

Oral Squamous Cell Carcinoma in Pediatric Moroccan Population: A Retrospective Study

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ABSTRACT

PURPOSE: Study the epidemiological and histopathological profile of the oral cancers in children in care at the Pediatric Hemato-Oncology, Stomatology and Maxillofacial Surgery departments at the 20 August hospital in Casablanca, and at the hospital of children in Rabat. Our aim is to define the importance of the pediatric oncology, especially the one affecting the oral cavity, and also to describe the oral cancers in children, their frequencies and their histopathological characteristics. We did collect 71 patients consultation records and files between 2004- 2012. **MATERIALS AND METHODS:** This is a retrospective study of 126 children hospitalized between 2010 and 2013 of the Pediatric Hemato-Oncology Department, Stomatology and Maxillofacial Surgery department at the 20 August hospital in Casablanca, and also in the Pediatric Hemato-Oncology Department at the hospital of children in Rabat, and in which we did diagnose a confirmed cancer of the oral cavity. **RESULTS:** In our sample, all age groups were affected by the disease process, but ages between [0-4] years and between [13-16] were the most affected with an average of age of 8 years, and extremes ranging from 4 months to 16 years. In our population sex sample, we noted a slight female predominance with 50.7% of cases. Non-Hodgkin lymphoma Burkitt was the most common histological type with 35.2% of cases. Cheeks represented the most frequent localization with 37.9% of cases, while the maxillary represented 19.7% of cases. Chemotherapy has been the exclusive therapeutic strategy most used in our sample in 67.6% of cases. **CONCLUSION:** Epidemiological, clinical and pathological characteristics of cancers of the oral cavity in our population are not different from the literature data. However, the parents lack of awareness and late diagnosis of these lesions appear to be responsible for the dramatic profile of oral cancers.

KEYWORDS: Children, Oral Cancer, Oral Cavity

INTRODUCTION

Squamous cell carcinoma (SCC) of the head and neck is uncommon in young adults, and rare in the pediatric age group. Approximately 1 in 1000 cases of head and neck SCC will occur in patients 20 years or younger.^{1,2} While there is an extensive literature examining outcomes of SCC in young adults (<40 years), to our knowledge, there is no literature on SCC in pediatric patients, other than isolated case reports.^{1,3-5} Squamous cell carcinoma in young patients is believed to be etiologically distinct from SCC in older adults owing to less significant exposure to risk factors such as tobacco and alcohol. This distinction in etiology would be especially pronounced in pediatric patients, in whom genetic syndromes such as Fanconi anemia, xeroderma pigmentosum, keratosis-ichthyosis-deafness (KID) syndrome, or other unidentified genetic risk factors may be contributory.⁵⁻⁷

Numerous early reports of SCC in young adults concluded that the disease was more aggressive, and prognosis was poorer, than in older adults.^{8,9} Several recent studies, however, have provided evidence for the

contrary.^{10,11} To our knowledge, outcomes data specific to the pediatric age group have not been reported and represent a missing piece of the literature on head and neck SCC in young patients.

In Moroccan young patients with head and neck SCC, the cheek seems to be the most common primary site.¹²⁻¹⁴ We report the outcomes of a pediatric retrospective study of patients with OSCC who were treated at a comprehensive cancer center.

MATERIALS AND METHODS

This is a retrospective study of 1070 Moroccan children records; 620 in Rabat Hospitals and 450 in Casablanca Hospitals, hospitalized between 2010 and 2013 in the Pediatric Hemato-Oncology Department, Stomatology and Maxillofacial Surgery department at the 20 August hospital in Casablanca, and also in the Pediatric Hemato-Oncology Department at the hospital of children in Rabat, and in which we did diagnose a confirmed cancer.

Only 126 records were selected for the study, patients with cancer of the oral cavity confirmed by histology. In

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the absence of records of some patients, it was based on the computerized database for the leadership of the service. Were excluded from the study all patients with pathological examination which did not allow to define the nature of cancer, patients with cancer in the nasopharynx, maxillary sinus and the parotid gland, patients having a cancer other than cancers of the oral cavity; and patients with benign tumors of the oral cavity.

Analysis of the results was performed using the software Statistic Package for Social Science (SPSS Version 13.0) for Windows. The implementation of graphics was done with Excel 2007 software. Quantitative variables were expressed as mean \pm standard deviation. Categorical variables were expressed as effective and valid percentage (%).

The comparison of categorical variables was performed using the "chi."2 Test. The difference is considered statistically significant if the p value is strictly less than 0.05.

RESULTS

For the annual prevalence, the year 2006 has seen a significant increase in the number of children with cancers of the oral cavity reported to August 20 Hospital in Casablanca, with a rate of 16.9% (Tab 1). However, the study conducted at the hospital of children of Rabat in 2012 found that the largest annual prevalence was marked in 2009 with a rate of 16.4%.

Year	Percentage %
2004	9.9
2005	9.9
2006	16.9
2007	15.5
2008	9.9
2009	11.3
2010	15.5
2011	7
2012	4.2

Table 1 : Annual Prevalence of OSCC Patients

About age in the 71 cases studied, all age groups were affected by the disease process, but ages from [0-4] years to [13-16] were the most affected with an average age of 8 years and extremes ranging from 4 months to 16 years (Tab 2). A study in Morocco carried out by Arfaoui & al in 2008, found that the age group [10-14] years was the most affected by cancer with 41.3% of cases. However, Tunisia Khiati & al in 2002 noted that the most affected age group is that of [0-4] years, with 39.8% of cases.

For the sex of our study population, there was a slight female predominance with 50.7% of cases (Tab3). Our results are similar to those found in the cancer records of the Grand Casablanca region in 2004, where females represented 53.8% of cases while Arfaoui & al in 2008 noted that the male represents 60.3% of cases.

Age Brackets (Years)	Percentage %
(0-4)	26.8
(5-8)	23.9
(9-12)	22.5
(13-16)	26.8

Table 2 : Age Distribution in OSCC Patients

Sex	Percentage %
Male	49.3
Female	50.7

Table 3 : Sex Distribution in OSCC Patients

The study of the employment situation in our children patient parents sample showed a significant majority of fathers without occupation and workers who accounted for 31.7% of cases, while 92.1% of mothers are housewives (Tab 4: a & b).

Fathers Profession	Percentage %
Unemployed	19
Worker	31.7
Liberal Profession	17.5
Public Profession	11.1
Farmer	14.3
Defunct	6.3

Table 4a: OSCC Patients fathers Professions

Mothers Profession	Percentage %
Housewives	92.1
Employee	4.8
Defunct	3.2

Table 4b : OSCC Patients Mothers Profession

In London, Saman & al in 2009 reported the highest rates of cancer in disadvantaged population groups. It is clear from these results that there is a close link between the occurrence of cancers of the oral cavity and the socioeconomic status of patients.

For the consultation period, patients who consulted in the first 4 months have been the majority of patients in our study and accounted for 80.3% of cases (Tab 5).

Age Brackets (Months)	Percentage %
(0-4)	80.3
(5-8)	9.9
(9-12)	9.9

Table 5 : Consultation period in OSCC Patients

Our results are comparable to those found at the Rabat children hospital where patients seen during the first four months accounted for 45.4% of cases. Nevertheless,

Donaldson & al in 2004 in the United States, found in his study that the consultation period was 3 to 6 months.

In Malaysia, the consultation period for Ajura in 2010 was between 1-5 months.

For the histological appearance of the tumor, non-Hodgkin lymphoma Burkitt (NHL) was the most common histological type with 35.2% of cases (Tab 6) . Our results are comparable to those of the children hospital in Rabat in 2012 where non-Hodgkin's lymphoma Burkitt accounted for 65.7% of cases. Similarly, in Nigeria, Ajayi & al in 2007 reported that lymphoma Burkitt was more frequent with 53.2% of cases.

Histological feature of the Tumor	Incidence (%)
Burkitt Malignant Non Hodgkin Lymphoma (MNHL)	35.2
Lymphoblastic Lymphoma T	1.4
Accurate Lymphoblastic Leukemia (ALL)	8.5
Rambdomyosarcoma (RMS)	16.9

Table 6 : Histology Feature of the Tumor in OSCC patients

For the location of the tumor, cheeks represented the most frequent localization with 37.9% of cases, while the maxillary only represented 19.7% of cases (Tab 7). However, in the hospital of children of Rabat, the mandible was the most frequent location with 37.1% of cases followed by tonsils with 25.7%.

Tumor Location	Incidence %
Maxillary	19.7
Mandible	15.5
Tonsil	21.1
Cheeks	37.9
Gum	21.1
Palate	4.2
Tongue	2.8
Lips	14

Table 7 : Tumor Location in OSCC Patients

Exclusive Chemotherapy is the most treatment strategy used in our sample in 67.6% of cases (Tab 8). Our results are similar to those found in the hospital of children Rabat where chemotherapy was performed in 69.1% of cases. In black Africa, Tonoh-Anoh & al in 2000, found that more than 50% f the cases were treated by chemotherapy alone. According to Khalifa & al in 2008, malignant diseases in children are to overwhelmingly sensitive to cytotoxic chemotherapy. That chemosensitivity is generally attributed to the rapidly proliferating character of these tumors and their ability to undergo apoptosis.

In our study, we found that 41, 9% had a complete remission. And complete remission was revealed in 72%

of cases of Burkitt's NHL. Our results are comparable to those of Rabat children hospital who revealed that 67.7% of cases of Burkitt NHL had a complete remission.

A study in Morocco carried out by Otmani & al in 2008, found that the complete remission was observed in 59% of cases of NHL Burkitt. Similarly Gentet & al in 2002 noted in his study that the complete remission was observed in 90.7% of cases of Burkitt's NHL (Tab 9).

Therapeutic Strategies	Percentage %
Chemotherapy	67.6
Surgery	14.1
Radiotherapy	1.4
Chemotherapy + Surgery	8.5
Chemotherapy + Radiotherapy	4.2

Table 8 : Incidence of Therapeutic Strategies on OSCC patients

Therapeutic Results	Incidence %
Complete Remission	41.9
Local Recurrence	3.2
Relapse Remote	9.7
Dead	17.7
Disappeared patient	19.7
Local + Distant Recurrence	4.8

Table 9: Treatment Outcome on OSCC Patients

DISCUSSION

Most malignant tumors in the head and neck region in childhood have a non-epithelial origin.¹⁵ In a nineteen year retrospective study, Cra Rapidis et al. reviewed 1007 tumors of head and neck in children and found that 308 (31%) were malignant. The malignant tumors included 52% lymphoma, 22.3% rhabdomyosarcoma, 5.8% squamous cell carcinomas, 5.8% neuroblastoma and 4.2% other sarcomas.¹⁶ In an extensive review of literature by various authors, only 4– 14% cases of SCC were reported below 40 years of age.¹⁷⁻¹⁹ Krolls et al.²⁰ in a study of 14,253 cases of squamous cell carcinomas found only 0.20% cases (n = 19) below 19 years of age while just 0.05% (n = 3) were found below 14 years of age.

According to the available literature until 1989, only 22 cases of SCC had been reported below 15 years of age; after that, until 2009, ten more such cases had been reported by various authors.^{15-19,21} Only 7 patients, in all these studies, were below 10 years of age.

The tongue is the most commonly reported site followed by the lip and cheek in pediatric squamous cell carcinoma.²¹⁻²³ The maxillary alveolus is involved in SCC in less than 10% of all oral carcinomas in adult population.²² Only 4 such cases of squamous cell carcinoma involving the maxillary alveolus in children below 10 years of age have been reported in the literature.^{15,24,25}

It is documented that there is equal gender predilection in pediatric patients with oral SCC but in the older population it is more common in males.^{23,24}

In the adult population, smoking, alcohol and tobacco chewing are etiological factors for SCC, but in the pediatric population, some genetic conditions like Fanconi syndrome, Xeroderma pigmentosa, Keratitis, Ichthyosis and Deafness syndrome; chewing and sucking of plastic containing polyvinyl chloride, which is carcinogenic in nature and exposure to the human papilloma virus have been suggested to be etiological factors⁽²²⁾. None of these etiologic factors were present in our study. Also in younger populations, SCC is thought to be more aggressive in nature with poorer prognosis, in comparison with older population, due to differences in etiology and histopathology.^{16,18,23} So it demands early diagnosis and definitive treatment.

Clinically SCC in younger patients can be misdiagnosed with lesions like Pseudo epitheliomatous hyperplasia, (this epithelial inflammatory lesion presents as a highly aggressive proliferative lesion with surface ulceration in the pediatric population), Solitary intraoral keratoacanthoma and Pyogenic granuloma (this presents as a rapidly growing, sometimes ulcerated overgrowth located on the surface of the gingiva).^{16,23,25,26} Histopathologically, there is a similarity between pseudoepitheliomatous hyperplasia and SCC. These lead to delay in diagnosis and treatment leading to change in prognosis.

Review of the literature suggests that chemotherapy and wide surgical resection are the preferred treatment modalities as radiotherapy in the children age group may have a significant negative impact on facial growth and soft and hard tissue development as well as increasing the risk of developing a second malignancy.^{22,24,27,28}

CONCLUSION

This study, together with previously reported studies and cases, indicates that oral SCC can occur in pediatric patients. SCC should not be overlooked as one of the differential diagnoses of oral lesions in pediatric patients as prognosis of the disease can be altered by early diagnosis and surgical treatment.

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