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Demographic and clinical criteria of intranasal lobular capillary hemangioma: as retrospective multicentric audit

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Abstract

Introduction: Lobular capillary hemangioma (LCH) is a benign vascular tumor that rarely affects the nasal cavity. Pregnancy and previous nasal trauma or surgery might be implicated in the pathogenesis of this tumor. However, in the majority of the cases, there is no identified cause. The objective of this study was to characterize the numerous demographic and clinical aspects of intranasal LCH and to find whether there is a link between its likely causation and other parameters. We retrospectively reviewed the medical records (from 2012 to 2021) of subjects with proven cases of intranasal LCH on histopathological evaluation at four main hospitals in Baghdad, Ramadi, Tikrit, and Samawah cities, Iraq. The detailed information concerning the age, gender, duration, clinical features, side, location, type of anesthesia, follow-up period, and recurrence was registered for each participant.

Results: Of the 82 patients, there were 73.2% females. The majority of the cases (79.3%) were from the age group of 18–40 years. The majority of the patients (74.4%) were seen within 1–2 months. Most of the cases originated from the left side (59.8%) and the nasal septum (81.7%). Epistaxis was the most common presenting symptom (91.5%). No cause was identified in 42.7% of the cases, followed by pregnancy (31.7%) and trauma (25.6%). There was a significant difference between the possible cause and the age and gender (P -value = 0.000). The recurrence rate was 3.66%.

Conclusion: Intranasal LCH shows a female predominance and mostly affects the age group of 18–40 years. Most of the cases involved the left side and nasal septum. Epistaxis was the chief complaint in the majority of cases. Age and gender can determine the possible cause of the lesion.

Keywords: Pyogenic granuloma, Lobular capillary hemangioma, Epistaxis, Nose, Nasal septum

Background

Nasal pyogenic granuloma, also known as lobular capillary hemangioma (LCH), is first reported by Mills et al. in 1980 [1]. It is a benign vascular proliferation of unknown causes that can arise in the nasal cavity. Acceptance of the name LCH over pyogenic granuloma goes back to the fact that the lesion did not develop in response to an infectious stimulus and also to the distinctive configuration of the capillary network in edematous lobules.

Despite epistaxis being a common presentation, nasal obstruction or pain may occur depending on the size of the lesion. Previous nasal trauma or surgery and pregnancy could be implicated in the pathogenesis of LCH. However, in the majority of intranasal LCH cases, there is no identified predisposing factor [2]. Many previous studies addressed the disease from the clinical, pathological, and therapy perspectives [2–4] or as case reports [5, 6] without concentrating on correlating the many parameters in the etiology, gender predominance, or age of incidence. This may be due to either a small number of cases [7] or due to studying nasal localization as part of the whole-body incidence of pyogenic granuloma [8]. Hence, we conducted this study at four centers to assess

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the different demographic and clinical characteristics of intranasal LCH and to determine if there is a correlation between its possible cause and various variables.

Patients and methods

This cross-sectional retrospective study was conducted at four hospitals in Baghdad (Al-Yarmouk Teaching Hospital), Ramadi (Al-Ramadi Teaching Hospital), Tikrit (Salahaddin General Hospital), and Samawah (Al-Hussein Teaching Hospital) cities, Iraq. The study covered a period of 10 years from 2012 to 2021. The study was approved by the Ethical Approval Committee of the University Of Anbar (reference number 18 on 18 April 2022). Patients with proven cases of LCH (histopathological characteristic arrangement of the capillary network in edematous lobules as described by Mills et al.) [1] of the nasal cavity of any age and of both sexes were enrolled in the current study. Patients with incomplete data, who lost to follow-up immediately following surgery, and those with other intranasal masses not proved to be LCH were excluded from the present study.

Data from the patients' medical records were collected, including demographic information (age and gender), clinical information (duration, clinical features, side, and site of the lesion), possible causes such as a history of nasal trauma or surgery, and anesthesia type (local or general). Radiological investigations in the form of computerized tomography and magnetic resonance imaging were performed in selected cases (large-sized tumors and in cases of exclusion of the other differential like malignancy).

All tumors were excised through an endoscopic approach with a few millimeters around the mass. Any complications were also recorded. The follow-up periods and recurrence were registered.

Data were entered and analyzed using Statistical Package for the Social Sciences (SPSS) version 25 for Windows. The mean and SD were calculated for the continuous variables and the frequency and percentage for categorical variables. The results were presented in tables or figures. The chi-square test was used to compare the categorical variables. A *P*-value of less than 0.05 was considered a statistically significant difference.

Results

With a male-to-female ratio of 1/2.73, 60 (73.2%) of the 82 patients with intranasal LCH were female (Table 1). The age ranged from 10–60 years with a mean age of 30.73 ± 11.366 years, while the median and mode were 27.5 and 25 years, respectively. The most affected age group was 18–40 years ($n = 65$, 79.3%) and the least < 18 years ($n = 5$, 6.1%). The mean duration from the onset of the disease was 2.32 ± 1.175 months (range 1–6 months).

Table 1 Demographic and clinical characteristics of 82 patients with LCH of the nasal cavity

Variable	Frequency	Percent
Age groups per years		
< 18	5	6.1
18–40	65	79.3
> 40	12	14.6
Gender		
Males	22	26.8
Females	60	73.2
Duration per months		
1–2	61	74.4
> 2	21	25.6
Clinical features		
Epistaxis	75	91.5
Nasal obstruction	44	53.7
Incidental finding	7	8.5
Side		
Right	33	40.2
Left	49	59.8
Site		
Septum	67	81.7
Lateral wall	15	18.3

The majority of the cases were presented within the duration group of 1–2 months ($n = 61$, 74.4%). Epistaxis was the most common presenting symptom ($n = 75$, 91.5%). The tumors originated from the left nasal cavity in 49 (59.8%) of the cases. Besides, the nasal septum was involved in 67 cases (81.7%) (Table 1).

The cause of the intranasal LCH was not detected in 35 patients (42.7%). While pregnancy and previous nasal trauma were found in 26 (31.7%) and 21 (25.6%), respectively. In each of the five pediatric cases, a historical trauma was mentioned as a potential tumor etiology. Eleven (91.7%) of the 12 patients in the over-40 age group did not have a known etiology. The age group 18–40 years showed that pregnancy was the possible cause in 26 (31.7%), while the least possible cause was nasal trauma ($n = 15$, 23.1%). The possible causes in males were nasal trauma in 11 cases (50%), while in the females, pregnancy was the most possible cause ($n = 26$, 31.7%). There were highly statistically significant differences between the possible causes and the age and gender (*P*-value = 0.000), while there was no significant difference between the possible cause and the duration of the disease, location, and side of the tumors (*P*-value > 0.05) (Table 2).

The surgical excision of the tumors was performed under general anesthesia in 29 patients (35.37%). The recurrence rate was 3.66% ($n = 3$), and all of them were subjected to local anesthesia. However, there was no

Table 2 The relationship between probable etiology and demographic and clinical factors in 82 individuals with intranasal LCH

Variable	Cause				P-value
	Pregnancy, N (%)	Nasal trauma, N (%)	Idiopathic, N (%)	Total, N (%)	
Age groups per years					0.000
< 18	0 (0)	5 (100)	0 (0)	5 (100)	
18–40	26 (40)	15 (23.1)	24 (36.9)	65 (100)	
> 40	0 (0)	1 (8.3)	11 (91.7)	12 (100)	
Total	26 (31.7)	21 (25.6)	35 (42.7)	82 (100)	
Gender					0.000
Males	0 (0)	11 (50)	11 (50)	22 (100)	
Females	26 (43.3)	10 (16.7)	24 (40)	60 (100)	
Total	26 (31.7)	21 (25.6)	35 (42.7)	82 (100)	
Duration per months					0.971
1–2	19 (31.1)	16 (26.2)	26 (42.6)	61 (100)	
> 2	7 (33.3)	5 (23.8)	9 (42.9)	21 (100)	
Total	26 (31.7)	21 (25.6)	35 (42.7)	82 (100)	
Side					0.755
Right	11 (33.3)	7 (21.2)	15 (45.5)	33 (100)	
Left	15 (30.6)	14 (28.6)	20 (40.8)	49 (100)	
Total	26 (31.7)	21 (25.6)	35 (42.7)	82 (100)	
Site					0.525
Septum	23 (34.3)	17 (25.4)	27 (40.3)	67 (100)	
Lateral wall	3 (20)	4 (26.7)	8 (53.3)	15 (100)	
Total	26 (31.7)	21 (25.6)	35 (42.7)	82 (100)	

statistically significant difference between the type of anesthesia used and the recurrence rate (P -value = 0.192) as shown in Fig. 1.

The follow-up period ranged from 6 to 15 months, with a mean of 9.94 ± 3.008 months. There were no complications in all the treated cases.

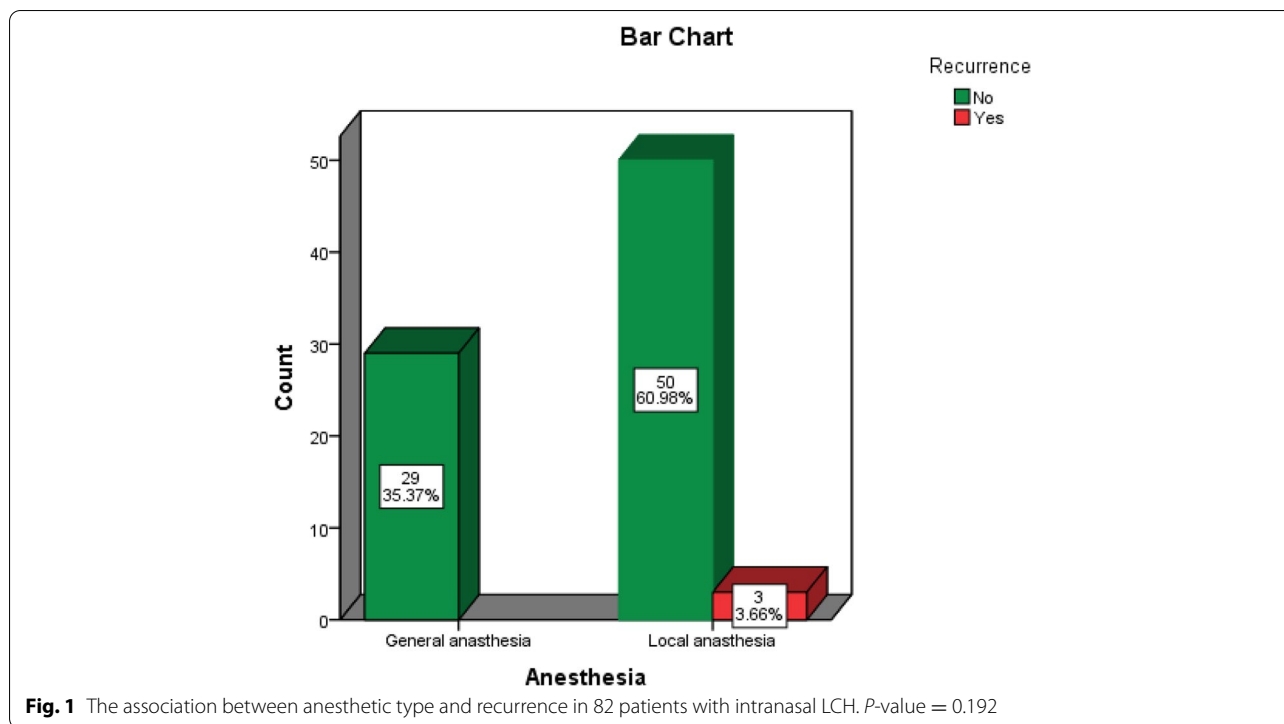
Discussion

LCH is a benign acquired vascular tumor that affects mainly the skin or the mucous membranes of the mouth and oropharynx. However, involvement of the nasal cavity occurs on rare occasions. The anterior part of the nasal septum is the commonest site involved in this lesion in comparison with other nasal parts. The exact pathogenesis of the LCH has not yet been recognized. However, there are four main mechanisms for the involvement of this rare tumor, including nasal trauma (surgical or accidental), hormonal changes such as pregnancy or contraceptive pills, viral oncogenes, and arteriovenous malformations [3, 6]. The LCH is still obscure to otolaryngologists regarding many aspects, particularly its occurrence, which remains underestimated. All the information in the literature comes from case reports or small case series [2, 4, 6, 9, 10]. To our best knowledge, the current study is the largest case

series in the world of LCH involving the nasal cavity. Besides, the study examined the relationship between the possible causes of this rare tumor with certain factors and showed a significant correlation between the cause and gender and age.

This study was consistent with previous studies in that the LCH is more prevalent in females than males [3, 11]. However, a study by Puxeddu et al. revealed a slight male predominance (21 males out of 40 cases) [2]. The female predominance in our study may be related to a considerable number of pregnant cases with LCH ($n = 26$, 31.7%). Moreover, the current study reported a highly statistically significant difference between the possible cause and gender (P -value = 0.000).

LCH could affect all age groups [2]. However, the most vulnerable age group is between the third and fifth decades [12, 13], while the disease is extremely rare in the pediatric age group [6]. Around 80% of our cases were in the age group of 18–40 years. Besides, our results reported that nasal trauma was a possible cause in children (5/5), pregnancy in the age group of 18–40 years (26/60), and no identified cause in the age group > 40 years (11/12) (Table 2). Therefore, the possible cause of the LCH might be determined by the age of the patient (P -value = 0.000).



Owing to the vascular nature of the tumor, epistaxis is the chief complaint in the majority of cases. However, nasal obstruction, rhinorrhea, facial pain, headache, and hyposmia might be the chief complaints either alone or associated with other features in different combinations [2]. Moreover, on rare occasions, intranasal LCH could be found as an incidental finding on a routine nasal examination [3]. The current study revealed that epistaxis was the presenting symptom in 75 cases, and 44 of them were associated with nasal obstruction. In general, the bleeding is usually slight and recurrent with a relatively long duration (month/s). But severe epistaxis which does not stop spontaneously due to LCH of the nasal cavity is also reported in the literature [9]. Therefore, it is of utmost importance to manage the patient promptly, as well as the exclusion of other differentials (bleeding nasal polyp, sinonasal tumor, angiofibroma, and vascular malformation) is essential too.

When we reviewed the literature, intranasal LCH occurs in the vast majority of cases on one side. However, the study by Lopez et al. reported 2 of their 38 cases with bilateral involvement, one of them with bilateral lesions and the other one with a midline tumor arising from the posterior rim of a nasal septal perforation [3]. The current study reported that the left side was involved in 60%, which was consistent with previous studies [4, 11]. Besides, the present study was consistent with other investigations regarding the nasal septum as

the commonest site of LCH [2–4], while the study by Chi et al. found only 7 out of 15 cases were originating from the nasal septum [11]. Furthermore, the study did not show a significant difference between the possible cause and the side and location of the LCH (P -value > 0.05).

The following case reports showed that the possible causes of LCH in the nasal cavity were due to anterior nasal packs [14], endoscopic transsphenoidal gonadotrophin-producing pituitary adenoma resection [15], and pregnancy [16, 17]. It is well-known that trauma and pregnancy are possible causes of LCH in the nasal cavity. Hormonal changes in pregnancy might be implicated as a possible cause of the increased incidence of intra-nasal LCH [3]. Another possible reason for such an increment is hyper-dynamic status during pregnancy. However, only a few cases were due to those two causes (pregnancy and nasal trauma) in many previous case series studies [2, 3, 7, 11]. Although our study revealed that there were considerable cases due to either pregnancy or nasal trauma, there were 42.7% of unidentified causes. We think that the higher percentage of an idiopathic cause may be related to either the patients' having forgotten trivial trauma or other general diseases or other still unknown causes. Therefore, a thorough history and proper physical examination, including systemic evaluation, are of utmost importance to search for possible causes of this rare entity of the nasal cavity. Knowing the possible causes of this rare tumor helps us more in understanding

the mechanisms of tumor initiation and better management of the patients.

It was reported in the literature that the incidence of intranasal LCH in pregnant women is ranged from 2 to 5% [3]. In our series, there was a much higher number of this tumor during pregnancy ($n = 26$, 31.7%) than in other case series studies [2–4, 7, 11]. This may be attributed to the fact that the pregnancy rate in Iraqi women is much higher than that in women from the countries of the abovementioned studies [18].

Surgical removal is an appropriate method of LCH treatment. There are different ways to achieve its excision, including removal by electrocautery, cryotherapy or laser, and excisional procedures either alone or after embolization [2, 11]. The operation is performed either under local or general anesthesia, depending on the size and site of the lesion [2]. In all cases of the current study, the LCH was excised using a zero-degree endoscope as well as the removal of a few millimeters of cuff of mucosa surrounding the lesion. The recurrence rate in the literature varies greatly, ranging from 0 to 42% [2, 4, 11]. This variation depends on the sample size of the case series and the period of follow-up. Our study reported that the recurrence rate was 3.66% (3/82), and all three cases were excised under local anesthesia. However, there was no statistically significant difference between the recurrence rate and the type of anesthesia used (P -value = 0.192).

Many limitations accompanied this study. The study did not take into consideration the exact site and the size of the LCH. The relatively short period of follow-up (mean 9.94 ± 3.008) had an impact on the accurate assessment of the recurrence rate. Lastly, the retrospective nature of the study was considered another limitation.

Conclusion

This study is considered the largest case series in the world. The study revealed that the intranasal LCH had a female predominance with the majority of the cases in the age group of 18–40 years. Epistaxis was the most common presenting feature of the LCH. Around 60% of the LCHs were on the left side. The majority (81.7%) of the cases originated from the nasal septum. About three-quarters of the patients were diagnosed within 1–2 months from the onset of the disease. The study revealed that there was no identified cause in 35 subjects, while trauma and pregnancy were the possible causes in 25.6% and 31.7%, respectively. Age and gender might play a role in determining the possible cause of the LCH. 3.66% (3/82) of the incidents occurred again. In every event that occurred repeatedly, local anesthetic was utilized.

However, the kind of anesthetic and the recurrence rate did not differ in a statistically meaningful way.

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Authors' contributions

RMA and OMB collected the data. OMB wrote the introduction and methodology sections and searched the related published studies. RMA revised the introduction and made the analysis, and he was a major contributor in writing the manuscript. Both authors read and approved the final manuscript.

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Availability of data and materials

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Written approval had been gained from the ethical committee at the University Of Anbar, Iraq (reference 52 number 18 on 18-4-2022). Study data/information was used for the research purpose only. Owing to the retrospective nature of the study, informed consents from the patients were waived.

Consent for publication

Not applicable (no individual personal data included).

Competing interests

The authors declare that they have no competing interests.

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