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Original Article

Basal cell adenoma of the parotid gland: clinical and pathological findings in 29 cases

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Abstract: *Objective*: To determine the clinical and pathological features of basal cell adenoma (BCA) of the parotid gland. *Methods*: This is a retrospective study of 29 parotid BCAs in 28 patients who underwent surgery at the Department of Otolaryngology Head and Neck Surgery, Xinhua Hospital, Shanghai Jiaotong University School of Medicine, between October 2000 and June 2013. The tumors were categorized according to their location in the parotid gland as superior superficial lobe, inferior superficial lobe and deep lobe. *Results*: The mean age was 57.0 years (range, 32-83 years). The clinical manifestations of parotid BCAs were consistent with those of other benign parotid tumors. There were no significant differences in age, average disease duration and tumor size among the three tumor groups. There were 11 deep tumors (11/29, 37.9%), and five of them exhibited cystic degeneration (5/11, 45.5%). A total of 15 patients underwent FNAB examination, and the results were positive in seven patients (7/15, 46.7%). Mild facial nerve function impairment occurred in five patients (House-Brackmann grade II), of whom, three had recovered by the 6-month follow-up. No cases of local recurrence or malignant transformation were observed during follow-up. *Conclusion*: The clinical features of BCA are consistent with those of other benign tumors. The deep lobe of the parotid gland is more likely to develop BCAs, and thus, this diagnosis should be considered in patients with deep-lobe tumors, especially when accompanied with cystic degeneration. FNAB can increase the rate of preoperative diagnoses.

Keywords: Basal cell adenoma, benign parotid tumor

Introduction

The majority of salivary gland tumors are benign, and parotid gland tumors are the most common salivary gland tumors [1, 2]. Basal cell adenoma (BCA) is a rare benign tumor of the parotid gland, and accounts for approximately 1%-3% of all benign parotid gland tumors [1, 3]. BCA was described as a new distinct histological entity by the WHO in 1991 [4] and has since attracted increasing research interest after being classified as one of nine salivary gland cancers in 2005 [5].

BCA is composed of basaloid cells, which lack a mesenchymal component or chondromyxoid stroma [2]. It is difficult to distinguish BCAs from other benign tumors of the parotid gland in the preoperative phase. The imaging features of BCA have been correlated with those of cystic lesions [6]. However, few reports have described the clinical diagnosis and treatment of these tumors [3, 7, 8]. In the present study, we investigated the diagnosis, computed tomography (CT) features, treatment, pathological features and follow-up results of 29 parotid BCAs in 28 patients.

Materials and methods

Patients and methods

This is a retrospective study of 29 parotid BCAs in 28 patients who underwent surgery at the Department of Otolaryngology Head and Neck Surgery, Xinhua Hospital, Shanghai Jiaotong University School of Medicine, between October 2000 and June 2013. The diagnosis of BCA was confirmed via pathological examination. Clinical data were collected from all the patients, includ-

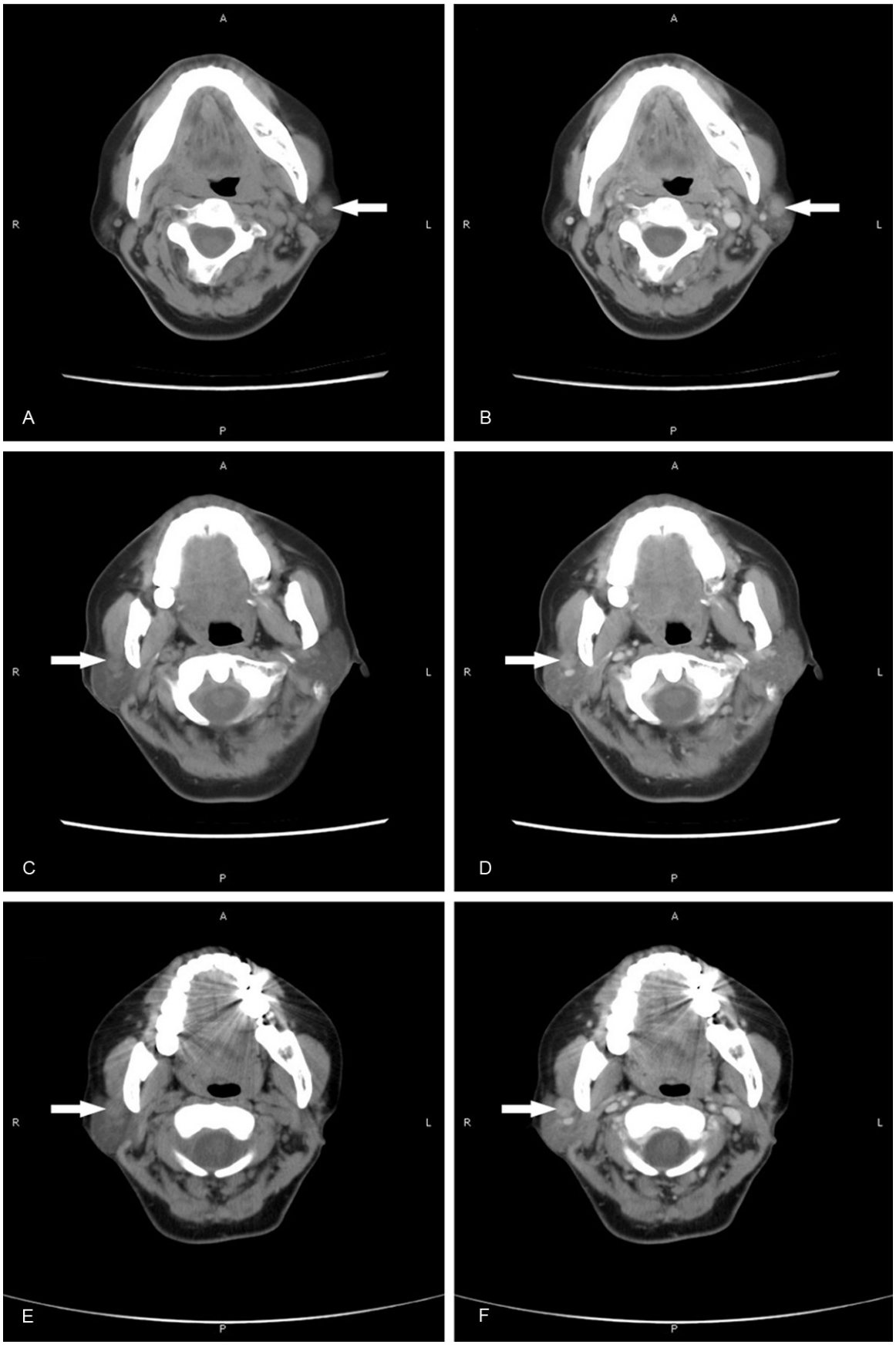
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Figure 1. Bilateral basal cell adenomas. A 54-year-old woman was diagnosed with bilateral parotid tumors. A and B: One tumor is seen in the lower part of the superficial lobe of the left parotid gland (white arrow) on a CT scan. C and D: The other tumor is located in the deep lobe of the right parotid gland, and shows an ill-defined border (white arrow). E and F: Two years later, CT shows a round, ill-defined, homogeneously enhancing lesion in the region of the deep lobe of the right parotid gland. This lesion is slightly larger than that observed 2 years ago (white arrow).

ing age, gender, symptoms, CT findings, surgical details, and pathology results.

The 28 study patients included 13 (46.4%) men and 15 (53.6%) women. The right parotid gland was involved in 37.9% (11/29) of cases, and the left gland was involved in 62.1% (18/29) of cases. The mean age was 57 years (range, 32-83 years). According to their location in the parotid gland, BCA tumors were categorized as deep or superficial. The latter were further classified into superior and inferior tumors on the basis of their relationship to the mandibular branch of the facial nerve [2].

All patients were followed up (duration: 6 months to 12 years) to evaluate facial nerve function and postoperative tumor recurrence.

Statistical analysis

Statistical analysis of the data was performed using SPSS software (ver. 19.0, SPSS Inc.). One-way analysis of variance was used for comparisons of categorical variables (age, duration of disease, tumor size). The significance level was set at $P < 0.05$.

Results

This study consists of 29 tumors in 28 patients; one patient had bilateral BCAs (**Figure 1**). Of the 29 tumors, 8 (27.6%) were located in the superficial lobe, superior to the mandibular branch of the facial nerve (superior tumors), 10 (34.5%) were located inferior to this branch in the superficial lobe (inferior tumors) and 11 (37.9%) tumors were located in the deep lobe (deep tumors; **Table 1**).

Superior tumors were present in four men and four women, who had an average age of 52.9 years (range, 32-71 years). Five of these tumors were in the left gland, and three were in the right gland. The average interval between the onset of symptoms and the administration of treatment was 18.5 ± 20.6 months. Four of these patients had localized pain prior to treatment. Ultrasonographic examinations showed a single, hypoechoic, well-defined solid tumor in each of these patients. CT showed a well-defined round mass, with an average maximum diameter of 19.8 ± 8.5 mm. Heterogeneous enhancement was also observed. Only two of the eight tumors (25%) showed cystic degeneration (**Table 1**). In these patients, the surgical treatment included dissection of the facial nerve and excision of the tumor and superficial lobe of the parotid gland. Three of the eight patients (37.5%) underwent preoperative fine needle aspiration biopsy (FNAB). The FNAB examination revealed a BCA in one patient (12.5%), while in the other two (25%) patients, it indicated the presence of benign tumors. The other five tumors were diagnosed on pathological examination. No facial nerve injury and no postoperative tumor recurrence occurred in this group (**Table 2**; **Figure 2A**, **2B**).

In the inferior tumor group, there were seven men and three women, with a mean age of 53.9 years (range, 36-75 years). Eight of these tumors were located in the left parotid gland, and two were located in the right gland. The average disease duration before surgery was 23.4 ± 36.7 months. Except for a local mass, there were no other clinical symptoms. The ultrasonographic features were similar to those of the superior tumors. The average maximum diameter of the tumors on CT was 22.1 ± 12.5 mm. Of the 10 tumors, 3 (33.3%) exhibited cystic degeneration (**Table 1**). The surgical treatment involved dissection of the facial nerve and excision of the tumor and superficial lobe of the parotid gland. Five patients (50%) underwent FNAB preoperatively, and two of these patients (20%) were diagnosed with BCA. The diagnosis was confirmed on intraoperative frozen-section examination in three patients and on postoperative pathological examination in other patients. In one patient (10%), the facial nerve function deteriorated to House-Brackmann (HB) grade II after the surgery, but recovered after 6 months. No patient developed recurrence after the operation (**Table 2**; **Figure 2C**, **2D**).

In the deep tumor group, there were two men and nine women, with an average age of 62.1

Table 1. Clinical characteristics of patients with basal cell adenoma

Tumor site[1]	Tumors[2]	Sex[3]	Age (years)	Side	Clinic symptoms	Duration of disease (months)	Tumor size[4] (mm)	Cystic formation
Superior (S)								
		M = 4		R = 3				
		F = 4		L = 5				
	8 (27.6%)		52.9 ± 13.6		4	18.5 ± 20.6	19.8 ± 8.5	2 (25.0%)
Inferior (S)								
		M = 7		R = 2				
		F = 3		L = 8				
	10 (34.5%)		53.8 ± 12.8		0	23.4 ± 36.7	22.1 ± 11.5	3 (33.3%)
Deep								
		M = 2		R = 6				
		F = 9		L = 5				
	11 (37.9%)		62.1 ± 12.8		1	10.5 ± 11.0	20.4 ± 5.2	5 (45.5%)
Total	29	M = 13 (46.4%)	57.0±13.2	R = 11 (37.9%)	4	17.1 ± 24.8	20.8 ±8.4	
		F = 15 (53.6%)		L = 18 (62.1%)				
		/	P > 0.05	/	/	P > 0.05	P > 0.05	/

[1]Tumor site. Upper Superior: The tumor is superior to the mandibular branch of the facial nerve in the superficial lobe. Inferior: The tumor is inferior to the mandibular branch of the facial nerve in the superficial lobe. Deep: The tumor is in the deep lobe of the parotid gland. [2]Tumors. One of the 28 patients had bilateral tumors, so the total number of tumors was 29. [3]Sex. M: male; F: female. [4]Tumor size. The mean and standard deviation of the maximum tumor diameter on CT imaging were calculated.

Table 2. Surgical and pathological findings in patients with basal cell adenoma

Tumor site	Surgery[1]	FNAB[2]	FSB[2]	Histological Subtype	Follow-up Facial nerve function	Follow-up Tumor recurrence
Upper (S)						
8 tumors	(1)	Not performed = 5	Not performed: = 1	Solid = 6	/	0
		Not confirmed: = 0	Not confirmed: = 2	Solid Tubular = 0		
		Benign = 2	Benign = 2	Solid Trabecular = 1		
		BCA confirmed = 1	BCA confirmed = 3	Membranous = 1		
Lower (S)						
10 tumors	(1)	Not performed = 5	Not performed = 1	Solid = 7	One case was facial nerve function HB II after surgery.	0
		Not confirmed: = 2	Not confirmed: = 3	Solid Tubular = 2		
		Benign = 1	Benign = 3	Solid Trabecular = 1		
		BCA confirmed = 2	BCA confirmed = 3	Membranous = 0		
Deep						
11 tumors	(2)	Not performed = 4	Not performed = 4	Solid = 4	Four cases were facial nerve function HB II after surgery and two of them failed to recover.	0
		Not confirmed: = 2	Not confirmed: = 0	Solid Tubular = 4		
		Benign = 1	Benign = 1	Solid Trabecular = 2		
		BCA confirmed = 4	BCA confirmed = 6	Membranous = 1		
Total	(1) or (2) surgery[1]	Not performed = 14	Not performed = 6	Solid = 17	5:24	Not recurrent
		Not confirmed = 4	Not confirmed = 5	Solid Tubular = 6		in all of them
		Benign = 4	Benign = 6	Solid Trabecular = 4		
		BCA confirmed = 7	BCA confirmed = 14	Membranous = 2		

[1]Surgery. (1) The surgeon dissected the facial nerve trunk and its branches, and then excised the superficial lobe of the parotid gland and the tumor. (2) The surgeon dissected the facial nerve trunk and its branches, and then excised the superficial and deep lobes of the parotid gland and the tumor. [2]FNAB: Fine needle aspiration biopsy; FSB: frozen-section biopsy.

years (range, 43-83 years). The mean age in this group was slightly higher than that in the other two groups, but the difference was not statistically significant. Five tumors were located in the left parotid gland, and six were located in the right gland. The average disease duration prior to surgery was 10.5 ± 11.0 months. One patient received treatment because of local pain. In these patients, ultrasonography revealed a hypoechoic mass located in the deep lobe of the parotid gland. CT examination showed that the maximum tumor diameter was 20.4 ± 5.2 mm. Cystic degeneration was present in 5 of the 11 tumors (45.5%), which was higher than the incidence in the other two groups (**Table 1**). The surgical treatment included dissection of the facial nerve and excision of the tumor, and the superficial and deep lobes of the parotid gland. FNAB was conducted in seven cases (63.6%), and four tumors were confirmed to be BCA on FNAB (4/11, 36.4%). Six tumors (6/11, 54.5%) were confirmed on intraoperative frozen-section biopsy, and the others were confirmed on the final pathological diagnosis. In four cases, the facial nerve function deteriorated to HB grade II after the surgery. Two of them recovered within 6 months and in other two cases, this function failed to recover. Two patients died of other diseases during follow-up. The remaining patients had no local recurrence or malignant transformation during the follow-up (**Table 2**; **Figure 2E**, **2F**).

BCA is composed of basaloid cells, which can be divided into the following categories: solid tubular type (**Figure 3A**), solid trabecular type (**Figure 3B**), membranous type cells (**Figure 3C**), and solid type (**Figure 3D**). The histological subtype was evaluated by examination of paraffin sections and Hematoxylin and Eosin staining. The histological subtypes found in this study were as follows: solid tubular lesions, 6 tumors; trabecular lesions, 4 tumors; membranous lesions, 2 tumors; and solid lesions, 17 tumors; (**Table 2**).

Discussion

In the present study, the clinical features of 28 patients (29 BCAs) were characterized according to the tumor position in the parotid grand. The proportion of women in the study group was 53.6% (15/28), which was slightly lower than the percentage in previous reports (> 70%) [3, 8, 9]. Furthermore, in the present study, the sex ratio was different among the three groups, with the highest proportion of female patients being seen in the deep tumor group (9/11, 81.8%). The average age was 57.0 years (range, 32-83 years), which is similar to that in previous reports [2, 7]. Although this study did not find statistically significant differences in the average age among the three groups, the average age in the deep tumor group seemed slightly higher than the average ages in the other two groups. Therefore, we speculate that a diagnosis of BCA should be considered in elderly female patients with tumors located in the deep lobe of the parotid gland.

In this study, left-sided tumors accounted for 62.1% (18/29) of the total number of tumors, which is slightly higher than the proportion of right-sided tumors. These results were consistent with the findings of other studies [10].

BCAs have the general characteristics of benign tumors, being slow growing and painless. In this study, five patients experienced localized pain, a symptom which has not been mentioned in previous reports. Among these patients, four had superior tumors, and one had a deep tumor. Pain was not present in the patients with inferior tumors. We thus speculated that the superficial lobe of the parotid gland is more prone to clinical symptoms. As inflammation tends to occur in this site, a rapid increase in tumor-related edema may lead to local symptoms.

The average disease duration was 17.1 months (range, 1-120 months), which is similar to that previously reported [3, 9]. Moreover, the duration did not significantly differ among the three groups, but we noticed that the average disease duration was slightly shorter in the deep group than in the other two groups. A possible reason for this trend is that deep lobe tumors were more likely to be noticed by a physician, leading to earlier diagnosis and treatment.

The imaging features of BCAs mainly included contrast enhancement and well-defined borders. We found that it was difficult to clearly distinguish between benign parotid tumors and BCAs on CT, which is consistent with previous reports [7, 9, 11-12]. The average maximum diameter was 20.8 ± 8.4 mm, which is similar to that in previous reports, and there was no

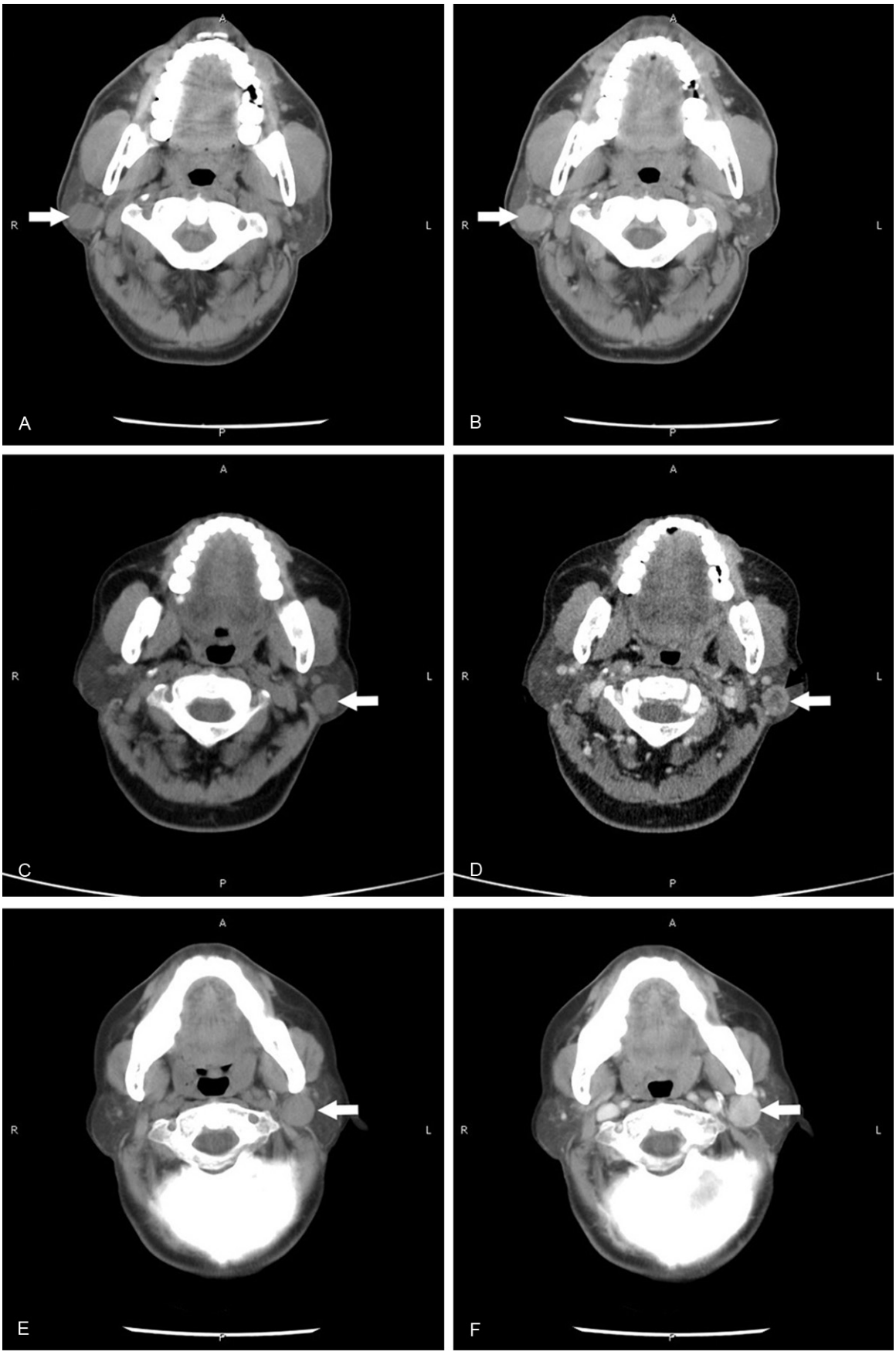
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Figure 2. Representative images of tumors in the three study groups. A and B: A 45-year-old man with a basal cell adenoma in the upper part of the superficial lobe of the left parotid gland. B: CT shows a round, well-defined homogeneously enhancing lesion (white arrow). C and D: A 60-year-old woman with a basal cell adenoma in the lower part of the superficial lobe of the left parotid gland (white arrow). D: The tumor shows slight heterogeneous enhancement (white arrow) on contrast-enhanced CT. E and F: A 76-year-old woman with a basal cell adenoma in the deep lobe of the left parotid gland. F: CT shows a round, well-defined, homogeneously enhancing lesion in the deep lobe of the left parotid gland (white arrow).

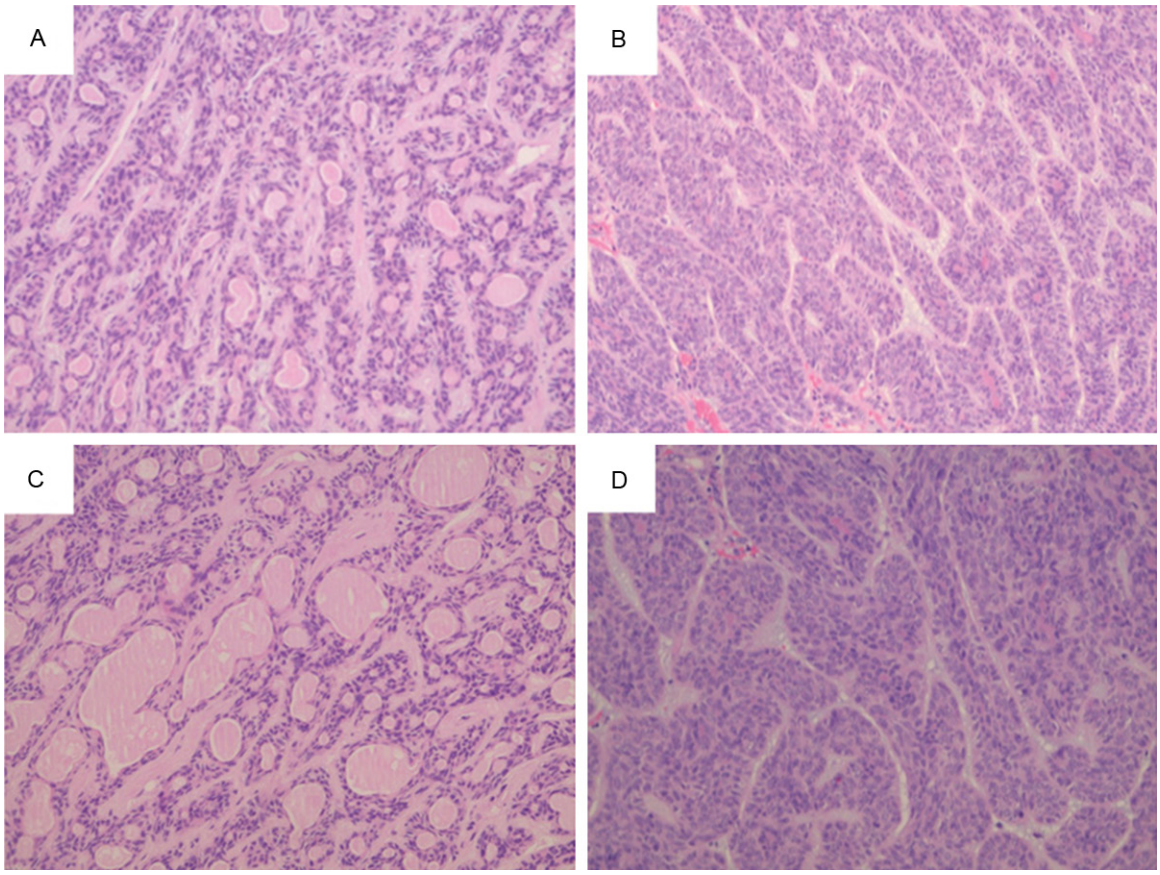

Figure 3. Histogicalsubstypes of basal cell adenoma tumor in the parotid gland. A: Solid tubular type; B: Solid trabecular type; C: Membranous type; D: Solid type.

significant difference in this diameter among the three groups. In our study, 37.9% of tumors (11/29) were located in the deep lobe. This ratio was higher than that previously reported [12]. In addition, the proportion of cystic degeneration was 34.5% (10/29) in this study, which is similar to that in earlier reports [3, 13]. However, it is worth noting that the proportion of cystic tumors in the deep lobe of the parotid grand was high (45.5%, 5/11). Therefore, physicians may consider the possibility of BCA in patients with cystic tumors in the deep lobe of the parotid gland.

FNAB is recommended for preoperative diagnosis [3, 13]. However, the accuracy of FNAB is limited by the tumor location and the number of extracted cells. Some BCAs are simply identified as benign tumors on FNAB, as they are difficult to distinguish from pleomorphic adenomas (PAs) [13]. In the present study, only 7 tumors were detected on FNAB, and 4 of these were identified as benign tumors. Thus, the rate of detection was lower than that previously reported [3]. However, considering that only 15 patients underwent FNAB examination, the rate of diagnosis on FNAB might be slightly higher than 50% (15/29). Therefore, we believe that wider use of FNAB can increase the rate of the preoperative diagnosis of BCA.

The surgical method was the same as that used for other benign parotid gland tumors. Facial nerve function was impaired (HB grade

II) postoperatively in five patients. At 6 months after surgery, three of these patients had recovered, while two patients continued to have facial paralysis. The incidence of facial nerve impairment was higher in the deep group than in the other two groups. This is because the facial nerve is more likely to be damaged during the resection of the deep lobe.

The clinical characteristics of BCA patients were similar to those of PA patients, but their pathological features were significantly different. BCAs consist of palisading basaloid cells and basement membrane thickening. According to the cell arrangement, BCAs can be divided into the solid type, solid tubular type, solid trabecular type and membranous type [3, 8] (**Figure 3**). It has been reported that BCAs of the salivary gland have a multifocal origin, and a case of malignant transformation has been reported in a patient with membranous-type BCA [14]. There was no recurrence or malignant transformation during the postoperative follow-up in the present study. This result may be attributable to the low number of membranous-type BCAs in our study.

A rare case of bilateral parotid BCAs was observed in our study. One of the tumors was located in the lower superficial lobe of the left parotid gland, and the other was located in the deep lobe of the right parotid gland (**Figure 1**). The mechanism of bilateral BCAs is unclear. It is assumed that these tumors were caused by genetic and environmental factors.

In conclusion, the clinical manifestations of parotid BCAs were consistent with those of other benign parotid tumors. There were no significant differences in age, average disease duration and tumor size among the three study groups. The diagnosis of BCA should be considered in patients with tumors located in the deep lobe of the parotid gland, especially when the tumor exhibits cystic degeneration. FNAB can increase the rate of preoperative diagnoses. These findings may facilitate the clinical diagnosis and treatment of BCA.

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Disclosure of conflict of interest

None.

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