bitewing radiographs	500	41.8	257	38.1	243	45.7	2180	12.3
9	Al-Ghurabi	2012	Iraq	OPG	390	34.8	169	36	221	33.9	10 510	2.6
10	Udoe	2011	Nigeria	Periapical radiograph	300	21	130	21.5	170	20.6	1154	9.9
11	Kannan	2015	Malaysia	Periapical radiograph	361	44.9	156	38.5	205	49.8	1779	15.7
12	Sisman	2012	turkey	Bitewing radiographs	469	57.6	167	30.3	302	69.6	6926	15
13	Ravanshad	2015	Iran	Periapical+bitewing radiographs	652	46.9	202	37.6	450	51	8244	11.25
14	Hekmatian	2014	Iran	Periapical radiograph	500	25.6	198	27.2	302	24.5		
15	Kazemizadeh	2008	Iran	Periapical+bitewing radiographs	800	20	318	21.7	482	18.9	2681	7.9
16	Javadzadeh	2014	Iran	Bitewing radiographs	373	20.9	120	21.7	253	20.5	6094	3.2

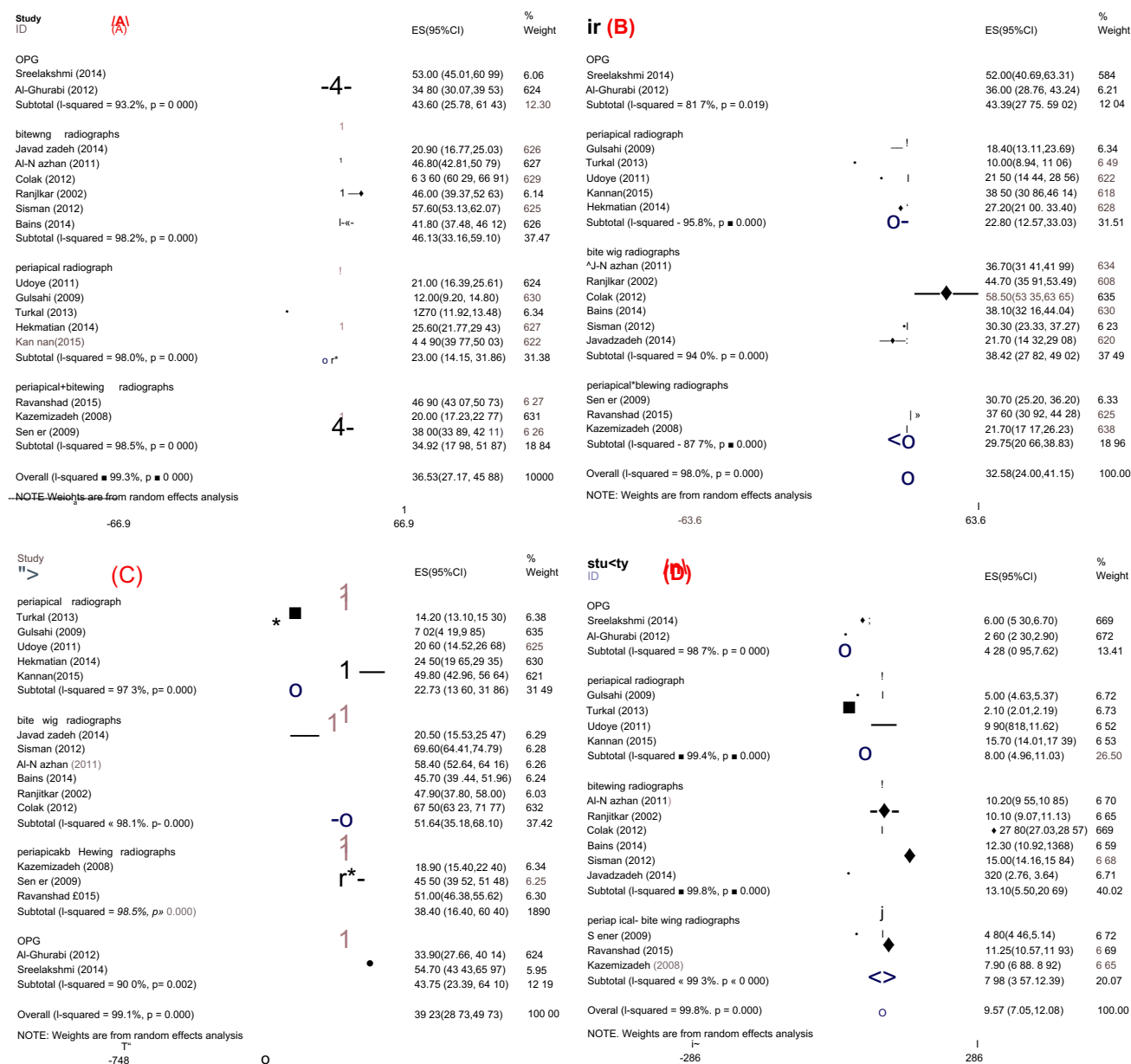


FIGURE 2 Prevalence of pulp stones by meta-analysis. A, Forest plot for point and pooled prevalence of pulp stones with 95% confidence interval. B, Point and pooled prevalence of pulp stones among men. C, Point and pooled prevalence of pulp stones among women. D, Point and pooled prevalence of pulp stones per studied teeth

Several studies showed that pulp stone is more common in noninfect teeth compared to the whole ones.^{26,27,31} Pulp calcification occurs in nonrepaired decayed teeth indicating that chronic pulp stimulation can be a reason for developing pulp stones.²⁶ Moreover, an association between periodontal diseases and pulp calcification has been observed by other researchers.²⁵ On the other hand, the combination of periodontal diseases and pulp stimulation increases the incidence of pulp degeneration and inflammation.²⁵

Few studies have been conducted to assess the association between pulp stones and systematic diseases. The previous findings of the correlation between renal and pulp stones are controversial.^{10,11,13,14} Moreover, the association between cardiovascular diseases and pulp stone has not been observed elsewhere.²⁷

Pulp stones are more common in molar teeth compared to premolar ones. It is also more common in premolar teeth than incisor teeth. The most common pulp stones develop in the first molar and premolar teeth.³² The reason is this tooth is the first tooth located in the mandibular bone,²⁵ therefore, has a longer exposure to degenerative changes. Also, it has a broader texture and more blood perfusion than the other teeth.²⁵ As reported in most studies,^{2,24,27,29,32,34} the maxilla is more affected than the mandible. However, the exact mechanism is unknown.

In general, pulp stones cause no limitation for endodontic procedures. Although, larger stones can limit access to the root canal. Also, the endodontic devices may be broken by the attached stones makes it difficult to access the canal.²⁵ Therefore, it is recommended to

provide appropriate preoperational imagings and using ultrasonic procedures for effective stone removal. Some of the idiopathic toothaches might be due to the pressure of the larger stones on the sensory nerves within the pulp.²⁵ In general, pulp stones do not need any treatment.

The present study suffers from limitations which are common in the meta-analysis on descriptive and prevalence studies. First, this meta-analysis has a high level of heterogeneity. It seems that heterogeneity a norm rather than an exception when many primary studies are including in a meta-analysis.³⁷ Second, our primary studies included limited data regarding important determinant factors of the heterogeneity such as family history of disease of interest (ie, pulp stone), the social status of individuals and medical comorbidity. Thus, the role of these factors in the heterogeneity could not be investigated by meta-regression models.³⁸ Third, results of meta-regression models indicate observational associations and was limited by ecological fallacy.³⁹ Therefore, our findings cannot identify a temporal causality in this meta-analysis. More longitudinal studies are required because they can determine the temporal association between predisposing factors and the risk of developing pulp stones. Lack of sufficient evidences from different countries is another limitation of the current study limiting the generalizability of the results. The majority of the primary studies were conducted in Middle East countries. Therefore, cultural and ethnic differences in lifestyle and genetic factors may affect the pooled estimates.⁴⁰ Moreover, we could not recruit studies published by languages other than English. Despite these limitations, this meta-analysis provides the most up-to-date information on the global prevalence of pulp stones.

The current meta-analysis showed that more than one-third of the world population especially women have pulp stones. We also revealed that approximately 10% of teeth are suffering from this problem. The results of meta-analysis on cross-sectional studies can provide several clinical implications including screening, prompts referral and early interventions.⁴¹ It is recommended to conduct further studies in term of pulp stone prevalence in all countries to provide better global estimates of the problem. Finally, designing studies assessing the potential risk factors of pulp stone can play a critical role in the preventive actions during the health policymaking.

CONFLICTS OF INTEREST

None.

REFERENCES

- Gulsahi A, Cebeci AI, Özden S. A radiographic assessment of the prevalence of pulp stones in a group of Turkish dental patients. *Int Endod J*. 2009;42(8):735-739.
- Çolak H, Çelebi AA, Hamidi MM, Bayraktar Y, Çolak T, Uzgur R. Assessment of the prevalence of pulp stones in a sample of Turkish central anatolian population. *Sci World J*. 2012;2012:804278.
- Udoye CI, Sede MA. Prevalence and analysis of factors related to occurrence of pulp stone in adult restorative patients. *Ann Med Health Sci Res*. 2011;(1):9-14.
- Moss-Salentijn L, Hendricks-Klyvert MS. Calcified structures in the human dental pulps. *J Endod*. 1988;14(4):184-189.
- Abbot PV, Yu C. A clinical classification of the status of the pulp and the root canal system. *Aust Dent J*. 2007;52(1):17-31.
- Moosazadeh Mahmood, Jannati Rez, Afshari Mahd. A protocol for estimating prevalence of pulp stone: a meta-analysis. PROSPERO 2017:CRD42017071651. Available from http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42017071651 (Accessed July 09,2017).
- Vandenbroucke JP, von Elm E, Altman DG, et al. Strengthening the reporting of observational studies in epidemiology (STROBE): explanation and elaboration. *IntJSurg*. 2014;12(12): 1500-24.
- Higgins JP, Thompson SG. Quantifying heterogeneity in a meta-analysis. *Stat Med*. 2002;21(11):1539-58.
- Cheung MW, Ho RC, Lim Y, Mak A. Conducting a meta-analysis: basics and good practices. *Int J Rheum Dis*. 2012;15(2):129-35.
- Moudi E, Kazemi A, Madani Z, Haghani S, Moudi E. A radiographic correlation between the presence of pulp stones and kidney stones. *CaspJAppl Scienc Res*. 2015;4(3):1-7.
- Kumar T, Puri G, Aravinda K, Laller S, Jatti D, Gupta R. Correlation between prevalence of pulp stones and renal stones in Panchkula region of India. *SRM J Res Dent Sci*. 2015;6(3):150-154.
- Dagistan S, Özkan M. The relationship of the chronic renal failure with carotid artery calcifications, dental pulp calcifications and dental pulp stones. *Atatürk Üniversitesi Diş Hekimliği Fakültesi Dergisi*. 2015;25:3.
- Patil SR. Prevalence of and relationship between pulp and renal stones: a radiographic study. *J Oral Biol Craniofac Res*. 2015;5(3):189-192.
- Tarim Ertas E, İnci M, Demirtas A, et al. A radiographic correlation between renal and pulp stones. *West Indian Med J*. 2014;63(6): 620-625.
- Goga R, Chandler NP, Oginni AO. Pulp stones: a review. *Int Endod J*. 2008;41(6):457-468.
- Nayak M, Kumar J, Prasad LK. A radiographic correlation between systemic disorders and pulp stones. *Indian J Dent Res*. 2010;21(3): 369-373.
- Tomczyk J, Komarnitki J, Zalewska M, Wisniewska E, Szopiński K, Olczyk-Kowalczyk D. The prevalence of pulp stones in historical populations from the middle euphrates valley (Syria). *Am J Phys Anthropol*. 2014;153(1):103-15.
- Talla HV, Kommineni NK, Yalamancheli S, Avula JS, Chillakuru D. A study on pulp stones in a group of the population in Andhra Pradesh, India: an institutional study. *J Conserv Dent*. 2014;17(2): 111-114.
- Swathy S, Gheena S, Varsha SL. Prevalence of pulp stones in patients with history of cardiac diseases. *Res J Phar Tech*. 2015;8(12): 1625-1628.
- Ezoddini Ardekani F, Mohammadi Z, Hashemian Z, SadrBafghi M, Hedayati A, Baghemalek R. Exploring the relationship between dental pulp stones and ischemic cardiovascular diseases. *J Dent Med*. 2009;22(1):74-80.
- Horsley SH, Beckstrom B, Clark SJ, Scheetz JP, Khan Z, Farman AG. Prevalence of carotid and pulp calcifications: a correlation using digital panoramic radiographs. *Int J Comput Assist Radiol Surg*. 2009;4(2): 169-173.
- Satheeshkumar PS, Mohan MP, Saji S, Sadanandan S, George G. Idiopathic dental pulp calcifications in a tertiary care setting in South India. *J Conserv Dent*. 2013;16(1):50-55.
- Yeluri G, Kumar CA, Raghav N. Correlation of dental pulp stones, carotid artery and renal calcifications using digital panoramic radiography and ultrasonography. *Contemp Clin Dent*. 2015;6(1): 147-151.
- Sreelakshmi N, Nagaraj T, Sinha P, Goswami RD, Veerabasaviah BT. A radiographic assessment of the prevalence of idiopathic pulp

- calcifications in permanent teeth: a retrospective radiographic study. *J Indian Acad Oral Med Radiol*. 2014;26(3):248-52.
25. Al-Nazhan S, Al-Shamrani S. A radiographic assessment of the prevalence of pulp stones in Saudi adults. *Saudi EndodJ*. 2011;1(1):19-19.
 26. Ranjitkar S, Taylor JA, Townsend GC. A radiographic assessment of the prevalence of pulp stones in Australians. *Aust Dent J*. 2002;47(1):36-40.
 - TJ. Şener S, Cobankara FK, Akgünlü F. Calcifications of the pulp chamber: prevalence and implicated factors. *Clin Oral Investig*. 2009; 13(2):209-215.
 28. Turkal M, Tan E, Uzgur R, Hamidi MM, Çolak H, Uzgur Z. Incidence and distribution of pulp stones found in radiographic dental examination of adult Turkish dental patients. *Ann Med Health Sci Res*. 2013;3(4):572-576.
 29. Bains SK, Bhatia A, Singh HP, Biswal SS, Kanth S, Nalla S. Prevalence of coronal pulp stones and its relation with systemic disorders in northern Indian central Punjabi population. *ISRN Dent*. 2014;2014:617590.
 30. Al-Ghurabi ZH, Najm AA. Prevalence of pulp stone (Orthopantomographic-based). *J Baghdad Coll Dent*. 2012;24(2):80-84.
 31. Kannan S, Kannepady SK, Muthu K, Jeevan MB, Thapasum A. Radiographic assessment of the prevalence of pulp stones in Malaysians. *J Endod*. 2015; 41(3):333-337.
 32. Sisman Y, Aktan A, Tarım-Ertas E, Çiftçi M, Şekerçi A. The prevalence of pulp stones in a Turkish population. A radiographic survey. *Med Oral Patol Oral Cir Bucal*. 2012;17(2):212-217.
 33. Ravanshad S, Khayat S, Freidonpour N. The Prevalence of pulp stones in adult patients of Shiraz dental school, a radiographic assessment. *J Dent (Shiraz)*. 2015;16(4):356-361.
 34. Hekmatian E, Shokrgozar A, Maleki V. Assessment of prevalence of pulp stones on digital panoramic radiographs of patients referring to Isfahan dental school in 2013. *J Isfahan DentSch* 2015;11(2):163-169.
 35. Kazemizadeh Z, Zargarpoor R, Ahmadi KJ. A Radiographic assessment of the prevalence of pulp stones in patients referred to Rafsanjan faculty of dentistry in 2008. *Qom Univ Med SciencJ*. 2011;2(18):29-33.
 36. Javadzadeh A, Mohtavipour ST, Vosooghi Y, PourHabibi Z, Nemati S, Dalili Z. Prevalence of Pulp stones in radiographs of patients, referred to Guilan school of dentistry in 2011. *J Mashhad Dent Sch*. 2014;38(2):99-106.
 37. Quek YH, Tam WWS, Zhang MWB, Ho RCM. Exploring the association between childhood and adolescent obesity and depression: a meta-analysis. *Obes Rev*. 2017;18(7):742-754.
 38. Lim GY, Tam WW, Lu Y, Ho CS, Zhang MW, Ho RC. Prevalence of depression in the community from 30 countries between 1994 and 2014. *Sci Rep*. 2018;8(1):2861.
 39. Ho RC, Cheung MW, Fu E, et al. Is high homocysteine level a risk factor for cognitive decline in elderly? A systematic review, meta-analysis, and meta-regression. *Am J Geriatr Psychiatry*. 2011;19(7):607-17.
 40. Zhang MW, Ho RC, Cheung MW, Fu E, Mak A. Prevalence of depressive symptoms in patients with chronic obstructive pulmonary disease: a systematic review, meta-analysis and meta-regression. *Gen Hosp Psychiatry*. 2011;33(3):217-23.
 41. Loh AZ, Tan JS, Zhang MW, Ho RC. The global prevalence of anxiety and depressive symptoms among caregivers of stroke survivors. *J Am Med Dir Assoc*. 2017;18(2):111-116.

How to cite this article: Jannati R, Afshari M, Moosazadeh M, Allahgholipour SZ, Eidy M, Hajihoseini M. Prevalence of pulp stones: a systematic review and meta-analysis. *J Evid Based Med*. 2018;1-7. <https://doi.org/10.1111/jebm.12331>