Ameloblastoma with Infratemporal Extension: A Review of the Literature

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Abstract

Ameloblastomas are benign tumors of odontogenic epithelium. They are locally aggressive with the tendency to recur, and sometimes with metastatic behavior. Recurrences often happen due to incomplete treatment and they can occur at difficult sites such as temporal and infratemporal fossa. Recurrences in the temporal area are very rare and are related to the type of primary treatment.

***Aim**: This literature review aims to answer the question on how common recurrent ameloblastoma extends to the infratemporal fossa and how this is related to the site of the primary lesion.*

***Materials and methods**: Web search for case reports, and case series of ameloblatoma with temporal, infratemporal extension, published in the English literature were carried out. Search results were further scrutinised for age, sex, location of lesion, histology, treatment modalities, and recurrence, following the adopted treatment modalities and treatment outcome.*

***Result:** A total 15 full length articles were included in this study. Twelve were case reports and three were case series. Of 28 patients with ameloblastoma in the articles, only 22 were recorded to have presented with ameloblastoma with infratemporal or temporal fossa involvement. All the cases of ameloblastoma involving the infratemporal/temporal fossa were recurrent tumors and the average time from first surgical intervention to recurrence was 11.36 years. Most of the primary cases were seen in the mandible (73%) with the body/ramus region being the commonest location. Only five cases were reported to be primarily maxillary ameloblastoma.*

***Conclusion**; This review has shown that temporal/infratemporal extension of ameloblastoma occurs commonly with recurrent lesions, although the overall reported incidence is relatively low. Aggressive primary tumor resection, especially for extensive mandibular lesions, may be key to preventing this tumor extension.*

***Keywords**: Ameloblastoma, temporal, infratemporal extension*

Introduction

Ameloblastomas are odontogenic lesions characterised by local invasiveness and the potential for direct involvement of vital structures with high tendency for recurrence, leading to extensive local morbidity and mortality. Ameloblastoma is the second most common odontogenic tumor of the jaws. It commonly occurs in the mandible and in the third to fifth decades of life.[1–9] Eighty percentage of ameloblastomas arise in the mandible, it infrequently involves the maxilla[2,7]. Only about 5 to 20% occur in the maxillary bone, with majority of these occuring in the molar region.[10,11] Some authors reported no gender predilection,[4,12,13] some others reported male predilection,[1,2] while others documented female predilection.[3]

Due to its tendency to cause extensive destruction of jaw bones, various treatment modalities of "conservative" and "radical" surgery have been described.[14,15] Conservative surgical approaches may be favored due to benign histology, however, these treatment modalities have very high recurrence rates (90% for mandibular tumors, 100% for maxillary tumors).[9,15] Recurrences often occur due to incomplete treatment and they can occur at difficult sites such as temporal and infratemporal fossa, orbit, anterior cranial base, paranasal sinuses, etc.[7,16-19] Recurrences in the temporal area are very rare and are related to the type of primary treatment. Most of the studies done on the temporal and/or intra cranial extension of ameloblastoma are mostly case reports and case series with little or no reviews of cases so far published. This literature review therefore aims to answer the question on how common recurrent ameloblastoma extends to the infratemporal fossa, and how this is related to the site of the primary lesion.

Materials and methods

We conducted systematic searches for published articles in PubMed (NLM), Cochrane, Ovid Medline, and OpenGrey databases up till December 2021 using the keywords: "ameloblastoma," "temporal," and "infrate-mporal extension." Additional searches for relevant studies were done via the following methods: hand-search of the reference section of eligible studies and purposeful Google Scholar searches. Only articles written in English or with English language translations were considered for the review. Both authors independently screened the titles and abstracts (when available) of all reports identified through electronic searches. The search was designed to be sensitive to include all available studies. For studies appearing to meet the inclusion criteria, or for which there was insufficient data in the title and abstract to make a clear decision, we obtained the full report. The full reports were also independently assessed by the two authors to establish whether the publication met the inclusion criteria or not. Disagreements were resolved through discussion between the two authors.

This search returned 37 articles in PubMed and 207 articles in PubMed Central. The initial

screening process resulted in 29 articles and these articles were retrieved and reviewed for relevance of content by the two authors (OAO and AAA). A total of 16 full articles were included in the final list for review (Table 1).

Data retrieved from search results included number of patients, age, gender, location of lesion and histology, treatment modalities carried out, any recurrence following the adopted treatment modalities, and treatment outcomes. Furthermore, other odontogenic tumours such as KCOT, ameloblastic fibroma, adenomatoid odontogenic tumour, etc. were excluded. Articles with cases more than two were adopted as case series.

Result

Out of the 15 papers found in the literature, 12 were case reports and 3 were case series. The total number of patients with ameloblastoma in the review was 28. However, only 22 were recorded to have presented with ameloblastoma with infratemporal or temporal fossa involvement and reviewed for this study. Sixty four percent (n = 14/22) were females, and their ages ranged between 18-73 years (mean = 43.10, SD ±17.39). All the cases of ameloblastoma involving the infratemporal/temporal fossa were recurrent tumors and the average time from first surgical intervention to recurrent lesion/involving the infratemporal/temporal fossa was 11.36 years. Seventy-three percent of the cases with infratemporal/temporal extension were found in the mandible (n = 16/22), with body/ramus region being the commonest location. Only 5 cases were reported to be primarily maxillary ameloblastoma.

Table 1. Case Series and Case Reports of Ameloblastoma and Temporal/Infratemporal Extension

Author	Title	Type of report	No of patients	Primary location	Secondary location	No of patients with infratemporal/temporal extension	Initial treatment	Time to temporal involvement	Treatment outcome	Age	Sex	Remarks
Zwahlen et al., 2002[11]	Maxillary ameloblastomas: a review of the literature and of a 15-year database	CS	5	Maxilla	maxilla	1	resection	NA	6yrs follow up	26	F	Ameloblastoma?
				Maxilla	ethmoid Sphenoid			NA		33	F	
				Maxilla	temporal	1		0.17		73	F	
				Maxilla				NA		42	M	
				Maxilla				NA		44	M	
Weiss et al., 1985[20]	Maxillary Ameloblastoma with Orbital Invasion A	CR	1	Maxilla	infratempo	1	resection	5	6 yrs and died same yr	72	M	follicular

	Clinicopathologic Study				ral and sphenoidal							
To et al., 2002[18]	Recurrent Ameloblastoma Presenting in the Temporal Fossa	CR	1	Mandible	temporal	1	curettage, resection	25	2.5 yrs follow--up	18	F	Ameloblastoma?
Al-Bayaty et al., 2002[17]	Soft Tissue Recurrence of a Mandibular Ameloblastoma Causing Facial Deformity in the Temporal Region: Case Report	CR	1	Mandible	temporal	1	resection	4	tumour free 2yrs follow-up	32	F	follicular
Faras et al., 2016[21]	Multi-recurrent invasive ameloblastoma: A surgical challenge	CR	1	Mandible	infratemporal	1	repeated resection	23	NA	56	F	follicular

Sharma et al., 2009[22]	Recurrent Unicystic Ameloblastoma of the Infratemporal and Temporal Fossa	CR	1	Mandible	temporal	1	enucleation and later resection	2.6	NA	20	F	follicular
Auluck et al., 2007[16]	Recurrent ameloblastoma of the infratemporal fossa: diagnostic implications and a review of the literature	CR	1	Mandible	infratemporal	1	resection	6	NA	44	F	follicular
Ferretti et al., 2000[23]	Recurrent Ameloblastoma Report of 2 Cases	CR	1	Mandible	temporal	1	resection	1.5	2 yrs tumour free	50	M	ameloblastoma
			1	Mandible	temporal	1	resection	25	3 yrs tumour free	42	M	ameloblastoma

Scaccia et al., 1991[24]	Maxillary Ameloblastoma Case Report	CS	1	Maxilla	ethmoidal , sphenoidal, infra temporal and intracranial	1	resection	NA	2 yrs follow-up	16	F	Ameloblastoma
			1	Maxilla	ethmoidal, sphenoidal,		resection	NA	2 yrs. follow-up	66	M	
			1	Maxilla	infra temporal and intracranial			17	Recurrence after 2 yrs	53	M	
			1	Maxilla	infra temporal and intracranial , ethmoidal, sphenoidal,	1	resection	2	2 yrs tumour free	36	F	Ameloblastoma
Luc et al., 1988[25]	Late Ioco-regional recurrences after radical resection for mandibular	CS	5	mandible	Maxilla	1	Resections	NA	Recurrence	51	F	Ameloblastoma

	ameloblastoma											
				mandible	Infratemporal	1	Resection	29	18 months	49	F	Ameloblastoma
				mandible	maxilla	1	Resections	NA	Recurrence after 2 yrs	46	M	Ameloblastoma
				Mandibular angle	infratemporal	1	hemimandibulectomy including the condyle	8	Recurrences and resections	67	M	Ameloblastoma
				Mandibular angle	temporal	1	hemimandibulectomy including the condyle and coronoid process	29	Recurrences and resections ,1 yr tumour free	50	F	Ameloblastoma
Aramanadka et al., 2018[26]	Recurrent Ameloblastoma: A Surgical Challenge	CR	1	Mandible		1	Resection	NA	2 yrs follow-up	56	M	Follicular Amelolastoma

			1	Mandible	Infratemporal		Hemimandibulectomy	6	NA	45	M	Follocular Ameloblastoma
Vaishampayan et al., 2014[4]	Recurrent ameloblastoma in temporal fossa: A diagnostic dilemma	CR	1	Mandible	temporal	1	resection, hemimandibulectomy	5	tumour free in 1.5 yrs	32	F	Ameloblastoma
Phillips et al., 1992[27]	Ameloblastoma of the Mandible With Intracranial Metastasis A Case Study	CR	1	mandible	temporal and intracranial	1	resections	13	NA	65	M	Ameloblastoma
Oka et al., 1986[7]	Mandibular ameloblastoma with intracranial extension and distant metastasis	CR	1	mandible	temporal and intracranial and femur	1	resections	19	femur mets chemo recurrence and died 43 yrs later	25	M	ameloblastoma

Rauso et al., 2010[12]	Recurrence of Ameloblastoma in Temporal Area: Primary Treatment Influences Recurrence Rate	CR	1	mandible	temporal fossa	1	enucleation and currettage	3	5 yrs follow up tumor free	29	F	Acanthomatous ameloblastoma

CR- case report; CS – case series

Discussion

In this review, 22 out of 28 patients were recorded to have presented with ameloblastoma with infratemporal or temporal fossa involvement. The initial site of involvement for majority was the mandible (73 percent) with the mandibular body and ramus being the most affected. The increased prevalence of infratemporal and temporal involvement of mandibular lesions compared to maxillary lesions could be attributed to higher prevalence of ameloblastoma in the mandible than in maxilla as reported in the literature.

Furthermore, 12 of the articles selected for this study were case studies while 3 articles accounted for case series with no cohort study. This could be due to rarity of this ameloblastoma with temporal and intra cranial extension. All reported cases involving the infratemporal/ temporal fossa were recurrent tumors and the average time from first surgical intervention to recurrent lesion involving the infratemporal/ temporal fossa was 11.36 years. Recurrence may be attributed to factors such as inadequate tumor removal, "seeding", aggressive histology, and spread along the muscle attachment.[17,23] Treatment of recurrence often mandates extensive ablative and reconstructive surgery with inherent morbidity, even in expert hands.[4,16] Recurrences of ameloblastoma often occur at difficult sites, and has been documented to recur in sites such as temporal and infratemporal fossa, orbit, anterior cranial base, paranasal sinuses etc.[16-19,11, 28, 29]

Due to the tendency of ameloblastoma to cause extensive destruction of jaw bones, various treatment modalities of conservative" and "radical" surgery have been described.[28] Other treatments described in literature include electrocautery, cryosurgery, chemotherapy, and radiotherapy.[3,7,13] Conservative surgical approach has been reported to have very high recurrence rates (90% for mandibular tumors, 100% for maxillary tumors).[4] The gold standard of care for ameloblastoma is complete surgical excision; aggressive surgical resection is advocated in patients with maxillary ameloblastoma to ensure recurrence-free outcome.[13] Although some authors have reported successful results with radiotherapy,[30,31] its use is however considered more in inoperable cases, primarily in the posterior maxilla.[31] Furthermore, chemotherapy as treatment modality has also been employed for inoperable lesions.[3] It is important to know that spread of the lesion from the infratemporal fossa and temporal region to adjacent structures to involve the pterygopalatine fossa or maxillary sinus, the skull base, and into the intracranial cavity or orbit makes radical surgical treatment more difficult.[16] Nastri et al[3] reported preoperative radiographic evidence of tumour in all of the cases in which surgical treatment failed to control the tumour, suggesting residual lesion. Therefore, early and aggressive surgical treatment is key in the management of ameloblastoma.

Treatment of maxillary ameloblastoma is inherently more difficult compared to its mandibular counterpart.[13] This is reported to be

due to the insidious nature of the lesion within the thin bones and hollow spaces of the midfacial bones, as the tumor easily spreads to the skull base, and, occasionally, may extend into orbit and/or the intracranial cavity by destroying the bones.[4] Numerous surgical approaches have been employed to access the infratemporal region, some of them being the coronal,[23] trans-oral, trans nasal, trans palatine, trans zygomatic, trans cervical, and extended maxillectomy approach.[16,26] Others include subtemporal epidural approach, and combined transcranial and transcervical approach.[32] The surgical approach to the lesion is often determined by clinical presentation, extent and location, as well as histopathological findings.[16] In addition, involvement of adjacent tissues requires collaborative surgical care[13] that would be provided by the oral and maxillofacial surgeons, otolaryngologists, plastic and reconstructive surgeons, ophthalmologists, and neurosurgeons.

This review has shown that temporal/infra-temporal extension of ameloblastoma occurs commonly with recurrent lesions, although the overall reported incidence is relatively low. Aggressive primary tumor resection, especially for extensive mandibular lesions may be key to preventing this tumor extension.

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