

Survey of opinions on the management of pleomorphic adenoma among United Kingdom oral and maxillofacial surgeons

Büyük Britanya'daki oral ve maksillofasyal cerrahlar arasında pleomorfik adenomların tedavisi ile ilgili anket araştırması

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Objectives: This study aims to highlight the current points of view regarding the management of pleomorphic adenomas among oral and maxillofacial surgeons in the United Kingdom.

Patients and Methods: A questionnaire was drafted and sent to all the consultant members of the British Society of Oral Maxillofacial Surgery (n=263). The survey evaluated the surgical experience of the surgeon, the preferred surgical treatment of a newly diagnosed 3 cm diameter pleomorphic adenoma in the superficial lobe of the parotid gland in an adult and a child, and the treatment options of pleomorphic salivary adenoma (PSA) with the four following scenarios: (i) PSA with complete excision or incomplete excision; (ii) PSA with carcinoma in situ (atypical pleomorphic adenoma) with complete excision or incomplete excision; (iii) PSA with non-invasive malignant transformation with complete or incomplete excision; and (iv) carcinoma in PSA with complete or incomplete excision. The initial response was low so two reminders were sent to increase the rate of response.

Results: The final response rate was 67%. The results showed that there was no significant difference in the opinions of the respondents regarding treatment of pleomorphic adenoma in both adults and children. Seventy-five (56.4%) of 133 surgeons were treated the multinodular recurrence in older patients with surgery combined with radiotherapy. One-hundred and five (78.9%) surgeons were treated older patients with solitary recurrence with surgery alone. There was a consensus on complete excision for the treatment of carcinoma in situ (atypical pleomorphic adenoma) or PSA with non-invasive malignant transformation.

Conclusion: This survey shows that superficial parotidectomy is the standard treatment method for primary pleomorphic adenoma in children and adults. Surgery combined with radiotherapy is the preferred option in the case of multinodular recurrence, and surgery alone is recommended in the case of a solitary recurrence.

Key Words: Carcinoma in pleomorphic adenoma; pleomorphic adenoma; superficial parotidectomy.

Amaç: Bu çalışmada Büyük Britanya'daki oral ve maksillofasyal cerrahlar arasında pleomorfik adenomların tedavisi ile ilgili güncel noktalar vurgulandı.

Hastalar ve Yöntemler: Bir anket hazırlandı ve İngiliz Oral Maksillofasyal Cerrahi Derneği'ne kayıtlı tüm danışman üyelerine (n=263) gönderildi. Ankette, cerrahın cerrahi deneyimi, bir yetişkin ve bir çocuktaki parotis bezinin süperfisyel lobunda yeni tanı konulan 3 cm çapındaki pleomorfik adenomda tercih edilen cerrahi tedavi ve pleomorfik tükürük bezi adenom (PTA)'ların tedavi seçenekleri, aşağıdaki dört senaryo ile değerlendirildi: (i) PSA ile birlikte tam eksizyon ya da tam olmayan eksizyon; (ii) PTA ve karsinom in situ (atipik pleomorfik adenom) ile birlikte tam eksizyon ya da tam olmayan eksizyon; (iii) PTA ile invaziv olmayan malign transformasyon ile tam eksizyon ya da tam olmayan eksizyon; ve (iv) PTA içinde karsinom ile tam eksizyon ya da tam olmayan eksizyon. Başlangıçta yanıt oranı düşük idi ancak iki katılımcının katkısıyla yanıt oranında artış oldu.

Bulgular: Son yanıt oranı %67 oldu. Sonuçlar, katılımcıların yetişkin ve çocuklardaki pleomorfik adenom tedavisiyle ilgili görüşleri arasında anlamlı fark bulunmadığını gösterdi. Yüz otuz üç cerrahın 75'i (%56.4), multinodüler nüksü olan yaşlı hastaları radyoterapi ve cerrahiden oluşan kombine tedavi yöntemi ile tedavi etmişlerdi. Yüz beş (%78.9) cerrah tekli (soliter) nüksü olan yaşlı hastaları yalnız cerrahi yöntem ile tedavi etmişlerdi. Karsinom in situ (atipik pleomorfik adenom) ya da invaziv olmayan malign transformasyon olan PTA tedavisi ile ilgili tam eksizyon üzerine bir fikir birliği vardı.

Sonuç: Bu anket, çocuklarda ve yetişkinlerdeki primer pleomorfik adenom tedavisi için süperfisyel parotidektominin standart tedavi yöntemi olduğunu göstermiştir. Radyoterapi ile birlikte kombine cerrahi tedavisi multinodüler nüks olgularında tercih edilen seçenektir ve tek başına cerrahi tedavi ise tekli nüks olgularında önerilmektedir.

Anahtar Sözcükler: Pleomorfik adenom karsinomu; pleomorfik adenom; süperfisyel parotidektomi.

There is a lack of clear evidence-based data regarding the management of pleomorphic salivary adenoma (PSA), particularly in uncommon situations such as malignant transformation or those arising in childhood.^[1] Even in straightforward cases, capsular dissection, superficial parotidectomy, subtotal parotidectomy and total parotidectomy may be used to treat pleomorphic adenomas. When there is a recurrence, the surgeon is faced with the often-difficult task of re-operating to remove multiple recurrent tumor nodules in a scarred operative field containing the facial nerve. Proper surgical management is still controversial. Some authors advocate resection of the entire tumor and surrounding tissue along with the involved facial nerve.^[2] Mendenhall et al.^[3] have indicated that the optimal treatment of pleomorphic adenoma is superficial or total parotidectomy with facial nerve preservation, which results in local control rates of 95% or higher.

Although Samson et al.^[4] for example, report low recurrence rates of 1% to 3% following superficial parotidectomy, others still recommend a complete parotidectomy with preservation of the facial nerve.^[5]

The study aimed to determine the current opinion and experiences regarding the management of pleomorphic adenomas among practising oral and maxillofacial surgeons in the United Kingdom. The survey also evaluated the influence of clinical factors such as age at presentation, and the influence of the histopathological diagnosis in transforming and malignant lesions.

PATIENTS AND METHODS

A questionnaire was sent to all the consultant members of the British Society of Oral Maxillofacial Surgery. This involved 263 oral and maxillofacial surgeons in the United Kingdom. The questionnaire was submitted beforehand and formal permission to circulate the questionnaire was obtained from the society. All the addresses of the surgeons were provided by the British Association of Oral and Maxillofacial Surgery.

Before submitting the questionnaire, a pilot study was performed with five consultants in oral and maxillofacial surgery. The questionnaire was modified according to their recommendations. Modifications included a better definition of "incomplete excision" (this referred to demonstration of tumor cells at the margin of the resected

specimen) and clarification that there was no nodal involvement by the tumor.

Statistical analysis

The data were described using frequency distributions and cross-tabulations. The associations between the number of PSA cases that the surgeons estimated they had treated since being appointed as consultants and the percentage of spillage or rupture during the removal of PSA were assessed by Pearson's correlation coefficient.

RESULTS

One-hundred and seventy-seven out of 263 surgeons participated in the main study (response rate 67.3%); 133 surgeons (75%) out of the 177 had previously operated on salivary gland tumors and were included in the analysis. Forty-two were non-eligible because they had not operated on salivary gland tumors in the last five years, and two responses were not clear. The mean of number of pleomorphic adenomas that the surgeons had treated since being appointed as consultants was 71, the maximum was 300, and the minimum was two. The mean number of pleomorphic adenomas that the surgeons had treated during the last five years was 33, the maximum was 150, and the minimum was one. The mean number of borderline malignant PSAs that the surgeons had treated during the last five years was only 3.2, with a maximum of 20, and a minimum of one (Table 1).

The current treatment of pleomorphic adenoma in children

Ninety-five (71.4%) surgeons out of 133 advocated superficial parotidectomy as the optimal treatment in children for PSA, while 18 (13.5%) surgeons advocated capsular dissection. Only 20 (15%) surgeons out of 133 did not choose either of these treatments (Table 2).

Table 1. Frequency test shows the mean, maximum, minimum of the treated cases

Number of treated cases	Mean	Maximum	Minimum
N1	71	300	2
N2	33	150	1
N3	3.2	20	1

N1: Number of pleomorphic adenomas which the surgeons had treated since becoming a consultant; N2: Number of pleomorphic adenoma which the surgeons had treated during the last five years; N3: Number of borderline malignant which the surgeons had treated during the last five years.

Table 2. Frequency test shows the different treatment options for pleomorphic adenoma in children

The surgeons' preferred treatment option	Frequency	Percent
Capsular dissection	18	13.5
Superficial parotidectomy	95	71.4
<i>Total</i>	113	85.0
No answer	20	15.0
<i>Total</i>	133	100.0

The surgical treatment of pleomorphic adenoma in the superficial lobe of the parotid gland in adults and children

The recommended treatment of primary pleomorphic adenoma with a 3 cm diameter was similar in both an adults and children. One-hundred and eleven (83.5%) surgeons out of 133 treated the adult patients with superficial parotidectomy and eight (6%) surgeons applied capsular dissection, but only one surgeon (0.8%) performed enucleation with radiotherapy. Six surgeons (4.5%) applied other treatments such as partial parotidectomy. Eighty-six (64.7%) surgeons treated the children with a superficial parotidectomy, 18 (13.5%) with capsular

dissection, and five (3.8%) applied other treatment such as partial parotidectomy. Only one surgeon used enucleation with radiotherapy (Table 3).

The cross-tabulation of frequencies shows that 86 out of 110 surgeons preferred superficial parotidectomy in children and in adults, nine would use superficial parotidectomy in adults and capsular dissection in children, eight would use capsular dissection in both adults and children, and one would use enucleation with combined radiotherapy in adults and children. Only one would prefer other a treatment such as partial parotidectomy for an adult but capsular dissection in a child, and five would use another treatment, such as partial parotidectomy, in both children and adults.

Treatment of recurrent tumor

The results indicated that 75 (56.4%) treated multinodular recurrence in older patients with surgery combined with radiotherapy. Forty-four (33.1%) would use surgery alone. In cases of solitary recurrence, 105 (78.9%) treated older patients with surgery alone while 16 (12.0%) would use surgery combined with radiotherapy (Table 4).

Table 3. Frequency test shows the different treatment options for a new 3 cm diameter pleomorphic adenoma in the superficial lobe of the parotid gland in adults and children

The surgeons' preferred treatment option	Frequency in an adult	Percent in an adult	Frequency in children	Percent in children
Capsular dissection	8	6.0	18	13.5
Enucleation with radiotherapy	1	0.8	1	0.8
Superficial parotidectomy	111	83.5	86	64.7
Other	6	4.5	5	3.8
<i>Total</i>	126	94.7	110	82.7
No answer	7	5.3	23	17.3
<i>Total</i>	133	100.0	133	100.0

Table 4. Frequency test shows the different treatment options of a healthy 50-year-old patient in the case of multinodular and solitary recurrence

The surgeons' preferred treatment option	Frequency in multinodular recurrence	Percent in multinodular recurrence	Frequency in solitary recurrence	Percent in solitary recurrence
Surgery alone	44	33.1	105	78.9
Surgery with radiotherapy	75	56.4	16	12.0
<i>Total</i>	119	89.5	121	90.9
No answer	14	10.5	12	9.1
<i>Total</i>	133	100.0	133	100.0

Table 5. The different treatment options and number of surgeons who treated a middle-aged patient with 3 cm diameter tumor from the superficial lobe of the parotid gland after receiving a pathology report of pleomorphic adenoma with diagnosis of either complete or incomplete excision shown as a cross-tabulation of frequency

	Incomplete excision				<i>Total</i>
	Follow-up	Further surgery	Further surgery with radiotherapy	Radiotherapy alone	
Complete excision					
Follow-up	45	29	12	31	117
Radiotherapy alone	1	–	–	–	1
<i>Total</i>	46	29	12	31	118

The cross-tabulation of frequency shows that 55 out of 116 surgeons would use surgery combined with radiotherapy in cases of multinodular recurrence, but would use surgery alone to treat the solitary recurrence. Forty-three surgeons would prefer surgery alone in the case of either multinodular or solitary recurrence. Sixteen out of 116 surgeons would use surgery combined with radiotherapy for both multinodular or solitary recurrences. Only one would use surgery alone to treat solitary recurrence, and another treatment to treat multinodular recurrence. Another respondent would use another treatment in a case of solitary recurrence, and surgery with radiotherapy in a case of multinodular recurrence.

Influence of the histopathological diagnosis on selection of treatment options for pleomorphic adenoma, its transforming variants and carcinoma arising in pleomorphic adenoma

1- Pleomorphic adenoma with complete or incomplete excision

Forty-five respondents prefer following up their patients in cases of pleomorphic adenoma with either complete or incomplete tumor excision (Table 5). Thirty-one would prefer follow-up in the case of complete tumor excision, and radiotherapy alone in the case of incomplete excision. Twenty-nine surgeons advocated further surgery in the case of incomplete excision, and follow-up in the case of complete excision. Twelve respondents would use further surgery with radiotherapy in the case of incomplete tumor excision, and follow-up in cases of complete excision. Only one would use radiotherapy alone in the case of complete excision, and follow-up in the case of incomplete excision.

2- Carcinoma in situ (atypical pleomorphic adenoma) with complete or incomplete excision

Thirty-six respondents would use radiotherapy in the case of carcinoma in situ with incomplete excision, and follow-up with complete excision (Table 6). Twenty-seven would prefer further surgery combined with radiotherapy in the case of

Table 6. The different treatment options and number of surgeons who treated a middle-aged patient, with a 3 cm diameter tumor of the parotid gland after receiving a pathology report with carcinoma in situ (atypical pleomorphic adenoma) and either complete or incomplete excision shown as a cross-tabulation of frequency

	Incomplete excision				<i>Total</i>
	Follow-up	Further surgery	Further surgery with radiotherapy	Radiotherapy alone	
Complete excision					
Follow-up	12	27	27	36	102
Further surgery	–	1	1	–	2
Radiotherapy alone	–	–	2	5	7
<i>Total</i>	12	28	30	41	111

Table 7. The different treatment options and number of surgeons who treated a middle-aged patients with a 3 cm diameter tumor from the superficial lobe of the parotid gland after receiving a pathology report describing non-invasive malignant transformation and either complete or incomplete excision, shown as a cross-tabulation of frequency

	Incomplete excision				<i>Total</i>
	Follow-up	Further surgery	Further surgery with radiotherapy	Radiotherapy alone	
Complete excision					
Follow-up	6	25	26	37	94
Further surgery	–	1	2	–	3
Further surgery with radiotherapy	–	–	1	–	1
Radiotherapy alone	–	–	3	11	14
<i>Total</i>	6	26	32	48	112

incomplete excision, but only follow-up in the case of complete excision.

Twenty-seven surgeons would undertake further surgery in the case of incomplete excision, and follow-up in the case of complete excision. Twelve respondents preferred follow-up in the case of complete or incomplete excision.

Only one respondent would use further surgery in the case of complete or incomplete excision. One other preferred further surgery with radiotherapy in the case of incomplete excision, and further surgery in the case of complete excision.

Two surgeons advocated further surgery with radiotherapy in the case of incomplete excision, and radiotherapy alone in the case of complete excision. Five respondents used radiotherapy alone in cases of complete or incomplete excision.

3- Pleomorphic adenoma with non-invasive malignant transformation with complete or incomplete excision

Thirty-seven out of 112 surgeons would use radiotherapy alone in the case of incomplete excision, and follow-up alone in the case of complete excision (Table 7). Twenty-six respondents preferred further surgery combined with radiotherapy in the case of incomplete excision, and follow-up alone in the case of complete excision. Twenty-five recommended further surgery in the case of incomplete excision, and follow-up in the case of complete excision. Eleven used radiotherapy alone in either complete or incomplete excision, while six surgeons would use follow-up in both complete and incomplete excision. Three respondents used further surgery with radiotherapy for incomplete excision, and radiotherapy alone in the case of complete

Table 8. The different treatment options and number of surgeons who treated a middle-aged patients with a 3 cm diameter tumor from the superficial lobe of the parotid gland after receiving a pathology report describing carcinoma in pleomorphic adenoma with either complete or incomplete excision, shown as a cross-tabulation of frequency

	Incomplete excision				<i>Total</i>
	Follow-up	Further surgery	Further surgery with radiotherapy	Radiotherapy alone	
Complete excision					
Follow-up	1	12	32	24	69
Further surgery	–	1	2	–	3
Further surgery with radiotherapy	–	–	6	–	6
Radiotherapy alone	–	–	15	18	33
<i>Total</i>	1	13	55	42	111

excision. Two recommended further surgery with radiotherapy in the case of incomplete excision, and further surgery in the case of complete excision while there was one who would use further surgery with radiotherapy in the case of complete or incomplete excision. Only one would use further surgery in either complete or incomplete excision.

4- Carcinoma in pleomorphic adenoma with complete or incomplete excision

Thirty-two respondents would use further surgery with radiotherapy in the case of incomplete excision, and follow-up alone in the case of complete excision (Table 8).

Twenty-four surgeons would undertake radiotherapy alone in the case of incomplete excision, and follow-up alone in the case of complete excision while 18 would use radiotherapy alone in either complete or incomplete excision. Fifteen preferred further surgery combined with radiotherapy in the case of incomplete excision, and radiotherapy alone in the case of complete excision and 12 used further surgery in the case of incomplete excision, and follow-up in the case of complete excision. Six surgeons would use further surgery with radiotherapy in the case of complete or incomplete excision while there were two who would use further surgery with radiotherapy in the case of incomplete excision, and further surgery in the case of complete excision. Only one preferred further surgery in cases of complete or incomplete excision. One out of 111 would advocate follow-up alone in either complete or incomplete excision.

Surgeons' estimated rates of spillage or rupture

The percentage of operations for PSA removal where spillage or rupture occurred during removal of pleomorphic adenoma was variable: 110 surgeons out of 125 estimated spillage rates ranging between 0-5% but only 15 surgeons estimated rupture or spillage rate that ranged from 10-20%.

DISCUSSION

The treatment of pleomorphic adenoma is controversial especially regarding the management of recurrent tumors and cases where malignant transformation has occurred. The results have shown that 71.4% of the oral and maxillofacial surgeons in the United Kingdom preferred superficial parotidectomy for straightforward cases of PSA in children. Extracapsular dissection was the next most preferred option. There was no signifi-

cant difference in the opinions of the respondents regarding treatment of a pleomorphic adenoma of 3 cm diameter in both adults and children. Also our results have shown that 83.5% of the respondents surgeons would use superficial parotidectomy in adults and 64.7% in children.

Despite the preference for this technique, several series report a small number of recurrences after superficial parotidectomy.^[6-8] One study, McGurk et al,^[9] compared the techniques of capsular dissection and superficial dissection in two groups and they found the rate of recurrence was similar at around 2%. Extracapsular dissection is not commonly favored in the United Kingdom, according to the findings of the survey, but some surgeons prefer this method to superficial parotidectomy, in order to reduce the rates of complications such as paralysis of the facial muscles.^[10,11] An important consideration is the rate of spillage or rupture, as the evidence suggests that in skilled hands low spillage and rupture rates can be achieved using extracapsular dissection. The survey participants' experience noted very low rates of rupture and spillage (with some exceptions) and it may be that superficial parotidectomy is preferred because of a perceived risk of perforation of the capsule during extracapsular dissection when the procedure is first used.

The survey revealed differences in opinion regarding the provision of postoperative radiotherapy for cases of primary disease. Postoperative radiotherapy was not advised for primary pleomorphic adenoma when complete removal had been reported. However when excision was incomplete, opinion was divided regarding further surgery with and without radiotherapy and radiotherapy alone as possible modalities. This variation reflects the lack of evidence-based management for this situation. In general, oral and maxillofacial surgeons were more inclined to advocate postoperative radiotherapy for pleomorphic adenomas which showed signs of transformation, but only when excision was reported as incomplete. When a diagnosis of invasive carcinoma in PSA was made then the majority of respondents advocated the use of radiotherapy, even when primary excision was reported as complete. However, there were still varied opinions regarding further surgery for incomplete excision, with many preferring radiotherapy alone. This is probably a reflection of the known poor prognosis of carcinoma arising in PSA.

It is generally recognised that the treatment of recurrent tumor following previous local excision involves total parotidectomy with preservation of the facial nerve as the minimum procedure. There are two main problems in the treatment of recurrent cases: first is facial nerve damage, which is much more common than in primary surgery while the second is multiple recurrence. Care must be taken to distinguish multinodular recurrence from solitary recurrence. The survey revealed that opinion was divided regarding the use of postoperative radiotherapy for recurrent disease. Surgery with postoperative radiotherapy was favored by 56.4% for multinodular recurrence and by only 12% for solitary recurrence. This difference is consistent with evidence published by Renehan et al,^[12] who showed that surgery combined with radiotherapy was better than surgery alone in the treatment of the multinodular recurrence of PSA. In the case of the solitary recurrence, surgery alone gives the same result as surgery combined with radiotherapy and minimizes the opportunity for malignant transformation of PSA. It is of interest that around one-third of participants would not advise postoperative radiotherapy following surgery for multiple recurrence, indicating the need for randomised controlled trials to evaluate the use of postoperative radiotherapy in the management of multinodular recurrence.

In this study the response rate was 67% which is accepted as adequate for a postal survey. It may also be that some of the eligible surgeons restrict their practice and do not perform salivary tumor surgery. Such oral and maxillofacial surgeons are less likely to respond.

This survey reveals inconsistencies in the general approach to the use of postoperative radiotherapy and surgical technique in a number of situations. This is not surprising as only a limited number of clinical guidelines regarding the management of pleomorphic adenoma and its sequelae.

This study has shown that there is a lack of published evidence regarding the management of pleomorphic adenomas, although there is a consensus that surgery which ensures complete removal with the capsule is the mode of choice for primary pleomorphic adenomas. There is divided opinion regarding the most appropriate treatment for recurrence and for when transformation is in progress or has occurred. Randomised controlled

clinical trials could be used to address these issues but are difficult to perform because of likely low accrual rates and the need for prolonged follow-up times to assess clinical outcomes. The extremely poor prognosis of carcinoma arising in pleomorphic adenoma in whichever provided treatment points to a need for translational research leading to innovative therapies for these rare but generally lethal malignancies. Such therapies would have to be tested in a small number of cases within a global network because of the rarity of these lesions.

The responses of 13 surgeons in this study who treated at least 200 cases since they were appointed as a consultant have been analysed as a subgroup. All preferred superficial parotidectomy (3 cm PSA in the superficial lobe of parotid gland) except one who preferred the extracapsular dissection technique in adults and these findings are corresponding with Donati et al,^[6] Woods^[13] and Donovan et al.^[14] The interesting point was that the percentage of estimated incidence spillage was 7% for seven surgeons, 5% for one surgeon, 2% for one surgeon, nil for three surgeons with one who did not respond to that question.

This subgroup of most experienced salivary operators expressed similar opinions to the overall group, except that they estimated lower spillage rates.

It can be concluded that oral and maxillofacial surgeons in the United Kingdom currently advocate superficial parotidectomy as the standard treatment for primary pleomorphic adenoma in children and adults. Surgery combined with radiotherapy is the preferred option in the case of multinodular recurrence, and surgery alone is recommended in the case of a solitary recurrence. Also there is agreement about the philosophy for the treatment of carcinoma in situ or PSA with non-invasive malignant transformation with complete excision. In both these cases, most of the surgeons advocate following up their patients as a first option before making any decision to use surgery alone or surgery combined with radiotherapy or radiotherapy alone. The results revealed no complete agreement in relation to the treatment of carcinoma in PSA.

Inconsistent views revealed by the survey, whilst recognising its limitations, indicate that there may be a need for production of consensus

guidelines. Additional data would be necessary and a broad expert panel would need to oversee their formulation.

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