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The divided zygoma: a meta-analysis of its prevalence with a review of the literature

D. Plutecki et al., The divided zygoma

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ABSTRACT

Background: Divided zygoma (DZ) is an important structure in the midfacial region. The anatomy of DZ is poorly researched, but knowledge about this entity could be useful during posttraumatic facial reconstructions. The aim of this study was to estimate the prevalence and anatomy of DZ in different regions around the world. Therefore, the authors performed a meta-analysis, including all studies that report extractable data on the DZ.

Materials and methods: The main online medical databases such as PubMed, EBSCO, ScienceDirect, Web of Science, SciELO, BIOSIS, Current Content Connect, Korean Journal Database and Russian Citation Index, were utilized to gather all studies on anatomical characteristics, prevalence, symmetry, and a number of divisions of zygomatic bone.

Results: A total of 20 studies were included in this meta-analysis. Data were grouped and analyzed in 5 categories: (1) prevalence of DZ bone, (2) prevalence of DZ skulls, (3) gender prevalence of DZ with sides, (4) divisions of zygomatic bone, (5) symmetry of DZ.

Conclusion: In conclusion, the authors of the present study believe that this is this study can be considered an up-to-date meta-analysis regarding the prevalence, divisions, and symmetry of the DZ. The data provided by the present study may be useful information for physicians in recognizing the DZ of the fracture and may be important information during

zygomatic bone osteotomy. Detailed anatomical knowledge of the midfacial region can prevent surgical complications when operating in this area.

Key words: os japonicum, divided zygoma, zygomatic bone, bipartite zygomatic bone, tripartite zygomatic bone, facial surgery, facial reconstruction

INTRODUCTION

Divided zygoma (DZ), also called os japonicum (OJ) because of its relatively high frequencies among modern human populations in Japan [15], is a division of zygomatic bone into two or more partitions. The division of the malar bone is accepted as an epigenetic variation [27]. Hilgendorf observed two zygomatic bones out of 11 Japanese skulls showing bipartition, and that was the reason for the name OJ [14]. Although some authors pointed out that the frequency of DZ in the Japanese population is too low to consider this character as a Japanese trait [22], many authors still use this synonym. Typically, the zygoma is a single midfacial bone and plays a significant role in the support and integration of the craniofacial skeleton, and the masticatory apparatus by its attachment to the masseter muscle [38]. It also contributes to the formation of the lateral wall and floor of the orbit, parts of the temporal and infratemporal fossa [36]. The zygomatic bone has three surfaces: malar, temporal and orbital. The malar surface has a small aperture for the passage of zygomatico-facial vessels and nerves [19].

There are great differences in opinion regarding the ossification of the zygomatic bone. Generally, it is thought that the human zygoma has only one ossification center, which appears in the fetus at eight weeks [5, 8, 20, 23, 37]. However, Buchanan's Manual of Anatomy [17] emphasizes that three ossification centers of the anterior, posterior, and inferior parts of the zygoma fuse to form the mature bone. Consequently, the DZ will occur if these three ossification centers fail to fuse, resulting in zygomaticum bipartium or more divisions [17].

The sutures of the craniofacial skeleton are described as fibrous joints, serving as important loci of craniofacial growth through their interactions with surrounding tissues and structures, especially the zygomatico-facial vessels and nerves [26, 30]. Biomechanically, sutures are relatively weak sites in the otherwise rigid skull [33–35]. Zygomatic fractures are

one of the most common facial bone fractures due to the prominent location of the zygoma that makes it prone to injuries [4]. This type of injury is common in contact sports or car accidents, which cause extensive and multiple fractures [19]. Supernumerary sutures could be misinterpreted as fracture lines in the radiograph plain, even if these lines do not have a typical appearance of a zygomatic fracture [24, 40]. It is crucial to differentiate fracture lines from sutures on the malar bone during craniofacial, maxillofacial, or zygomatic bone osteotomy reconstruction procedures [21].

The “norm” in anatomy is not as precise a concept as one would wish, and can be considered an approximation [41]. Understanding the variability of the zygomatic bone can be of great clinical significance when performing facial plastic and reconstructive surgeries, such as posttraumatic facial reconstructions. Detailed and precise anatomical knowledge is essential for surgeons in order to minimize potential complications associated with the craniofacial area. Therefore, the objective of the present study was to provide physicians, especially surgeons, with useful data on the prevalence of DZ and its anatomical features. To achieve this, a systematic search of the literature and a meta-analysis were performed.

MATERIALS AND METHODS

Search strategy

Online medical databases such as PubMed, ScienceDirect, EBSCO, Web of Science, SciELO, BIOSIS, Current Content Connect, Korean Journal Database, and Russian Citation Index, were used to gather all studies on anatomical characteristics, prevalence, symmetry, and a number of divisions of zygoma. The study collection ended in May 2022. In agreement with the Boolean technique, the following search terms were employed: (os japonicum) OR (divided zygoma) OR (bipartite zygomatic bone) OR (tripartite zygomatic bone). Search terms were individually adapted to each database to minimize potential bias. Neither the date, language, type of article, nor text availability conditions were applied. An additional search was conducted through the references of the identified studies at the end of the search stage to ensure the accuracy of the process. During the study, the Preferred Reporting Items for Systematic Reviews and Meta-Analyzes (PRISMA) guidelines were followed. Furthermore, the Critical Appraisal Tool for Anatomical Meta-analysis (CATAM) was used to provide the highest quality findings [6].

Eligibility assessment

The database search and the manual search identified a total of 347 studies that were initially evaluated by two independent reviewers. In addition, nine studies were added through reference searching. After removing duplicates and irrelevant records, a total of 22 articles were qualified for full text evaluation. To minimize potential bias and maintain accurate statistical methodology, articles such as case reports, case series, conference reports, reviews, letters to editors, and studies that provided incomplete or irrelevant data were excluded. The inclusion criteria consisted of original studies with extractable numerical data on the subject of this study. Finally, a total of 20 studies were included in this meta-analysis. Additionally, the AQUA Tool, which was specifically designed for anatomical meta-analyses, was used to minimize the potential bias of included studies [13].

Data extraction

Data from qualified studies were extracted by two independent reviewers. Qualitative data, such as year of publication, country and continent of origin, data collection methodology, and information on diseases in the studied groups, were collected. Quantitative data, such as sample size, numerical data on anatomical characteristics, prevalence, symmetry, and divisions of zygomatic bone, were also collected. Any discrepancies between studies identified by the two reviewers, were resolved by contacting the authors of the original studies whenever possible or by consensus with a third reviewer.

Statistical analysis

To perform the meta-analyses, STATISTICA version 13.1 software (StatSoft Inc., Tulsa, OK, USA) and MetaXL version 5.3 software (EpiGear International Pty Ltd, Wilston, Queensland, Australia) were used. A random-effects model was used in all analyses. The heterogeneity among the studies was evaluated, using both the chi-square test and the I-square statistic. The I-squared statistic was interpreted as follows: 0-40% as “might not be important”; 30%-60% as 'may represent moderate heterogeneity; 50%-90% as “may represent substantial heterogeneity”; 75%-100% as “may represent considerable heterogeneity”. The P-value <0.05 and the confidence intervals (95% CI) were used to find statistically significant differences between the studied groups. In the case of overlapping confidence intervals, differences were considered statistically insignificant.

RESULTS

Search results

After the 22 selections of the initially accepted studies, a total of 2 studies were excluded. They were disqualified due to the lack of relevant data. Finally, a total of 20 studies were included in this meta-analysis. According to the PRISMA guidelines, an overall data collection process is presented in Figure 1. In addition, the characteristics of all the submitted studies are collected in Table 1.

Prevalence of divided zygoma on the sides

A total of 49734 zygomatic bones were analyzed in relation to the prevalence of DZ. Furthermore, 14324 left and 14324 right zygomatic bones were analyzed. The pooled prevalence of any DZ was 1.69% (95% CI 0.73% - 3.01%). The pooled prevalence of any DZ on the left was shown to be 0.72% (95% CI: 0.28% - 1.36%) and on the right 0.98% (95% CI: 0.44% - 1.70%). Despite the general results, additional regional analyzes were also enrolled. All the results mentioned above and the more detailed results are gathered in Table 2.

Prevalence of divided zygoma in skull

A total of 17790 skulls were analyzed concerning the prevalence of DZ. The pooled prevalence of any DZ was shown to be 1.36% (95% CI: 0.76% - 2.12%). Despite the general results, additional regional analyzes were also enrolled. All the results mentioned above and the more detailed results are gathered in Table 2.

Divided zygoma prevalence according to the gender

Analysis of the prevalence of DZ according to sex and side was performed on a total of 1,414 zygomatic bones of the female group and 1,084 zygomatic bones of the men's group. The zygomatic bones were divided into two subgroups: the left and right side. The pooled prevalence for each group was 1.08% (95% CI: 0.00% - 4.02%) for the women and 0.87% (95% CI: 0.00% - 2.50%) for the men's group. Despite the general results, additional

analyzes were also enrolled in the left and right subgroups. All the results mentioned above and the more detailed results are gathered in Table 3.

Divisions of zygomatic bone

An analysis of the number of divisions of the zygomatic bone in DZ was performed on a total of 271 zygomatic bones. Bipartite zygomatic bone was found to be the most common, with a prevalence of 95.08% (95%CI: 88.75% - 99.06%). On the other hand, tripartite zygomatic bone was found, with a prevalence of 2.77% (95%CI: 0.75% - 5.82%). Despite the general results, additional regional analyzes were also enrolled. All the results mentioned above and the more detailed results are gathered in Table 4.

Symmetry of divided zygoma

An analysis of the symmetry of the occurrence of DZ was performed on a total of 46 DZ. The bilaterally DZ was found to be the most common, with a prevalence of 60.18% (95%CI: 46.27% - 73.34%). However, unilateral DZ was found, with a prevalence of 39.82% (95%CI: 26.66% - 53.73%). Despite the general results, additional regional analyzes were also enrolled. All the results mentioned above and the more detailed results are gathered in Table 4.

DISCUSSION

The prevalence of DZ has been extensively discussed in the literature. The DZ was first described in 1779 by Sandifort as a single case report [15]. In the nineteenth and twentieth centuries, some researchers had examined the DZ [9, 18]. Hilgendorf was the first author to give the name 'os japonicum' for DZ due to his research. In his study, two DZ were found in 11 Japanese skulls [14]. However, this synonym was not well received by everyone because of its occurrence in other populations, especially in a study conducted by Martin, where it was pointed out that the prevalence of DZ in the Japanese population is too low to consider that this structure has a Japanese trait [9, 22]. Despite this controversy, many researchers still use the name 'os japonicum' for this structure [2, 12, 16, 29, 32]. Although, in 1998, a Japanese researcher, Hanihara et al. made a huge analysis of DZ prevalence around the world. After this examination, they concluded that the trait of DZ is not specified for

Japanese but for the East Asian population [10]. The os zygomaticum bipartitum was also called “os ainonicum” by Belz, a German pathologist [25]. However, Koganei et al. found that no individuals who had a complete division of the zygomatic bone in the skulls of Hokkaido (homeland of the Ainu people) and Hanihara confirmed that Asian people have a higher frequency of DZ than Ainu people [10, 18]. In 1984, Pardoe, who was the only one who made the exploration in Australia, stated that searching for a complete OJ is essentially fruitless since only one DZ was found in that region [29]. In addition, other geographic regions, except Asian populations, have small elucidated data by the authors up to the present times. Some researchers collected their data from the skulls from the old days, such as: Kozintsev, who used skulls from the bronze age to find the posterior trace of the OJ, or Anil, who found DZ among Anatolian skulls from the 18th century, and Wang and Zhang, who described many DZs in museums collections and archeological sites [1, 33, 38]. The majority of the studies investigating the DZ focused purely on the prevalence of this structure, rather than its morphometric properties [33].

There has been a lot of controversy regarding the prevalence of DZ in many different geographical groups. The DZ was previously described as a Japanese trait, which caused the name ‘os japonicum’, however, not all authors agreed with this statement. Some researchers argued that DZ seems to be an East Asian trait [10]. The results of the present meta-analysis show that there is no statistically significant difference between the overall population and the Asian population group ($p > 0.05$). Moreover, other region groups (Europe, North and South America, and Australia and Oceania) have similar statistical results. This score applies to both the prevalence of DZ in the skull and single zygomatic bone. Therefore, a change in the nomenclature of the DZ seems redundant. Wang and Dechow claimed that the prevalence of DZ in Rhesus macae is significantly lower than in humans, especially in East Asia and South Africa [33]. Some researchers have also seen no evidence of the occurrence in Homo Sapiens, Homo erectus, Australopithecus, and other fossil hominids of DZ traits [15, 39]. The results of DZ lateralization show that the right side of the zygomatic bone appears to be more affected by DZ than the left side (0.98% for the right side and 0.72% for the left side). The DZ was prevalent more often in the female group (1.08%). In both sexual groups, the right side was more frequent than the left side; however, all the above results did not show any statistical significance ($p > 0.05$). Similarly, Anil et al. did not observe any differences in gender and side preferences in the cadaveric and radiograph groups [1]. Zhang also reported

that the existence of bipartite or tripartite zygomatic bones was not related to gender, age, or side [38].

The divisions of the zygomatic bone depend on many sutures that form during prenatal time. Hauser and De Stefano categorized the sutures on the zygomatic bone into four subgroups for bipartite zygomatic bone and into two for tripartite zygoma: type I, simple horizontal inferior; type II, simple oblique lateral; type III, simple horizontal superior; type IV, simple oblique medial; type V, horizontal inferior + oblique medial; type VI, horizontal superior + oblique lateral [12]. The same criteria could be used in radiological findings [1]. Anil et al. observed that type I and IV were significantly frequent. This classification was not included in the present meta-analysis because hardly any authors used this division in their studies. Some researchers observed that the horizontal division could be complete or incomplete [1, 29]. Interestingly, there is a high frequency of a vestigial (incomplete) transverse zygomatic suture in the zygomatic bone, mainly in the posterolateral part of the zygoma in human populations worldwide (10-25%) [7, 11, 28]. The bipartite zygomatic bone is much more common than the tripartite zygomatic bone ($p < 0.05$). However, there are no statistically geographical traits of the bipartite zygoma. The bilateral symmetry of the DZ is often that of the unilateral one (60.18%), but there are no statistically significant results ($p > 0.05$). The probable explanations for this situation could be the small amount of studies on this subject; data are obtained from a small number of sources, and therefore, the data is homogeneous ($I^2 = 0.00$).

Knowledge about the existence of DZ, its prevalence, symmetry, sexual dimorphism, and lateralization in different populations could be of great importance in neurosurgery and reconstructive procedures. This knowledge can be especially important in patients with facial trauma, particularly fractures of the buccal surface of the zygomatic bone shaft, as the zygomatic bone is the most prominent part of the facial skeleton [19, 31]. After zygomaticomaxillary complex fracture all patients have performed the 3-dimensional analysis of the degree of recovery of malar asymmetry [3]. This 3D image is essential before reconstruction operation and the DZ could be seen in the frontal view, which could be misunderstanding. It is crucial to not confuse potential fracture lines with the sutures, which divide the zygomatic bone, for a correct diagnosis. Furthermore, the DZ is also a clinically important structure for the plastic or maxillofacial surgeon during the osteotomy operation, which is used in patients with deformed viscerocranium, malocclusion, speech defects, or

nasal defects. The said procedure is usually focused on the nasal bone and nasal septum to improve the patient's respiration and aesthetics. In both aspects, having knowledge that the zygomaticofacial vessels and nerves pass through the zygomaticofacial foramen, could be clinically significant to prevent undesirable complications [19].

This study is not without limitations and is burdened with potential bias, as the results of this meta-analysis are only as accurate as the results from the submitted studies. A potential morphometry dimorphism in the anatomical features of the DZ was not established due to the lack of data. Analogically, no relation was enrolled with respect to the other structures surrounding the zygomatic bone statistics.

CONCLUSIONS

In conclusion, the authors of the present study believe that this is this study can be considered and up-to-date meta-analysis regarding the prevalence, divisions, and symmetricity of the DZ. The most common type of DZ is the bilaterally DZ bone (60.18%). The data provided by the present study may be useful for doctors to recognize the DZ from potential fractures and during zygomatic bone osteotomy. Detailed anatomical knowledge of the midfacial region can prevent surgical complications when operating in this area.

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List of abbreviations: OJ - Os Japonicum; DZ- divided zygoma

Table 1. Characteristics of the studies included in this meta-analysis

First Author	Year	Continent	Country	Methodology	Number of zygomatic bones studied	Number of zygomatic bones with Os Japonicum
Anil, A.	2000	Asia	Turkey	Cadaveric	2614	51
Bhargava, K.	1960	Asia	India	Cadaveric	200	13
Dimowski, N.	2012	Europe	Serbia	Cadaveric	616	3
Ding, S.	1961	Asia	China	Cadaveric	1638	21
Gong, S.	1965	Asia	China	Cadaveric	2036	10
Hanihara, T.	1998	Multipopulational study		Cadaveric	19582	102
Hu, X.	1985	Asia	China	Cadaveric	1600	11
Jeyasingh, P.	1982	Asia	India	Cadaveric	1000	40
Jit, I.	1960	Asia	India	Cadaveric	200	5
Kozintsev, A.	1999	Multipopulational study		Cadaveric	11202	777
Kundu, B.	2016	Asia	India	Cadaveric	286	6
Li, Y.	1985a	Asia	China	Cadaveric	400	9
Li, Y.	1985b	Asia	China	Cadaveric	664	24

Mangalgi, A.	2015	Asia	India	Cadaveric	228	1
Nikolova, S.	2017	Europe	Bulgaria	Cadaveric	2746	1
Pardoe, C.	1984	Australia	Australia	Cadaveric	2576*	1*
Soni, J.	2016	Asia	India	Cadaveric	486	21
Wang, Q.	2016	Asia	China	Cadaveric	280	3
Yang, Y.	1987	Asia	China	Cadaveric	1666	4
Zhang, Q.	2019	Asia	China	Cadaveric	2290	24

* number of skulls studied and number of skulls with Os japonicum
Table 1 | Characteristics of the studies included in this meta-analysis

Table 2. Results established in this meta-analysis regarding the prevalence of the Os Japonicum in each category. LCI – lower confidence interval. HCI – higher confidence interval. Q – Cochran’s Q.

Source of data	Category	N	Prevalence	LCI	HCI	Q	I ²
Results obtained analyzing a number (n) of zygomatic bones	<i>Os Japonicum prevalence</i>						
	Overall	4973 4	1.69%	0.73 %	3.01 %	1380.4 1	98.7 0
	In Asian population	2814 3	2.07%	1.07 %	3.38 %	657.53	97.4 1
	In European population	1021 2	0.96%	0.00 %	3.52 %	316.26	98.7 4
	In South and North American populations	2796	1.00%	0.00 %	5.16 %	64.77	96.9 1
	<i>Prevalence of Os Japonicum occurring on the left side</i>						
	Overall	1432 4	0.72%	0.28 %	1.36 %	47.57	83.1 8
	In Asian population	6546	1.13%	0.77 %	1.57 %	7.79	35.8 3
	In European population	3104	0.18%	0.00 %	0.54 %	7.47	59.8 2
	<i>Prevalence of Os Japonicum occurring on the right side</i>						
	Overall	1432 4	0.98%	0.44 %	1.70 %	49.02	83.6 8
	In Asian population	6533	1.49%	1.00 %	2.07 %	9.90	49.5 2
In European population	3094	0.27%	0.04 %	0.66 %	5.47	45.1 3	
Results obtained analyzing a number (n) of skulls	<i>Os Japonicum prevalence</i>						
	Overall	1779 0	1.36%	0.76 %	2.12 %	130.43	89.2 7
	In Asian population	7689	2.11%	1.34 %	3.04 %	47.46	76.8 2
	In Australian and Oceanian populations	4591	0.07%	0.01 %	0.18 %	1.72	0.00

	In European population	3232	0.27%	0.05%	0.64%	5.37	44.16
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Table 3. Results of the prevalence of the Os Japonicum in each sex. LCI – lower confidence interval. HCI – higher confidence interval. Q – Cochran’s Q.

Category	N	Prevalence	LCI	HCI	Q	I ²
Females						
Os Japonicum overall in women	1414	1.08%	0.00%	4.02%	12.06	75.13
Os Japonicum on the left side in women	705	1.25%	0.00%	3.64%	5.33	43.73
Os Japonicum on the right side in women	709	1.37%	0.00%	4.26%	6.37	52.91
Males						
Os Japonicum overall in men	1084	0.87%	0.00%	2.50%	30.53	90.17
Os Japonicum on the left side in men	857	2.02%	1.17%	3.08%	2.95	0.00
Os Japonicum on the right side in men	855	2.08%	0.00%	5.63%	7.75	61.27

Table 4. Results of this meta-analysis regarding the number of divisions of zygomatic bone and the symmetry of occurrence of the Os Japonicum. LCI – lower confidence interval. HCI – higher confidence interval. Q – Cochran’s Q.

Category	N	Prevalence	LCI	HCI	Q	I ²
Number of divisions of the zygomatic bone in Os Japonicum						
Overall prevalence of bipartite zygomatic bone	271	95.08%	88.75%	99.06%	28.03	57.19
Overall prevalence of tripartite zygomatic bone		2.77%	0.75%	5.82%	14.62	17.90

Prevalence of bipartite zygomatic bone in Asian population	165	93.69%	86.33%	98.52%	16.91	46.78
Prevalence of tripartite zygomatic bone in Asian population		4.70%	1.95%	8.48%	6.28	0.00
Symmetricity of occurrence of the Os Japonicum						
Overall prevalence of Os Japonicum occurring bilaterally	46	60.18%	46.27%	73.34%	6.17	0.00
Overall prevalence of Os Japonicum occurring unilaterally		39.82%	26.66%	53.73%	6.17	0.00
Prevalence of Os Japonicum occurring bilaterally in Asian population	42	63.98%	49.32%	77.47%	2.89	0.00
Prevalence of Os Japonicum occurring unilaterally in Asian population		36.02%	22.53%	50.68%	2.89	0.00

Figure 1. Flow diagram presenting process of collecting data included in this meta-analysis

Figure 2. Illustrations of the single, bipartite and tripartite zygomatic bone. A – Single zygomatic bone. B – Bipartite Zygomatic Bone (os japonicum / divided zygoma). C – Tripartite zygomatic bone (os japonicum / divided zygoma). Z – zygomatic bone. M – maxilla. N – nasal bone. F – frontal bone. S – sphenoid bone. P – parietal bone. T – temporal bone.

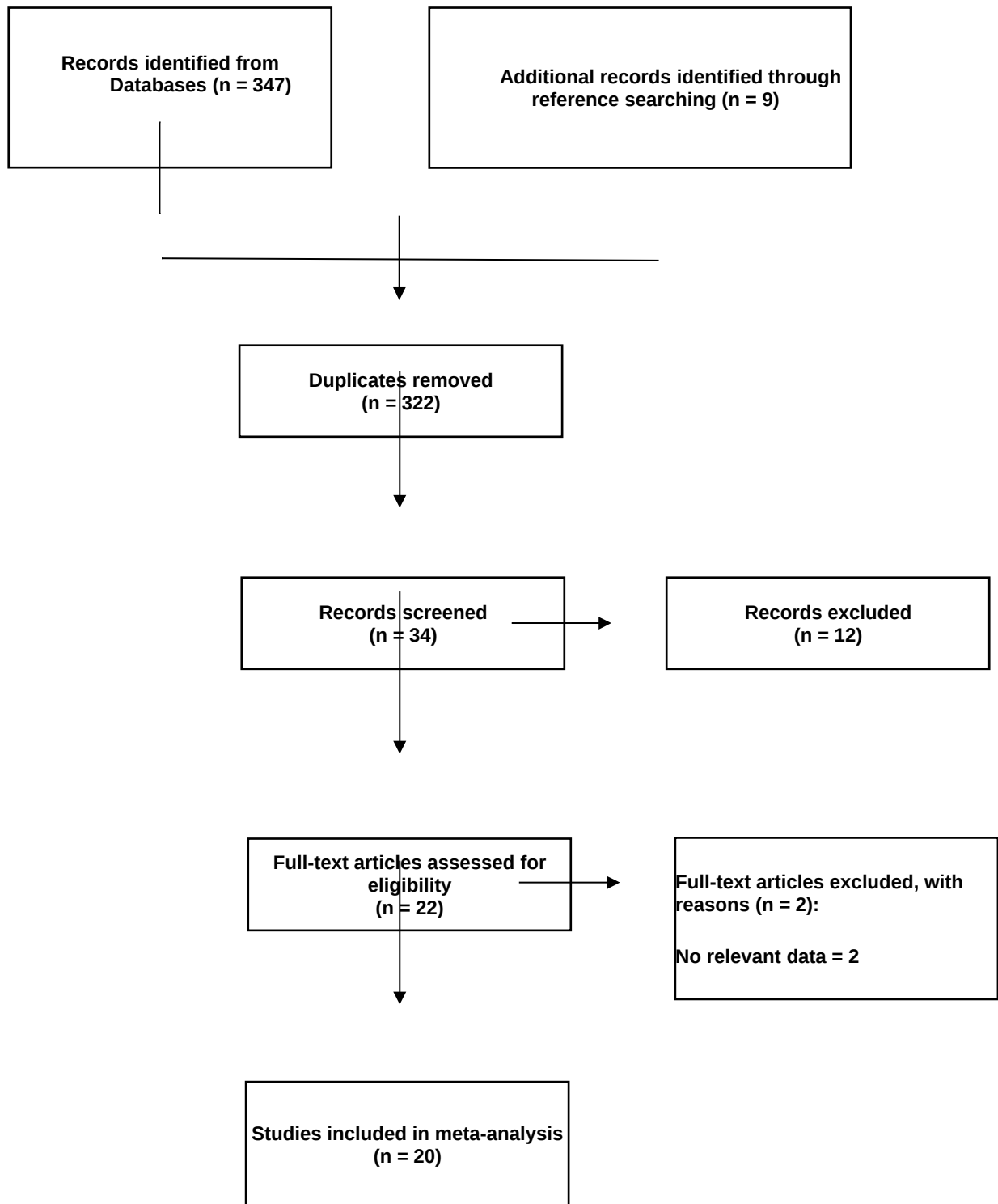


Figure 1 | Flow diagram presenting process of collecting data included in this meta-analysis

