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Review Article

A meta-analysis: could we predict the malignancy of solid pseudopapillary neoplasm?

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Abstract: Objective: To evaluate the risk factors influencing the malignant potential of solid pseudopapillary neoplasm (SPN). Methods: This meta-analysis used MEDLINE (PubMed), EMBASE and web of science including 14 cohort studies reporting the risk factors influencing the malignant potential after the initial operation on SPN up to March 2017. Review Manager Software 5.2 was used for meta-analysis. Results: 14 studies with a total of 763 patients were included in our meta-analysis. In all the variables, age and tumor size were significantly correlated with malignancy. Conclusion: Malignant SPNs tended to be larger in diameter and younger in age than benign type. In particularly, larger tumor size may be a crucial factor for decision of aggressive resection.

Keywords: Solid pseudopapillary neoplasm, pancreas, malignancy

Introduction

Solid pseudopapillary neoplasm (SPN) is a rare pancreatic tumor predominantly affecting young women with low malignant potential [1]. It usually has a favorable prognosis, with just over 95% of patients reported as disease free after surgical resection and with less than 2% mortality [2]. In the year of 2010, the World Health Organization (WHO) classified SPN as a low-grade malignant neoplasm. Approximately 10% to 15% cases of SPN are malignant, which could require the good survival with the resection [3], however due to higher risk of recurrence and mortality, aggressive surgical approach is warranted especially in the malignant cases such as local invasion, metastasis [4-7]. There has been a dramatic increase of reported SPN in the world over the past few decades [2]. Unfortunately the detection of risk factors of malignancy is of utmost importance, but remains unclear.

Materials and methods

Search strategy

Studies evaluating the factors influencing the malignancy of SPN were retrieved from the Embase, PubMed, and Web of Science before March 2017. We used the following free-text search terms in "All fields": "solid pseudopapillary neoplasm" and "risk factors" and "malignancy". There was no language restriction and no methodological filters. A recursive search of the reference of selected studies, review articles and guidelines were performed manually to identify the rest of potential relative articles.

Study inclusion/exclusion criteria

All titles identified by the search strategy were independently screened by two authors (Banghua Zhong and Wei Gao). Search results were compared, and disagreements were resolved by consensus. Abstracts of potentially relevant titles were then reviewed for eligibility, and full-length articles were selected for closer examination if there was a specific description on patients with SPN. The criteria for eligibility were as follows. First, any prospective or retrospective studies in English publication on patients with SPN only were included. Second, studies with evaluating the association between prognostic factors and the malignancy of postoperative SPN patients were included. Third, odds ratios (ORs) in case-control studies or relative risks in cohort studies were reported with the 95% confidence intervals (CIs) (or, if 95% CIs were not reported, the reported data were

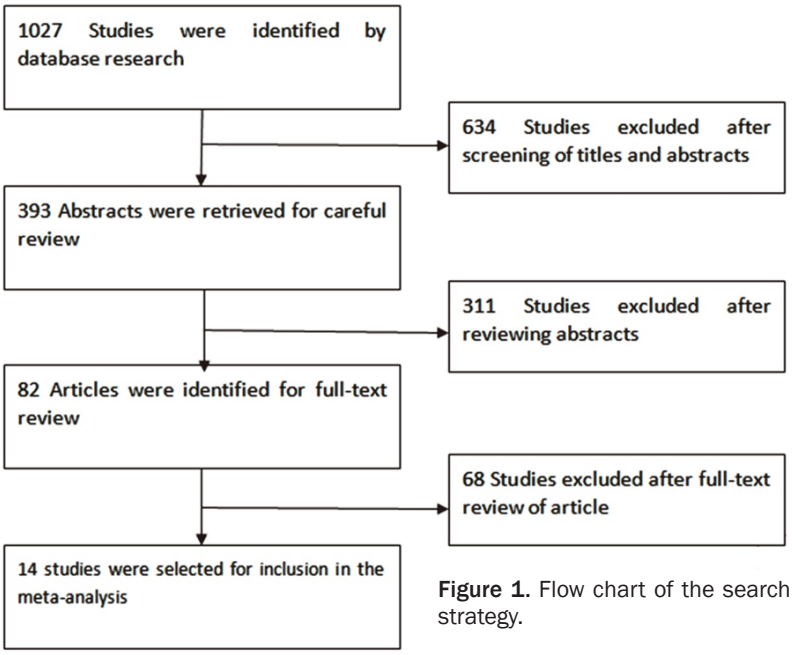

Figure 1. Flow chart of the search strategy.

Table 1. Characteristics of studies concluded

Referrance	Country	Year	Malignant/Total
Cai [8]	China	2011	17:33
Chuang [9]	Korea	2009	12:30
GOH [10]	Singapore	2007	9:16
Hokim [11]	Korea	2011	5:30
Hwang (child) [12]	Korea	2014	9:45
Jean [13]	U.S.	2010	9:45
Kang [3]	Korea	2006	11:33
Kim [5]	Korea	2014	17:106
Lee (adult and child) [14]	Korea	2008	10:62
Nakeeb [15]	Eygpt	2013	6:24
Yang [16]	China	2009	2:26
Yu [4]	China	2015	16:97
Yucai [17]	China	2014	35:116
Tang [18]	China	2015	24:100

sufficient to calculate them). Fourth, to avoid overlapping data that may result from duplications, only the articles with the largest sample size were included. Editorials, review articles or case series without association between risk factors and malignancy, duplicate publications and case reports were excluded. As is shown in the **Figure 1**.

Data extraction and management

All data were extracted onto a standardized form. The primary data extracted from each article included the first authorship, country of origin, year of publication (**Table 1**).

Assessment of risk of bias in included studies

Risk of bias across studies may be present, particularly with regard to publication bias. As the topic involves surgical procedures and outcomes, it is very likely that smaller-sample studies or those with negative outcomes may not be published in the literature. A funnel plot was created to assess publication bias.

Statistical analysis

If a specific factor was reported in at least three studies and supported by comparable methodologies, the odds ratios (OR) and 95% confidence intervals (CIs) were calculated to estimate the association between binary factors and malignancy of SPN. When mean values and SDs for a certain risk factor were provided, we calculated the mean differences (MDs) between patients with malignant SPN and with benign SPN. Depending on the presence or absence of significant heterogeneity, meta-analysis was conducted using the random-effects model or the fixed-effect model. Statistical heterogeneity of treatment effects between studies was formally tested with Cochran's Chi-squared statistics and with significance set at $P<0.10$. The I^2 statistic was used to quantify heterogeneity. All the individual outcomes were integrated with the meta-analysis software Review Manager Software 5.2 (Cochrane Collaborative, Oxford, United Kingdom).

Results

There were 14 eligible studies with the comparision of clinicopathlogical paremeter which were published from 2007 to 2015 (**Table 1**). One of these studies were from America, one from Africa, one from Australia, and eleven from Asia (six from Korea, five from China, one

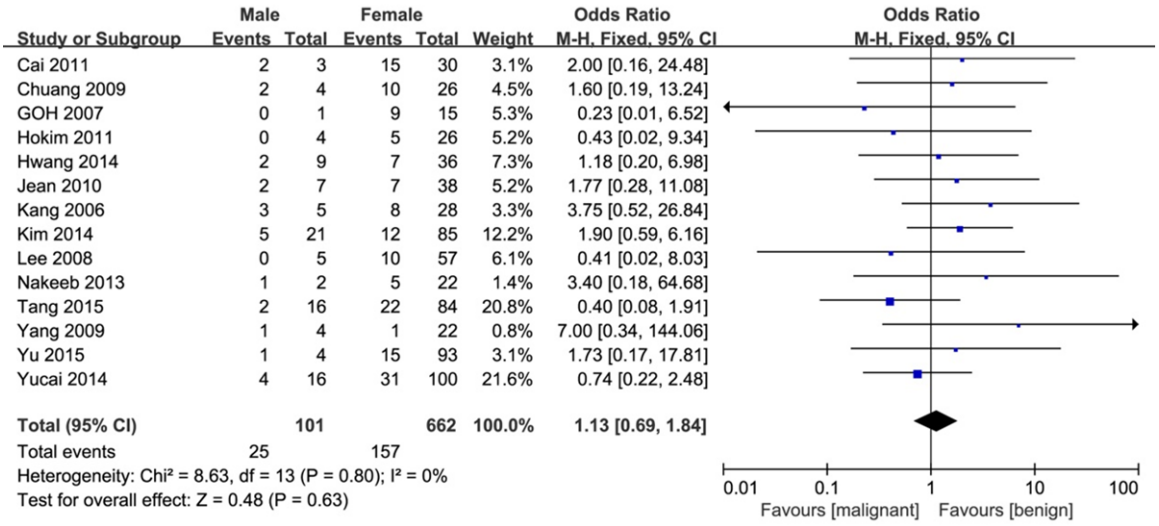

Figure 2. Forest plot for risk factors for malignancy of SPN categorized by male vs. female.

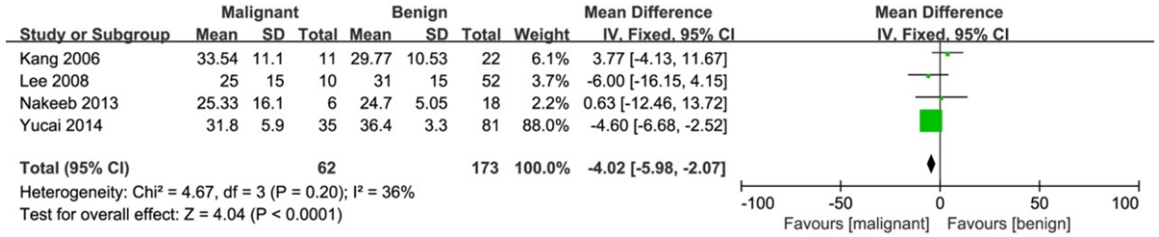

Figure 3. Forest plot for risk factors for malignancy of SPN categorized by age.

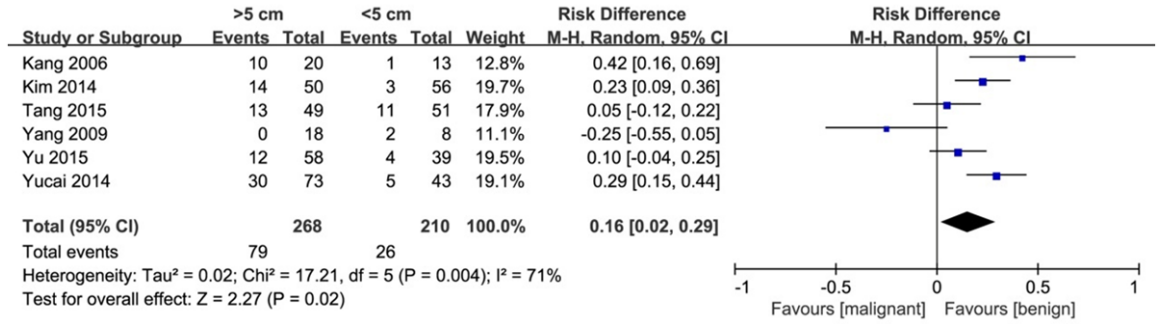

Figure 4. Forest plot for risk factors for malignancy of SPN categorized by tumor size in the dichotomous data type.

from Singapore). There were 763 patients in total and 101 patients of malignant SPN. There were 9 potential risk factors for malignant SPN identified for analysis.

Gender

With the statistical method of Mantel-Haenszel in the fixed-effect model, it showed no statistically significant difference in malignancy in the patients with male vs. female (OR = 1.09, 95% CI: 0.77-1.56) without heterogeneity (I^2 = 0%, P = 0.78) (**Figure 2**).

Age

With the statistical method of Inverse Variance in the fixed-effect model, it showed the statistically significant difference in malignancy in the patients about age (MD = -4.02, 95% CI: -5.98 to -2.07) without much heterogeneity (I^2 = 36%, P = 0.20) (**Figure 3**).

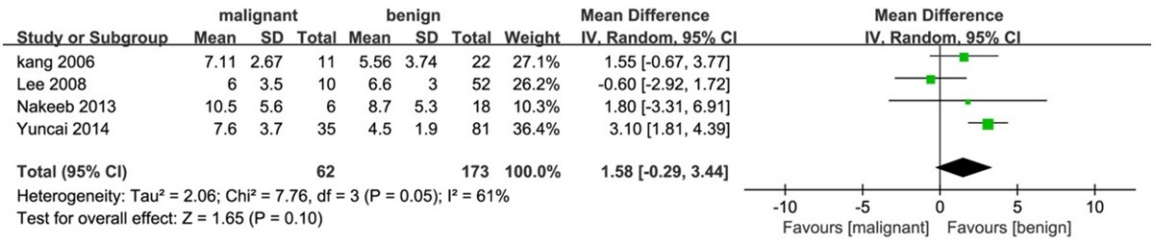

Figure 5. Forest plot for risk factors for malignancy of SPN categorized by tumor size in the continuous date type.

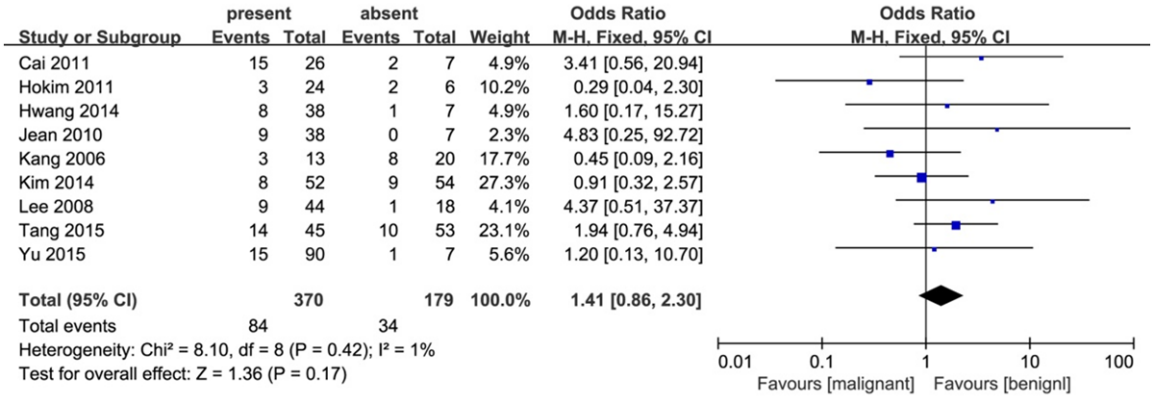

Figure 6. Forest plot for risk factors for malignancy of SPN categorized by symptom.

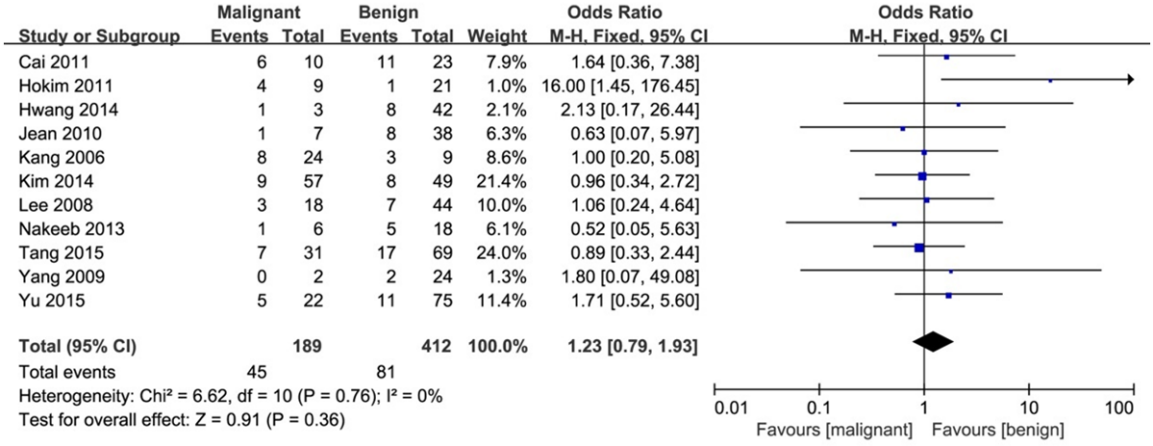

Figure 7. Forest plot for risk factors for malignancy of SPN categorized by calcification.

Tumor size

There were 2 meta-analyses to calculate the OR and MD respectively according to the dichotomous data type and continuous date type in the studies evaluated. In the dichotomous data type, tumor size were classified as two groups size >5 cm vs. size <5 cm. Analysis of the pooled data showed the risk factors of malignancy was significantly higher among the patients in the size >5 cm group (OR = 2.82, 95% CI: 1.13-7.01) with heterogeneity (I^2 = 61%, P = 0.03) using the statistical method of Mantel-Haenszel in the random-effect model (**Figure 4**).

In the continuous data type, the meta-analysis showed no statistically significant mean difference in malignancy in the patients about tumor diameter (MD = 1.58, 95% CI: -0.29 to 3.44)

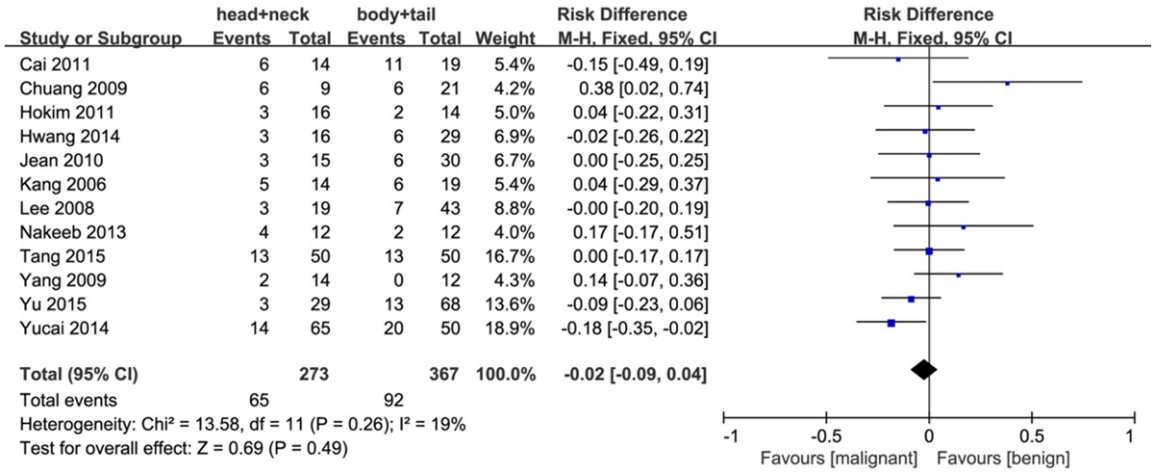

Figure 8. Forest plot for risk factors for malignancy of SPN categorized by tumor location.

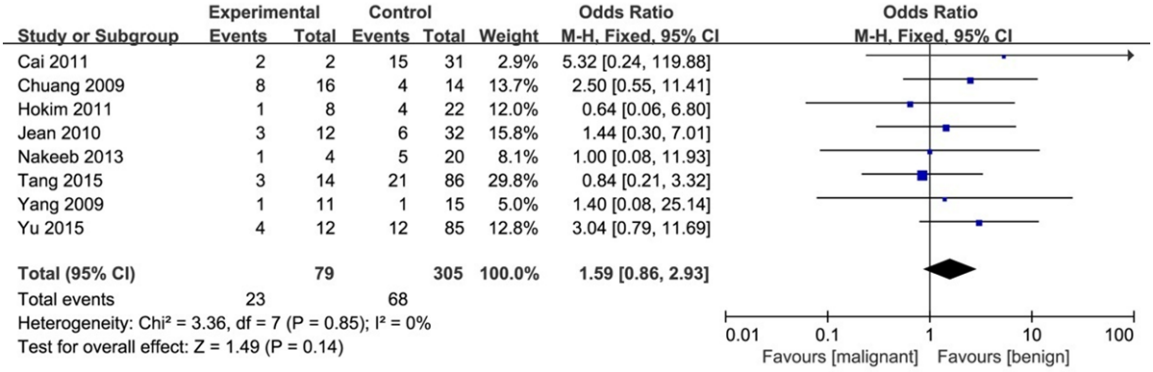

Figure 9. Forest plot for risk factors for malignancy of SPN categorized by tumor nature.

with heterogeneity (I^2 = 61%, P = 0.05) using the statistical method of Inverse Variance in the random-effect model (**Figure 5**). In the sensitivity analysis, removal of Lee study, there was significant mean difference (MD = 2.67, 95% CI: 1.57 to 3.76) without heterogeneity (I^2 = 0%, P = 0.47) (figure not shown).

Symptom

A meta-analysis showed no statistically significant difference in malignancy in the patients with present vs. absent (OR = 1.41, 95% CI: 0.86-2.30) without heterogeneity (I^2 = 1%, P = 0.42) using the statistical method of Mantel-Haenszel in the fixed-effect model (**Figure 6**).

Calcification

A meta-analysis showed no statistically significant difference in malignancy in the patients with present vs. absent (OR = 1.23, 95% CI: 0.79-1.93) without heterogeneity using the statistical method of Mantel-Haenszel in the fixed-effect model (I^2 = 0%, P = 0.76) (**Figure 7**).

Tumor location

The tumor location was defined as head+neck vs. body+tail. Comparisons of patients with head+neck vs. body+tail, there was no statistically significant difference between different tumor location (OR = 0.88, 95% CI: 0.60-1.28) without heterogeneity (I^2 = 4%, P = 0.41) using the statistical method of Mantel-Haenszel in the fixed-effect model (**Figure 8**).

Tumor nature

Tumor nature was defined as predominantly solid vs. predominantly cystic or mixed. The meta-analysis showed no statistically significant difference in malignancy in the patients about tumor nature (OR = 1.59, 95% CI: 0.86-

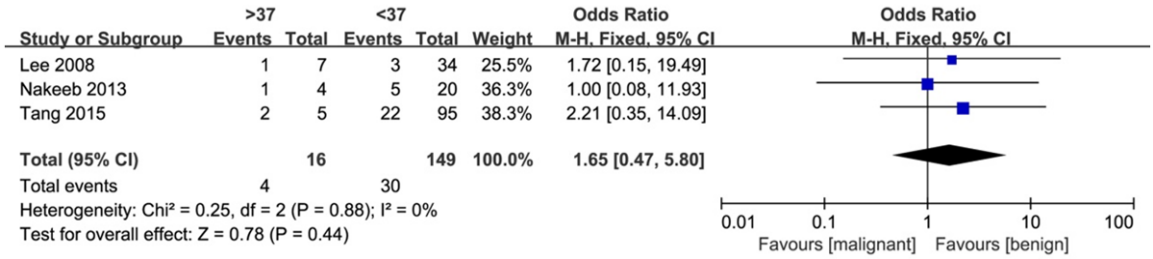

Figure 10. Forest plot for risk factors for malignancy of SPN categorized by tumor marker.

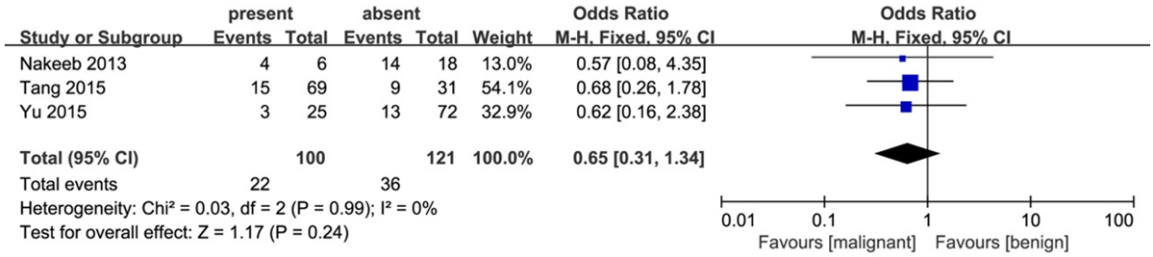

Figure 11. Forest plot for risk factors for malignancy of SPN categorized by tumor hemorrhage or necrosis.

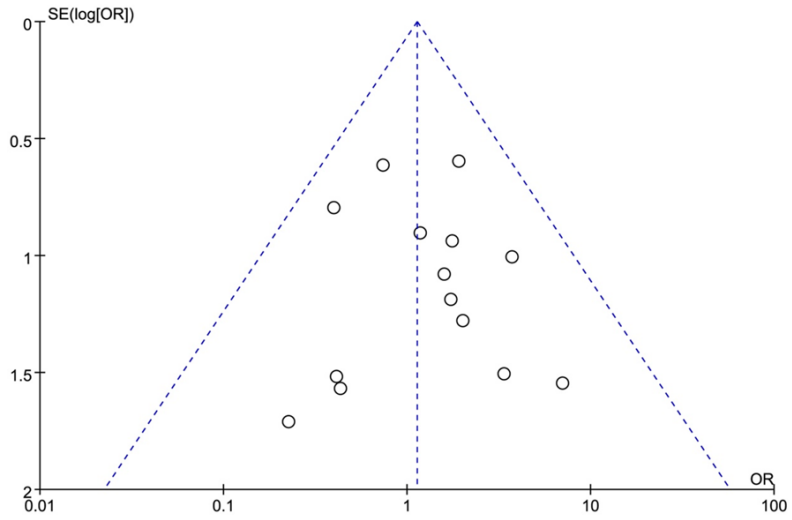

Figure 12. Funnel plot for risk factors for malignancy of SPN is symmetry.

2.93) without heterogeneity (I^2 = 0%, P = 0.85) using the statistical method of Mantel-Haenszel in the fixed-effect model (**Figure 9**).

Tumor marker CA199

The tumor marker CA199 was defined as elevated vs. normal. Comparisons of patients with elevated vs. normal, there was no statistically significant difference between different tumor marker (OR = 1.65, 95% CI: 0.47-5.80) without heterogeneity (I^2 = 0%, P = 0.88) using the statistical method of Mantel-Haenszel in the fixed-effect model (**Figure 10**).

Tumor hemorrhage or necrosis

A meta-analysis showed no statistically significant difference in malignancy in the patients with present vs. absent (OR = 0.65, 95% CI: 0.31-1.34) without heterogeneity (I^2 = 0%, P = 0.99) using the statistical method of Mantel-Haenszel in the fixed-effect model (**Figure 11**).

Sensitivity analyses

To test the strength of our results, we removed an individual study each time and calculated the pooled ORs of the rest of studies. No significant differences were observed between the corresponding results and the robust overall results (data not shown), except removal of Lee study leading to significant result in the continuous data type of the tumor diameter.

Publication bias

No obvious asymmetry was observed in the funnel plot of the meta-analysis evaluating the risk factors of malignancy in SPN (**Figure 12**).

Discussion

In the recent days, a relatively large case series of single center [19-25] or the multicenter [26] reporting SPN have tried to enrich the unsufficient clinical data in the current literature, but the results were limited to descriptive clinical analysis and failed to enclose the relation between clinicopathologic feature and potential malignancy. Many factors may affect malignancy of SPN, yet there still existed some controversy so far. The prevalence of malignancy in SPN patients should not be negligible, therefore malignancy-related variables involved in SPN patients' outcome are crucial. This study was the first meta-analysis of the literature on exploring the factors for SPN patients with malignancy. Included in our analysis were 14 unique studies from 2007 to 2017 with 763 patients. Data shows that age and tumor size were associated with a significantly increased risk of malignancy.

For the gender, the report of 34 cases [27] from Brazil regarded SPN were more aggressive in male patients. Men had a two-times higher incidence of metastases and a three-times higher death rate [28]. However, the similar tendency was not confirmative in our meta-analysis. There were no significant sex differences in histopathologic or immunohistochemical features of SPN [29, 30]. So maybe the report [27] was limited to the small sample case series to draw the conclusion.

For the age, although SPN had different clinical features in adults and children, children were not likely to appear malignant potential [14]. However, in our meta-analysis the younger age (in adults) could be related to the malignant potential. Only four studies were included, there would be more specific clinical data reporting age.

For the tumor size, there were several studies [3, 31] indicating that larger tumor size was related to malignancy, which is consistent with our meta-analysis. Moreover, tumor size is the significant clinical feature associated with metastatic disease and decreased disease-free survival [32]. Therefore, we suggest a more precise and aggressive resection of the SPN for larger tumors (>5 cm). Tumor (<5 cm) can be managed in conservative surgery such as enucleation or distal pancreatectomy preserving spleen. Especially, tumor locating in the head of pancreas more than 5 cm was likely proceeded with duodenum-preserving pancreatic head resection rather than enucleation [33]. In the sensitivity analysis, removal of Lee study led to significant result in the continuous data type of the tumor diameter. It was probable that Lee study covered the combination of data among both children and adults, children's clinical data will interfere with the overall results. In comparison with the adult population, children with SPN generally have a better prognosis and a different clinical feature [34].

Clinical presentation of the SPN is usually nonspecific. At the time of diagnosis, SPN may appear with a significantly enlarged size leading to abdominal pain or distention [35]. However, presence of symptom is not related to malignancy in our ananlysis.

Tumor location, tumor marker and presence of hemorrhage or necorsis were not associated with malignancy in our study, which has the similar results as the Tang [18].

There were some studies showing calcification [36] and tumor nature [12] were related to the malignancy which is not consistent with our meta-analysis.

Few studies reported other important risk factors, such as tumor capsule [9], Ki-67 [4] and peripancreatic lymphadenopathy [18], a meta-analysis of these factors could not be performed due to unsufficient data.

The malignancy incidence of SPN varies from 3.6 to 56% [37]. FDG-PET may help distinguish the malignant SPN from benign type, however no statistical analysis could be performed because the PET is not the routine examination [37, 38]. Maybe the accumulation of FDG in the imaging would be the new orientation for predictive factors of malignancy.

All the meta-analyses have limitations due to the quality of the primary studies. All studies in our meta-analysis were retrospective researches and limited number of cases. Some clinical data in the studies were not complete, and detailed data could not be required by contacting the relevant authors. We generated funnel plots for each variable. There were no obvious asymmetries. Based on our meta-analysis, we recommended establishment of standardized reporting guidelines for SPN.

In conclusion, malignant SPNs tended to be larger in diameter and younger in age than benign type. In particularly, larger tumor size may be a crucial factor for decision of aggressive resection.

Acknowledgements

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Disclosure of conflict of interest

None.

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