Evidence Based Case Report: Surgical Approach in Ameloblastoma with Odontogenic Keratocyst and Recurrence Rate

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Abstract

Ameloblastoma and odontogenic keratocyst are the major odontogenic neoplastic lesions. Ameloblastoma and odontogenic keratocyst are high reccurence. Treatment for ameloblastoma and odontogenic keratocyst is classified as conservative or radical approach. The purpose of the research to evaluate the surgical approach in treatment of ameloblastoma with odontogenic keratocyst and recurrence rate. A 36 years old woman with nasal obstruction in her right side for a year, blood stain rhinorea and facial swelling. Computerized tomography showed a isodens mass, non calsification, well defined margin, extending from nasal cavity to nasopharynx, bilateral maxillary sinus, zygoma area and bilateral buccal region. The mass was destructed nasal bone, bilateral zygomatic bone, bilateral alveolar maxillary bone and the wall of bilateral maxillary sinus. Pre-operative histological examination showed ameloblastoma with keratinizing odontogenic cyst (odontogenic keratocyst). Total maxillectomy with obturator was performed. Patient followed up for a year without recurrence. A PubMed database and ScienceDirect search were conducted. the search resulted in two useful articles were critically appraised. Result of the research that the risk of recurrence for ameloblastoma and odontogenic keratocyst following radical treatment was lower than conservative treatment. Radical approach can be a good choice for ameloblastoma with odontogenic keratocyst for prevent the recurrence. The patient should followed up regularly to observe recurrence.

Key Words: Ameloblastoma, Odontogenic Keratocyst, Radical Approach, Conservative Approach, Maxillectomy, Recurrence

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Introduction

Ameloblastoma, odontogenic keratocyst, dentigerous cyst, radicular cyst, and nasopalatine duct cyst are the major odontogenic neoplastic lesions in maxillomandibular regions. Odontogenic cysts and tumors constitute an important aspect of oral and maxillofacial pathology (Meng et al., 2019). Ameloblastoma is a tumor of odontogenic epithelial origin. It may arise from rests of dental lamina, from a developing enamel organ, from the epithelial lining of an odontogenic cyst, or from the basal cells of the oral mucosa (Kosmidou et al., 2020). Ameloblastoma is a benign, slow-growing, locally invasive tumor and high reccurance rate (Pitak-Arnnop et al., 2010). Ameloblastoma has a strong predilection to occur in the mandible than in the maxilla, with a mandible to maxilla ratio of 4:1. The ratio of male and female of 25 patients with ameloblastoma was 1,78:1 and 72% of ameloblastoma was located in the posterior area (Meng et al., 2019). Ameloblastoma appears most commonly in the third to fifth decades (Tranchina et al., 2012).

There are three classic varieties of ameloblastoma: solid/multicystic, unicystic and peripheral. Multicystic or solid type is prevalent in 86% of cases (Singh et al., 2014). Multicystic ameloblastoma is considered the most common variant and consists of a locally aggressive tumour that mainly affects the body and posterior portion of the mandible in patients in the third and fourth decades of life (de AC Almeida et al., 2016). The most common presenting symptom is a painless swelling. Other symptoms include, loosening of teeth, malocclusion and altered sensation in teeth and occasionally pain. Recurrence rates of ameloblastoma are reportedly as high as 15-25% after radical treatment and 75-90% after conservative treatment (de AC Almeida et al., 2016).

Odontogenic keratocyst (OKC), formerly known as keratocystic odontogenic tumor (KCOT), is a common odontogenic cyst that originates from the dental lamina or from its residue; occurring at any age, and is more common in men than women with a 2:1 ratio (Rahrotaban et al., 2018). Odontogenic keratocyst may present as solitary or multiple arising from remnants of the dental lamina. The occurrence of odontogenic keratocyst was found to be from the first to the ninth decades of life, with a peak incidence is however in the second and third decades. The mandible is mostly affected with the angle of mandible being a very common anatomical site. Involvement of the maxillary sinus by OKC is rare with <1% of cases reported in the literature (Sheethal et al., 2019). Because of the high proliferation rate, aggressiveness and recurrence rate of epithelial cells of this cyst, in 2005 WHO reclassified odontogenic keratocyst as benign intra-osseous neoplasia (Rahrotaban et al., 2018).

Treatment for ameloblastoma and odontogenic keratocyst is classified as conservative or radical. Conservative treatment includes marsupialization, enucleation, curettage, and surgical excision with peripheral osteotomy or other adjuvant therapy, such as cryotherapy or Carnoy's solution. Radical treatment consists of bone resection. In the mandible, resection can be completed through segmental osteotomy or a mandibulectomy, or can be marginal. In the maxilla, radical treatment comprises maxillectomy. Marsupialization can be considered as a definitive treatment modality for odontogenic keratocyst (de AC Almeida et al., 2016; Khan et al., 2019). Beside marsupialization alone, marsupialization followed by enucleation or curetage and excision of the lesion (resection) as a treatment for large and extensive odontogenic keratocyst (Gupta et al., 2016). This paper was aimed to evaluate the comparison of radical or conservative approach in treatment of ameloblastoma with odontogenic keratocyst and recurrence rate.

Case Report

A 36 years old woman was referred to Ear, Nose and Throat Clinic, Wahidin Sudirohusodo Hospital, Makassar, South Sulawesi, in May 2019. The chief complaint was an obstruction in her right nasal cavity for a year. She also complaint a blood stain rhinorea, a swelling in her palatum, difficult to open her mouth, errupted molar in a right side and swelling in her right cheek. The swelling was getting bigger and painless. The patient's picture showed in figure 1.

Figure 1. Patient's picture

Physical examination showed a swelling in her right bucal region. The swelling was hard, painless to palpation,unclear margin, colorish same with the surround and extending to her right nasal cavity. There were a trismus and no vision disturbance. From anterior rhinoscopy, there was mass in her right nasal cavity. It was hard and easy to bleed. The patient was consulted to opthalmologist and there was not any problem in vision or eyes.

Computerized tomography showed a isodens mass (45 HU), non calsification, well defined margin, size 11,6x7,7x10,5 centimeters, extending from nasal cavity to nasopharynx, bilateral maxillary sinus, zygoma area and bilateral buccal region expecially in right region. The mass was destructed nasal bone, bilateral (expecially right) zygomatic bone, bilateral

alveolar maxillary bone and the wall of bilateral maxillary sinus (Figure 2).

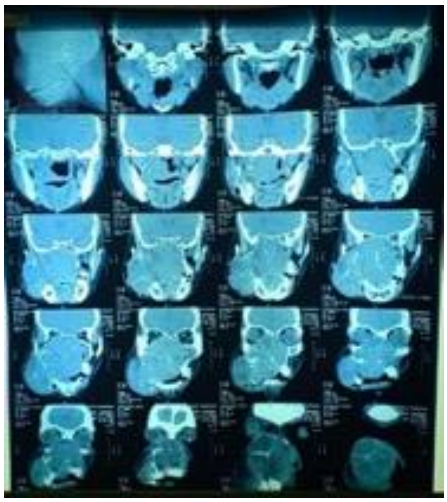

Figure 2.Patient's computerized tomography

Pre-operative histological examination showed the features of a ameloblastoma with keratinizing odontogenic cyst (odontogenic keratocyst).

Total right maxillectomy (without orbital exenteration) was performed with Weber Ferguson incision, from inferior right palpebra to the posterolateral maxillary bone until inferior zygomatic bone. The mass was found in right maxillary bone and extended to superior canine fossa. It was solid, ireguler, easy to bleed, and unclear margin. The deficiency of palate was packed with obturator. It was made by prostodontist.

Histological examination of the lesion showed a ameloblastoma with keratinizing odontogenic cyst (odontogenic keratocyst). Based on clinical presentation, computerized tomography and histopathology, the type of ameloblastoma is multicystic or solid ameloblastoma. The patient was followed up a year after operation and there was not a recurrence.

Clinical Question

Problem : Female, 36 years old with multicystic or solid ameloblastoma and odontogenic keratocyst

Intervention : Radical approach (total maxillectomy)

Comparison : Conservative approach

Outcome : Recurrence

"Does radical approach prevent the recurrence of multicystic ameloblastoma with odontogenic keratocyst than conservative approach?"

Methods

Journal was searched with PubMed via PICO with Spelling Checker and ScienceDirect in September, 9th, 2020. The inclusion criterias were free full text, published in the last 10 years, and in English, the exclusion criteria was research in animal. The strategy of journal search was list in table 1.

Table 1. Search Strategy

Database	Search Strategy	Articles
PubMed	P : ameloblastoma or odontogenic keratocyst I : radical C : conservative O : recurrence	59
Science Direct	ameloblastoma and odontogenic keratocyst AND radical AND conservative AND recurrence	133

Results and Discussion

From PubMed via PICO with Spelling Checker, we found 59 articles, from these 32 articles were published in the last 10 years and from ScienceDirect, we found 133 articles and 57 articles were published in the last 10 years.

We choose 2 articles that related with out aim and goes to the critical appraisal step. The articles were " Radical vs conservative treatment of intraosseus ameloblastoma: Systemic review and meta-analysis" written by Khan et al. (2019) and article written by Al-Moraissi, et al with title "What surgical treatment has the lowest recurrence rate following the management of keratocystic odontogenic tumor?: A large systematic review and meta-analysis"

The flowchart of journal result was showed in figure 3.

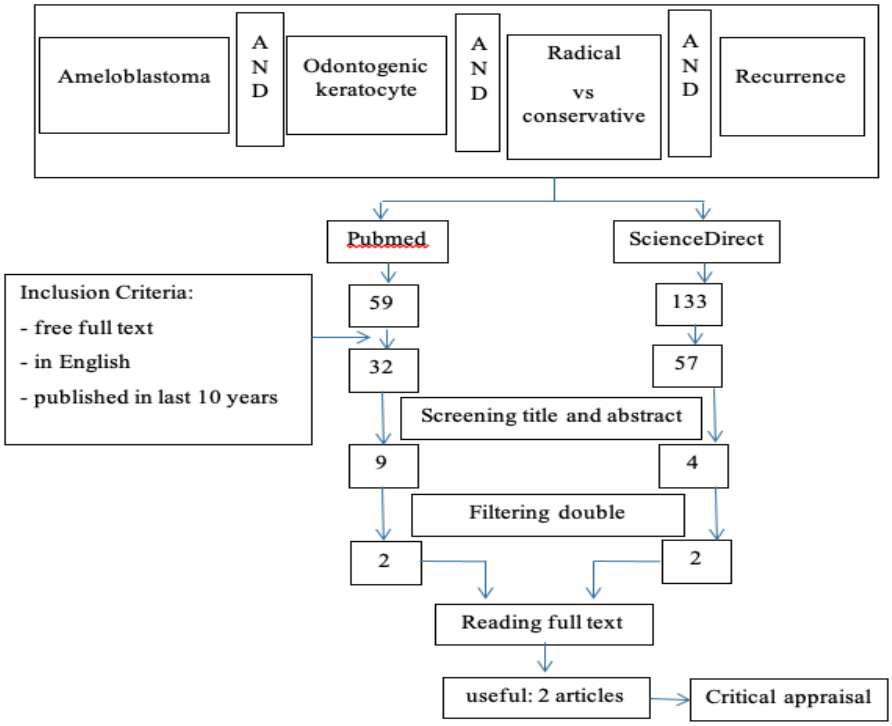

Figure 3. Flowchart of Search strategy

The result of critical appraisal of the systemic review and meta-analysis is shown on table 2. We conclude that the study is valid, important, and applicable to our patient with level of evidence Ia.

Table 2. Critical Appraisal

Critical appraisal	Hendra, et al	Al-Moraissi, et al
Validity		
What question (PICO) did the systematic review address?	Yes	Yes
Is it unlikely that important, relevant studies were missed?	Yes	Yes
Were the criteria used to select articles for inclusion appropriate?	Yes	Yes
Were the included studies sufficiently valid for the type of question asked?	Yes	Yes
Were the result similar from study to study?	Yes	Yes
Importancy		
What were the results?	For solid or multicystic ameloblastoma, the estimate combined relative risk was 0.35 (95% CI, 0.23-0.52; ρ < 0.00001) and for uncystic ameloblastoma, the estimate relative	A weighted event rate of the pooled recurrence in the enucleation group was 23.1% (95% CI = 17.3%, 30.2%). An overall weighted event rate of enucleation with a peripheral osteotomy was 17.4% (95% CI = 11.2%-26.1%). The overall weighted event rate of enucleation plus Carnoy's solution was 11.5% (96% CI = 6.8%,

	risk was 0.32 (95% CI, 0.12-0.82; ρ = 0.02)	18.8%). The overall weighted pooled event rate of enucleation plus liquid nitrogen cryotherapy was 14.5% (95% CI = 5.1%, 34.9%). The overall weighted pooled event rate of marsupialization or decompression alone was 32.3% 995% CI = 16.9%, 52.8%). The overall weighted pooled event rate of decompression followed by residual cystectomy was 14.6% (95% CI = 9.2%, 22.3%) and for resections, the overall weighted pooled event rate was 8.4% (95% CI = 3.5%, 18.8%)
How precise were the results?	Precise	Precise
Applicability		
Can he results be applied to my patient care?	Yes	Yes
Were all clincally important outcomes considered?	Yes	Yes
Are the benefits worth the harm and the cost?	Yes	Yes

We appraised these meta-analysis study by Hendra, et al and Al-Moraissi, et al for its validity, importance, and applicability.

Hendra et al performed a systematic review and meta-analysis to assess the recurrence rates of radical and conservative treatment approaches of solid or multicystic and unicystic ameloblastoma (Hendra et al., 2019). Hendra, et al found the pooled recurrence rate for solid ameloblastomas was 8% after radical, and 41% after conservative treatment. For unicystic ameloblastomas, these values were 3% and 21%. Hendra et al pooled recurrence rates in solid or multicystic ameloblastomas compared to unicystic ameloblastomas were higher following conservative as well as following radical treatment. This may indicate that the solid or multicystic type behaves more aggressive than the unicystic type. This study has several limitations. For examples, they could not consider the quality of life in the included studies and could not assess when recurrence occured after treatment of ameloblastoma (Hendra et al., 2019).

A systematic review and meta-analysis by Al-Moraissi, et al aimed to seek the best surgical treatment with the least recurrence rate. The weighted RR for various treatment techniques was as follows: enucleation alone (23.1%), enucleation with curettage (17.4%), enucleation and Carnoy's solution (11.5%), enucleation plus liquid nitrogen cryotherapy (14.5%), marsupialization alone (32.3%), decompression followed by residual cystectomy (14.6%), and resection (8.4%). They conclude that resection is the option for obtaining the lowest recurrence with odontogenic keratocyst, resections should be reserved for multiple recurrent lesions and possibly syndromic cases. Enucleation with Carnoy's solution or cryotherapy is the first-line treatment for primary odontogenic keratocyst tumor. Marsupialization (where indicated) must be followed by secondary cystectomy to minimize recurrence (Al-Moraissi et al., 2017). Ameloblastoma is a slow growing, locally aggressive tumor4. Sinonasal ameloblastoma are rare tumors of the sinonasal tract that arise from sinonasal epithelium. The sinonasal tract and oral cavity communicate, in fact, until closure of the palatine shelves (Tranchina et al., 2012). Clinical presentation of sinonasal ameloblastoma commonly includes nasal obstruction, sinusitis and/or epistaxis (Barrena et al., 2019). Less

common presenting signs and symptomps include facial swelling, headache and tearing (Barrena et al., 2019).

A systematic review of the literature published in 2011 by MacDonald-Jankowski showed that patients of odontogenic keratocyst in East Asian origin may present symptoms early, characterised by swelling and pain, while discharge and numbness of the inferior alveolar nerve are described more frequently (Borghesi et al., 2018). In our case, the patient was complain nasal obstruction, painless facial swelling, blood stain rhinorea. Preoperative assessment is important for planning treatment and management, we did a pre-operative biopsy and the result is ameloblastoma with keratinizing odontogenic cyst (odontogenic keratocyst). The occurrence of ameloblastoma and odontogenic keratocyst simultaneously in one patient is very rarely documented in English literature (Gupta et al., 2016; Firth et al., 2020).

From clinical presentation and radiologic findings we conclude that the type of ameloblastoma was mulicystic or solid. Based on clinical presentation, radiologic and histopathologic finding, we concluded to performed a radical approach (total maxillectomy without orbital exenteration). The defect of palate was covered by obturator. A year after operation, there is no recurrence. The recurrence is not only depand on the treatment but several factors like age, clinical presentation, life syle and etc.

Ameloblastomas and odontogenic keratocyst is a benign, rare and high recurrence odontogenic lesions. Radical approach can be a good choice for ameloblastoma withodontogenic keratocyst for prevent the recurrence. The patient should followed up regularly to observe recurrence.

References

1. Al-Moraissi, E. A., Dahan, A. A., Alwadeai, M. S., Oginni, F. O., Al-Jamali, J. M., Alkhutari, A. S., ... & Al-Sanabani, J. S. (2017). What surgical treatment has the lowest recurrence rate following the management of keratocystic odontogenic tumor?: A large systematic review and meta-analysis. *Journal of Cranio-Maxillofacial Surgery*, *45*(1), 131-144.
2. Barrena, B. G., Phillips, B. J., Moran, K. J., & Betz, S. J. (2019). Sinonasal ameloblastoma. *Head and Neck Pathology*, *13*(2), 247-250.
3. Borghesi, A., Nardi, C., Giannitto, C., Tironi, A., Maroldi, R., Di Bartolomeo, F., & Preda, L. (2018). Odontogenic keratocyst: imaging features of a benign lesion with an aggressive behaviour. *Insights into imaging*, *9*(5), 883-897.
4. de AC Almeida, R., Andrade, E. D. S., Barbalho, J. C., Vajgel, A., & Vasconcelos, B. D. E. (2016). Recurrence rate following treatment for primary multicystic ameloblastoma: systematic review and meta-analysis. *International journal of oral and maxillofacial surgery*, *45*(3), 359-367.
5. Firth, N., Alsarraf, A., Vujcich, N., & Kujan, O. (2020). Synchronous occurrence of odontogenic keratocyst and ameloblastoma: A case report and review of the literature. *Clinics and practice*, *10*(2), 1205..
6. Gupta, R. K., Dugal, A. G., Pawar, S. R., Khandelwal, S. G., & Iyengar, A. (2016). A rare simultaneous occurrence of odontogenic keratocyst and unicystic ameloblastoma in mandible: a case report. *Journal of Clinical and Diagnostic Research: JCDR*, *10*(8), ZD01.
7. Hendra, F. N., Natsir Kalla, D. S., Van Cann, E. M., de Vet, H. C., Helder, M. N., & Forouzanfar, T. (2019). Radical vs conservative treatment of intraosseous ameloblastoma: Systematic review and meta-analysis. *Oral diseases*, *25*(7), 1683-1696.
8. Khan, A. A., Al Qahtani, S., Dawasaz, A. A., Saquib, S. A., Asif, S. M., Ishfaq, M., ... & Ibrahim, M. (2019). Management of an extensive odontogenic keratocyst: A rare case report with 10-year follow-up. *Medicine*, *98*(51).
9. Kosmidou, P., Angelis, S., Papagianni, E., Kokkevi, I., & Filippou, D. (2020). Ameloblastoma on the maxillary sinus: cause of unilateral nasal obstruction. *Cureus*, *12*(1).

10. Meng, Y., Zhao, Y. N., Zhang, Y. Q., Liu, D. G., & Gao, Y. (2019). Three-dimensional radiographic features of ameloblastoma and cystic lesions in the maxilla. *Dentomaxillofacial Radiology*, *48*(6), 20190066.
11. Pitak-Arnnop, P., Chaine, A., Dhanuthai, K., Bertrand, J. C., & Bertolus, C. (2010). Unicystic ameloblastoma of the maxillary sinus: pitfalls of diagnosis and management. *Hippokratia*, *14*(3), 217.
12. Rahrotaban, S., Nikparto, N., Yousefizadeh, P., Kharazifard, M. J., & Derakhshan, S. (2018). Fascin Expression in Ameloblastoma, Odontogenic Keratocyst and Dentigerous Cyst. *Turk Patoloji Derg*.
13. Sheethal, H. S., Rao, K., Umadevi, H. S., & Chauhan, K. (2019). Odontogenic keratocyst arising in the maxillary sinus: A rare case report. *Journal of Oral and Maxillofacial Pathology: JOMFP*, *23*(Suppl 1), 74.
14. Singh, M., Shah, A., Bhattacharya, A., Raman, R., Ranganatha, N., & Prakash, P. (2014). Treatment algorithm for ameloblastoma. *Case reports in dentistry*, *2014*.
15. Tranchina, M. G., Amico, P., Galia, A., Emmanuele, C., Saita, V., & Fraggetta, F. (2012). Ameloblastoma of the sinonasal tract: report of a case with clinicopathologic considerations. *Case reports in pathology*, *2012*.