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Original article

Infratemporal fossa tumors: When to suspect a malignant tumor? A retrospective cohort study of 62 cases

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ABSTRACT

Objectives: Infratemporal fossa (ITF) tumors are rare and little is known about their general epidemiology, making it sometimes difficult for clinicians, who seldom encounter them, to distinguish between benign and malignant forms on the basis of the initial clinical and radiological work-up alone. The objectives of this retrospective study were: (i) to determine the respective prevalences of the various histologic types of ITF tumor, and (ii) to assess associations between certain clinical and radiological features and malignancy.
Methods: A single-center observational study in a university hospital included all new consecutive cases of ITF tumor treated from January 2000 to December 2016. Histologic type, demographics, clinical presentation and imaging findings were analyzed.
Results: In total, 62 patients were included. 74% of tumors were benign ($n = 46$) and 26% malignant. Juvenile nasopharyngeal angiofibroma, adenoid cystic carcinoma and schwannoma were the most frequent histologic types, accounting for 47%, 16% and 10% of cases, respectively. The only clinical or imaging signs significantly associated with malignancy were trismus, facial pain, facial hypoesthesia and neural invasion on magnetic resonance imaging (all P-values < 0.05).
Conclusion: This study provides general epidemiological data on ITF tumors, and identified several clinical and radiologic signs to help clinicians suspect malignancy.

1. Introduction

The infratemporal fossa (ITF) is a deep facial space behind the zygomatic bone and the maxillary tuberosity; in Legent et al.'s classification, it is subdivided into 3 regions: pterygopalatine fossa, retro-maxillo-zygomatic space, and pterygoid muscle region [1].

Although many anatomic studies and surgical series have been published [2–5], there are no descriptive series of the respective frequencies of the various types of ITF tumor. Most surgical reports have focused on one particular histologic type, so as to have a homogeneous study population [2–6]. Some reported more varied populations [2,7,8], but none included non-operated cases. Thus, although the studies provide useful information on treatment, there are almost no data on the general epidemiology of ITF tumors. In particular, the proportions of benign and malignant forms and the clinical and imaging signs that distinguish them have never been reported in a large series.

The aims of the present study were therefore: (i) to estimate the distribution of histologic types in a retrospective series of 62 patients, both operated and non-operated; and (ii) to attempt to assess clinical and radiological signs of malignancy that could guide clinicians in IFT tumor work-up.

2. Material and methods

An observational study included all consecutive cases of ITF tumor, managed surgically or not, in the Lariboisière University Hospital, Paris, France, between January 2000 and December 2016. Only tumors originating from the ITF were included; those originating in adjacent regions with secondary invasion were excluded.

General sociodemographic data, clinical history, symptoms and clinical examination findings were collated. All patients had cervicofacial computerized tomography (CT) scan, with and without contrast enhancement, studying soft tissues and any bone involvement. Magnetic resonance imaging (MRI) was also systematic,

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comprising at least: T1-weighted sequences with and without gadolinium injection, T2-weighted sequences and fluid attenuated inversion recovery (FLAIR) sequences. MRI focused on precise extension within the ITF and any extraorbital and/or intracranial extension or neural invasion.

In malignant tumors, the imaging work-up was completed by PET scan and/or thoraco-abdominopelvic CT.

2.1. Statistical analyses

Malignant versus benign groups were compared on Fisher exact test or Pearson Chi^2 for qualitative variables, and Student *t*-test or Wilcoxon test for quantitative variables. Odds ratios (OR) and 95% confidence intervals (95% CI) were calculated. The significance threshold was set at 5%. Analyses used SPSS software (SPSS, Chicago, IL). To avoid bias due to the large number of juvenile nasopharyngeal angiofibromas (JNA), which show clinical and radiologic specificities, analyses were performed firstly on the whole cohort and secondly excluding JNA.

3. Results

3.1. Demographic data

Between January 2000 and December 2016, 62 patients were managed for tumor originating from the ITF. There was male predominance (69%) but, if JNAs ($n = 29$) were excluded, the remaining cohort comprised 42% male and 58% female patients. Mean age at diagnosis was 31 years (median: 20.5 years; range: 9–80); excluding JNA, mean age was 45 years (median: 43 years; range: 16–80).

3.2. Histology

Histologic type was determined either on biopsy ($n = 20$) or on surgical specimen ($n = 33$, including 29 JNAs). In 9 cases (14%), the tumor was paucisymptomatic and imaging found typical signs of fibrous dysplasia, schwannoma or pleomorphic adenoma; for typical imaging aspects, see [9–12]. In these cases, biopsy was not performed, and diagnosis was presumptive; the patients were followed up regularly, clinically and on imaging, to check progression and indicate biopsy or resection in case of anomaly. None of these 9 cases in fact required subsequent surgery.

Most tumors were benign ($n = 46$, 74%). The most frequent histologic types were JNA ($n = 29$, 47%), adenoid cystic carcinoma (ACC) ($n = 10$, 16%) and schwannoma ($n = 6$, 10%); fibrous dysplasia accounted for 6% ($n = 4$) and neurofibroma 3% ($n = 2$); for all other types, $n = 1$. These data are presented in Table 1.

3.3. Clinical presentation

Table 2 shows presenting symptoms. The most frequent were nasal obstruction, facial pain and otologic signs (otalgia, tinnitus and chronic otitis media), in 53%, 35%, 18% and 16% of cases, respectively.

3.4. Imaging

In 56% of cases, the left side was involved. 40% of the 62 ITF tumors showed intraorbital, 26% intracranial and 16% cavernous sinus invasion. Table 3 presents imaging data.

CT found bone erosion in 41 cases (66%), including 28 JNAs. The most frequent bone erosion sites were pterygoid apophysis root (80%), basisphenoid (54%) and posterior maxillary sinus wall (36%); in most cases, all 3 sites were involved. Erosion of the carotid canal, clivus, ascending mandibular branch or orbital floor was found in less than 10% of cases.

Table 1
Histologic types of ITF tumors.

Histologic type	Number of patients (%)
Benign	46 (74)
Juvenile nasopharyngeal angiofibroma	29 (47)
Schwannoma	6 (10)
Fibrous dysplasia	4 (6)
Neurofibroma	2 (3)
Cavernoma	1 (1.6)
Eosinophilic granuloma	1 (1.6)
Pleomorphic adenoma	1 (1.6)
Epithelioid hemangioendothelioma	1 (1.6)
Meningioma	1 (1.6)
Malignant	16 (26)
Adenoid cystic carcinoma	10 (16)
Chondrosarcoma	1 (1.6)
Metastasis (primary: pulmonary squamous cell carcinoma)	1 (1.6)
Solitary fibrous tumor (high grade)	1 (1.6)
Lymphoma	1 (1.6)
Rhabdomyosarcomas	1 (1.6)
Plasmocytoma	1 (1.6)

Table 2
Clinical presentation of ITF tumors.

Symptoms	Histologic type			*P*-value
	All	Malignant	Benign	
Nasal obstruction	33 (53)	4 (27)	29 (62)	ns
Epistaxis	22 (35)	2 (13)	20 (42)	ns
Facial pain	11 (18)	6 (40)	5 (11)	0.02
Otologic signs	10 (16)	3 (20)	7 (15)	ns
Facial hypoesthesia	5 (8)	4 (27)	2 (4)	0.03
Trismus	4 (6)	3 (20)	1 (2)	0.04
Asymptomatic	3 (5)	1 (7)	2 (4)	ns
Diplopia	1 (2)	1 (7)	0 (0)	ns
Ptosis	1 (2)	0 (0)	1 (2)	ns

Data are presented as number (percentage). Comparisons are between benign and malignant types. ns: non-significant (*P*-value > 0.05).

Table 3
Imaging data of ITF tumors.

	Histologic type			*P*-value
	All	Malignant	Benign	
Intraorbital extension	25 (40)	6 (38)	19 (41)	ns
Intracranial extension	16 (26)	3 (19)	13 (28)	ns
Cavernous sinus invasion	10 (16)	0 (0)	10 (22)	ns
Bone erosion	41 (66)	7 (44)	34 (81)	ns
Neural invasion	9 (15)	9 (56)	0 (0)	$< 10^{-5}$

Data are presented as number (percentage). Comparisons are between benign and malignant types. ns: non-significant (*P*-value > 0.05).

MRI found neural involvement in 9 cases, all ACC.

3.5. Signs associated with malignancy

Associations between clinical and imaging signs and malignancy were assessed.

Gender showed no association (OR: 0.46; 95% CI: 0.12–1.80). Advanced age showed an association (*P*-value < 0.001), which, however, was no longer significant when JNAs were excluded (*P*-value = 0.90).

Three of the clinical signs were significantly associated with malignancy: trismus, facial pain, and hypoesthesia in a trigeminal nerve territory (all *P*-values < 0.05). Table 2 presents these findings.

On imaging, intraorbital, intracranial and cavernous sinus invasion were not associated with malignancy (Table 3). Bone erosion likewise was not associated with malignancy (OR: 0.34; 95% CI: 0.08–1.32). JNA often underlies bone erosion due to pterygoid

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