

Frequency of head and neck masses in the Iranian infants during a 21-year period

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Abstract

Aim: Head and neck masses are common clinical findings in infants, children and adolescents. This retrospective study aimed to assess the frequency of head and neck masses in children younger than 2 years of age.

Materials and methods: All medical files present in the Pathology Department of Children's Medical Center in Tehran from 1995 to 2016 with the pathology report confirming head and neck mass in children 0-2 years of age were evaluated in this study. The masses were divided into three groups of congenital/developmental, inflammatory/reactive/infectious and neoplastic (benign/malignant) lesions. Data were analyzed using SPSS® version 22.

Results: Among 15,572 children aged 0-2 years presenting to the Children's Medical Center in Tehran, Iran, 413 patients (2.7%) showed head and neck masses with a higher frequency in males (1.2:1; $p = 0.000$). About half of the lesions were in the neck, and about one-third of the remaining lesions were in the oral cavity and salivary glands. Most lesions were congenital (40.7%) or inflammatory (34.9%). Hemangioma was the most common benign tumor. Among congenital lesions, dermoid cyst and branchial cleft cyst and among inflammatory lesions, lymphadenitis

and lymphoid tissue hyperplasia were the most common. The overall prevalence of malignant neoplasms was 3.6%, and rhabdomyosarcoma had a high frequency. Mucocele was the most common oral inflammatory mass.

Conclusion: The results showed variations in the frequency of head and neck masses, especially malignancies, in children under the age of 2 compared with older children. Since children comprise one-fourth of the population, the frequency of head and neck masses must be separately determined for each age group.

Keywords

Mass, head, neck, infant.

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Introduction

Children comprise a significant portion of the population of developing countries [1]. According to the Food and Drug Administration, babies in their first 28 days of life are referred to as neonates, the time period from 29 days to less than 2 years is referred to as infancy and the time from 2 years to less than 12 is referred to as childhood [2]. Head and neck masses are common clinical findings in infants, children and adolescents. The differential diagnosis of these masses includes congenital, inflammatory and neoplastic (benign/malignant) lesions [3]. The prevalence of cancer increases by 1% to 2% annually; however, the prevalence of head and neck tumors in children, especially in infants, has not been well evaluated, and there is a gap of information in this respect [4]. In Iran, two studies assessed the head and neck tumors in children and both studies reported that non-Hodgkin lymphoma was the most common malignant tumor. The frequency of

malignant tumors was almost the same (85.5% and 89.5%) [4, 5]. On the other hand, another study carried out by us about the head and neck masses in children between 2-12 years showed that although lymphoma was the most common malignant lesion, the type of lymphoma was different [6].

Some studies have assessed the prevalence of brain tumors in children under the age of 2, but no study has evaluated the epidemiology of head and neck masses in this age group [7]. Considering the available statistics and the role of geographical location and ethnicity in the occurrence of these lesions, this study aimed to determine the frequency of head and neck tumors in infants presenting to the Children's Medical Center in Tehran, Iran (which is a referral center for children) from 1995 to 2016.

Materials and methods

In this descriptive, retrospective study, medical files of patients aged 0-2 years presenting to the Children's Medical Center in Tehran from 1995 to 2016 with the diagnosis of head and neck masses were evaluated. The Children's Medical Center in Tehran is a referral center for children with specific diseases such as cancer in Iran. Medical files with complete patient information and accurate histopathological report of the lesion were included. The exclusion criteria were intra-orbital masses, intracranial masses and central nervous system lesions. Age and gender of patients, location of the mass and histopathological diagnosis were all evaluated and recorded. Slides were re-evaluated to confirm the diagnosis. Data were analyzed using SPSS® version 22. The patients were divided into two age groups of 0 to < 1 and 1 to < 2 years. The masses were divided into three groups of congenital/developmental, inflammatory/reactive/infectious and neoplastic (benign/malignant) lesions.

Ethical considerations

This study was approved by the ethics committee of Tehran University of Medical Sciences with project number 1396.2169.

Results

Among a total of 15,572 children aged 0-2 years presenting to the Children's Medical Center

in Tehran from 1995 to 2016, 413 (2.7%) showed head and neck masses with a higher frequency in males (1.2:1, $p = 0.000$). The frequency of patients aged 0 to < 1 and 1 to < 2 years was 65.9% and 34.1%, respectively. Among all lesions, the frequency of the lesions in the neck, oral cavity and salivary glands was 51.35%, 11.45% and 4.35%, respectively. The highest frequency belonged to congenital masses (40.7%), followed by inflammatory lesions (34.9%). The prevalence of benign and malignant tumors was 20.8% and 3.6%, respectively (**Fig. 1**).

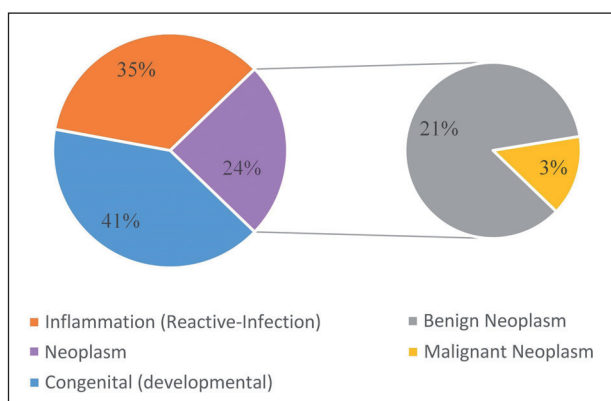


Figure 1. Frequency of different groups of head and neck masses in children under the age of 2.

Among congenital masses, the most frequent masses were the dermoid cyst (20.8%), the branchial cleft cyst (19%) and the cystic hygroma (17.3%) (**Tab. 1**). In this group, 71% of the patients were < 1 year of age and 56.5% were males. The most frequent lesion in patients aged 0 to < 1 and 1 to < 2 years was dermoid cyst and thyroglossal duct cyst, respectively.

Among inflammatory masses (reactive/infectious), the most frequent masses were lymphadenitis (22.9%), lymphoid tissue hyperplasia (22.9%) and epidermoid cyst (18.1%) (**Tab. 2**). Boys were more commonly affected than girls, and 63% of the patients were < 1 year of age.

Among benign neoplasms, hemangioma was the most common (44.2%), followed by Langerhans cell histiocytosis (15.1%) and lipoma (10.5%). Also, rhabdomyosarcoma (40%) followed by neuroblastoma (26.7%) and lymphoma (20%) were the most common malignant masses. The three cases of lymphomas included two non-Hodgkin and one Hodgkin lymphoma (**Tab. 3**).

Table 1. Frequency distribution of gender and location of congenital lesions in children under the age of 2 years.

Category	Pathologic diagnosis (n)	Sex (n)		Location (n)											
		Female	Male	Neck	Face	Ear	Nose	Head	Naso pharynx	Soft tissue	Mouth	Oro pharynx			
Congenital (developmental)	Dermoid cyst (35)	17	18	5	19	0	1	10	0	0	0	0	0	0	0
	Branchial cleft cyst (32)	11	21	31	0	0	0	1	0	0	0	0	0	0	0
	Cystic hygroma (29)	15	14	29	0	0	0	0	0	0	0	0	0	0	0
	Accessory ear lobe (25)	13	12	0	0	25	0	0	0	0	0	0	0	0	0
	Thyroglossal duct cyst (13)	4	9	13	0	0	0	0	0	0	0	0	0	0	0
	Teratoma (9)	4	5	4	0	0	0	0	1	3	1	0	0	0	0
	Lymphangioma (7)	2	5	4	0	0	0	0	0	0	3	3	0	0	0
	Salivary duct cyst (5)	3	2	0	0	0	0	0	0	0	0	4	0	5	0
	Oral lymphoepithelial cyst (4)	0	4	0	0	0	0	0	0	0	0	0	0	0	0
	Congenital epulis (3)	3	0	0	0	0	0	0	0	0	3	3	0	0	0
	Oral epidermoid cyst (3)	0	3	0	0	0	0	0	0	0	0	3	0	0	0
	Vascular malformation (1)	0	1	0	0	0	0	0	0	0	0	1	0	0	0
	Congenital staelectasia (1)	0	1	0	0	0	0	0	0	0	0	0	0	1	0
Eruption cyst (1)	1	0	0	0	0	0	0	0	0	0	1	0	0	0	
Total (n = 168)		73	95	86	19	25	1	12	3	16	6	0	0	0	

Table 2. Frequency distribution of gender and location of inflammatory lesions in children under the age of 2 years.

Category	Pathologic diagnosis (n)	Sex (n)		Location (n)											
		Female	Male	Neck	Face	Ear	Nose	Head	Naso pharynx	Mouth					
										Soft tissue	Salivary gland	Oro pharynx			
Inflammatory (reactive-infection)	Lymphadenitis (33)	12	21	29	0	3	0	1	0	0	0	0	0	0	0
	Hyperplastic lymphoid (33)	15	18	27	0	2	0	4	0	0	0	0	0	0	0
	Epidermoid cyst (26)	14	12	4	19	0	0	3	0	0	0	0	0	0	0
	Epidermoid inclusion cyst (8)	5	3	0	5	0	0	3	0	0	0	0	0	0	0
	Inflammation (19)	9	10	13	0	0	0	0	0	0	6	0	0	0	0
	Mucocele (11)	6	5	0	0	0	0	0	0	0	0	11	0	0	0
	Abscess (9)	5	4	8	0	0	0	0	0	0	1	0	0	0	0
	Hyperplastic adenoid (8)	1	7	0	0	0	0	0	0	0	2	0	0	0	6
	Xanthogranuloma (3)	2	1	1	1	0	0	1	0	0	0	0	0	0	0
	Irritation fibroma (2)	1	1	0	0	0	0	0	0	0	2	0	0	0	0
Total (n = 144)	65	79	82	20	5	0	9	0	0	11	11	6			

Also, 58% of the children were < 1 year of age, in this group of lesions the neck (43.5%) was the most common place of involvement.

Among 64 masses arising from the oral cavity (46 cases) and salivary glands (18 cases), inflammatory (43.8%) and congenital (34.4%) lesions had the highest frequency, and mucocele (17.2%) followed by adenoid tissue hyperplasia (12.5%) were the most common lesions. Also, 73% of the patients with oral cavity lesions were < 1 year of age. Rhabdomyosarcoma in a 1-year-old girl was the only malignant lesion in this area, found in the parotid gland.

Discussion

Epidemiologic studies based on histopathologic findings are necessary to achieve the true incidence of the lesions in different ages [8]. Numerous lesions can occur in the head and neck region in children, and the differential diagnosis of these lesions is highly important. On the other hand, early diagnosis of neoplastic lesions, especially in children, is necessary because it can increase the opportunity for early treatment and better response to treatment [9]. However, in our study, the most common head and neck masses in children aged 0-2 years were congenital lesions and only 3% of the lesions were malignant. In our study, the prevalence of head and neck masses in this age group was 2.7% with a significantly higher prevalence in males (1.2:1) and a mean age of 10 months compared with all patients with a mass evaluated in the Pathology Department of Children's Medical Center. The male to female ratio in studies by Showkat et al. and Lucumay et al., in India and Tanzania, was 1.2:1 and 2.5:1, respectively [10, 11]. This ratio was 1.7:1 in a previous study performed by our group about head and neck masses in children between 2-12 years and also in a study by Ragesh et al., in India [6, 12]. The frequency of involvement in boys in their study was higher compared with the frequency observed in our cohort of patients, but it could be due to the smaller size of their cohort (n = 50) [12] or the difference in the age range of the study populations [6, 12]. Similar to our study, in the study performed by Shengwei et al., in China, the most common lesions were congenital, followed by inflammatory lesions [13]. In the study by Lucumay et al., in Tanzania, congenital masses accounted for 85% of cervical masses in children under the age of 2 [11]. In the study by Ragesh et

Table 3. Frequency distribution of gender and location of neoplastic lesions (benign/malignant) in children under the age of 2 years.

Category	Pathologic diagnosis (n)	Sex (n)		Location (n)									
		Female	Male	Neck	Face	Ear	Nose	Head	Naso pharynx	Mouth			
										Soft tissue	Salivary gland	Oro pharynx	
Neoplasm (benign)	Hemangioma (38)	22	16	13	9	2	0	9	0	5	0	0	
	Langerhans cell histiocytosis (13)	5	8	5	1	0	0	5	0	2	0	0	
	Lipoma (9)	6	3	7	0	1	0	1	0	0	0	0	
	Hamartoma (8)	3	5	2	1	1	0	0	0	4	0	0	
	Piliomatixoma (4)	1	3	1	2	1	0	0	0	0	0	0	
	Hemangiomaendothelioma (3)	0	3	2	0	1	0	0	0	0	0	0	
	Dermatofibroma (3)	0	3	0	0	0	0	3	0	0	0	0	
	Skin tag (2)	1	1	0	0	1	0	0	0	1	0	0	
	Fibromatosis (1)	1	0	1	0	0	0	0	0	0	0	0	
	Angiofibroma (1)	1	0	0	0	0	0	1	0	0	0	0	
	Benign spindle cell tumor (1)	1	0	1	0	0	0	0	0	0	0	0	
	Chondroma (1)	0	1	1	0	0	0	0	0	0	0	0	
	Fibromatosis colli (1)	0	1	1	0	0	0	0	0	0	0	0	
	Granular cell tumor (1)	1	0		0	0	0	0	0	1	0	0	
	Neoplasm (malignant) Miscellaneous	Rhobdomyosarcoma (embryonal type) (6)	3	3	3	0	0	1	0	1	0	1	0
Nourblastoma (4)		0	4	3	0	1	0	0	0	0	0	0	
Lymphoma (3) Non-Hodgkin T-cell type (2) Hodgkin (1)		1	2	3	0	0	0	0	0	0	0	0	
Leukemia (acute myeloblastic) (1)		0	1	0	0	1	0	0	0	0	0	0	
Post transplantation lympho proliferative disease (malignant neoplasm) (1)		0	1	1	0	0	0	0	0	0	0	0	
Total (n = 101)	46	55	44	13	9	1	19	1	13	1	0		

al., in India, and in another study performed by our group, inflammatory lesions were the most common, while congenital lesions ranked second. This can be due to the different age ranges of the study populations [6, 12]. In a study by Shah et al., in India, dermoid cyst (18.1%) and branchial cleft cyst (18.1%) were the most common congenital lesions of the head and neck in children younger than 1 year of age [14]. These values were 20.8% and 19% in our study, respectively. In a study by Yoon and Park in South Korea, dermoid cyst (18.1%) after hemangioma was the most common head and neck lesion in children younger than 3 years of age [15]. In studies carried out by Ragesh et al., about head and neck masses in children in India, and Lucumay et al., about neck masses in Tanzania, cystic hygroma was the most common congenital lesion (8% and 18.2% prevalence rates among the total lesions, respectively) [11, 12]. This lesion in our study ranked third in terms of prevalence (17.3% among the congenital lesions). In our study, dermoid cyst (20.8%) was the most frequent congenital lesion. In the above-mentioned studies, the prevalence of dermoid cyst was low (4%) [11, 12]. The reason for different results may be the different number of cases evaluated in each study. On the other hand, in a study performed by Lucumay et al., only neck masses were evaluated, since the neck is a common place for cystic hygroma [11]. The dermoid cyst has a high prevalence in the head and scalp compared with the neck [15]. Therefore, the frequency of this cyst may be different in studies carried out only on the neck or head regions [11, 15]. In our study, although the neck was the most common place for the congenital lesions (86 out of 168 cases), and the dermoid cyst was the most common lesion in this group, only 14.3% of dermoid cysts were observed in the neck. In a study performed by Showkat et al., in India, and our previous study on children between 2-12 years, the most common congenital lesion was the thyroglossal cyst (6.5% and 11%, respectively, among all lesions) while in the current study, this lesion had a prevalence of 7.7% among the congenital lesions. The age range of patients in the aforementioned studies was wider, and the thyroglossal cyst was often diagnosed in children older than 2 years of age [6, 10].

In most previous studies, lymphadenitis and lymph node hyperplasia were the most common inflammatory lesions with similar prevalence rates [6, 12, 13], which was in line with our

findings. This shows that the frequency of inflammatory lesions in children and infants is approximately the same. In studies by Showkat et al. and Lucumay et al., the most common lesion was lymph node hyperplasia followed by cervical abscess [10, 11].

In line with our results, Yoon and Park, in their studies in South Korea, reported that the most common benign tumors (aside from congenital lesions) in 2- and 3-year-old children were hemangioma and Langerhans cell histiocytosis [15]. In a study by Saravani et al., conducted in Iran, the most common benign neoplasm of the maxillofacial region in children under the age of 5 was also hemangioma [16]. Most studies conducted on maxillofacial and head and neck lesions in children with a wider age range reported that hemangiomas were the most common benign neoplasms aside from the congenital and hard tissue (bone and teeth) lesions [4]. The prevalence of Langerhans cell histiocytosis in young children was very low or zero in other studies. However, in the study by Saravani et al., only the tumors of the oral and maxillofacial regions were evaluated [16]. Langerhans cell histiocytosis often involves the scalp and lymph nodes of the head and neck region. This explains the lower prevalence of this lesion in the aforementioned study. It should be noted that, in our study, 77% of the cases of Langerhans cell histiocytosis involved the neck and scalp.

In a study by Abdulai et al. conducted in Ghana, the prevalence of malignant tumors relative to all tumors in children under 5 years and under 3 years of age was 10% [17]. This value was close to the value obtained in our study (18%). Also, in a study conducted by Khademi et al., in Iran, the prevalence of malignant tumors in children aged 1 to 4 years was 95%. High frequency of malignancy in their study was probably due to the fact that the collected data mainly belonged to patients referred to the Oncology Department of Namazi Hospital in Iran (a referral center for pediatric cancer), which were mainly cases of malignancy. Moreover, children under the age of 1 were not included in their study, which may further increase the frequency percentage of malignancy reported in their study [5].

In studies by Adeyemo and Okolo and by Sengupta et al. conducted in South Africa and India, respectively, the most common malignancies in children under 5 years were lymphoma, followed by rhabdomyosarcoma [18, 19]. In the study by

Adeyemo and Okolo on infants younger than 1 year, the prevalence of rhabdomyosarcoma was higher than lymphoma, and neuroblastoma had a lower prevalence, but it was only reported in patients younger than 5 years [18]. In the study by Sengupta et al., on infants under 1 year of age, lymphoma and neuroblastoma had a higher frequency with equal prevalence rates followed by rhabdomyosarcoma [19]. It should be noted that neuroblastoma in our study was only seen in infants under 1 year of age. In a study by Albright et al., which was conducted according to the Surveillance, Epidemiology, and End Results Program, aside from the eye tumors, the most common malignancies in children under 1 and between 1 to 5 years of age were neuroblastoma and rhabdomyosarcoma, respectively [20]. Fattahi et al., in Iran, reported that lymphoma was the most common malignant tumor in children under the age of 12 years [4]. Abdulai et al. evaluated children younger than 16 years and reported that lymphoma, nasopharyngeal carcinoma and rhabdomyosarcoma were the most common [17]. In the study by Gosepath et al., in Germany, rhabdomyosarcoma, thyroid carcinoma and lymphoma were the most common cancers in patients under the age of 15 years. In their study, 52% of rhabdomyosarcomas occurred in patients younger than 5 years, and 67.3% of thyroid carcinomas occurred in patients between 10 to 15 years, which indicated a higher frequency of rhabdomyosarcoma and a lower frequency of thyroid carcinoma in young children [21]. In a study by Khademi et al., in Iran, the most common malignancies in children and young adults between 1 to 19 years were lymphoma and squamous cell carcinoma [5]. A possible reason for different frequency rates reported in previous studies and ours may be due to a wider age range of patients evaluated in previous studies and the effect of malignancies that are more common in older ages. According to this theory, Cunningham et al. showed that all thyroid carcinomas and nasopharyngeal cancers had occurred in patients over 6 years of age [22]. In the study by Albright et al., thyroid carcinomas were more commonly seen in the intermediate group of children with the highest mean age of 15.3 years [20]. In the study by Cunningham et al., the most common malignant tumors in children and young adults under the age of 19 years were lymphoma and rhabdomyosarcoma. The prevalence of neuroblastoma in their study was 5%, and all

cases were below 5 years of age with a mean age of 1.9 years; also, all cases of lymphomas were older than 2 years of age [22].

Conclusion

This study's results showed the different incidence rates of head and neck masses, particularly malignancies, in children under the age of 2 years compared with the values in older children reported in the literature. Also, variability exists in the prevalence rates for masses in different geographical locations and different populations. Considering the absence of a specific study on children younger than 2 years of age as well as the fact that children account for one-fourth of the population, the frequency of head and neck tumors must be evaluated in specific age groups of children to better elucidate this topic and find more reliable results.

Declaration of interest

There is no conflict of interest to declare. Financial support and sponsorship: none.

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