

# Branchial Remnants: A Retrospective Study of 50 Patients and Review of the Literature

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## Abstract

**Objectives:** To study the clinical spectrum of branchial remnants and to analyze the methods of diagnosis and management of these lesions.

**Methods:** Retrospective review of 50 patients presenting with 60 branchial remnants reported at Jordan University Hospital from 1986 – 2001.

**Results:** Fifty patients (23 males, 27 females) presented with 60 branchial remnants were identified. Of these, 23 on the right side, 22 on the left side, 5 bilaterally and 1 patient with 4 anomalies. Thirty one patients presented with branchial cysts, 12 cervical sinus, 10 fistulas and 3 patients with dual branchial remnants; cyst and sinus and one patient with branchial cartilage. Sixty eight percent of branchial cysts occurred below the age of 20 years whereas 76% percent of fistulas and sinuses occurred below the age of 10 years. The junction of the upper and middle third of sternocleidomastoid muscle was the most common site for branchial cyst (96%) and in one case the cyst was found inside the parotid gland. Two patients with branchial sinus had recurrence ranging between 1- 3 times and another 2 patients with fistulas had 3 times recurrence.

**Conclusions:** Branchial remnants have a variety of clinical presentations; cyst, fistula and sinus. Accurate diagnosis is crucial in preventing recurrences and redo surgeries. Good history and physical examination is enough in the diagnosis of most of these lesions. Preoperative radiological diagnosis preferably a computed tomography (CT scan) is helpful in determining the extent, nature and possible complications of these anomalies. Complete and careful surgical excision is recommended in all types of branchial remnants.

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## Keywords

Branchial anomalies, branchial remnants, cyst, fistula.

## Introduction

Branchial Remnants (BR) are congenital anomalies of the branchial arches. Six pairs of mesodermal condensation appear during intrauterine life surrounding the primitive pharynx. In between these arches, there are spaces lined internally by endoderm and are called branchial pouches. Externally these spaces are lined by ectoderm and are called branchial clefts (Figure-1).<sup>1</sup> Branchial remnants may present in the form of a cyst, sinus or fistula. If a portion of the cleft fails to involute completely, the entrapped remnant forms an epithelium-lined cyst which is called branchial cyst. A branchial sinus is a blindly ending pouch with a single opening. Branchial fistula is applied to the remnant when both external and internal openings are present. Depending on their anatomical location, branchial remnants are classified into first, second, third and fourth anomalies. Most branchial remnants are considered to be derived from the second and first branchial cleft. Initial surgical exploration is crucial because recurrence rate after incomplete surgical excision can be as high as 22%.<sup>2,3</sup> The aim of this paper is to describe our experience with 50 patients diagnosed to have BR and to analyze our methods of diagnosis and management of these lesions.

## Methods

The medical records of all patients treated at Jordan University Hospital (JUH) with the primary diagnosis of BR during the period 1986-2002 were reviewed. A total of 50 cases were found. Patients' data such as age and sex were recorded along with the clinical diagnosis. All patients underwent full clinical examination. Fistulography and Computed Tomography (CT) were performed as indicated (Figure-2).

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All cases were classified according to the site of the lesion, type of the lesion (cyst, sinus, or fistula). Diagnosis, surgical therapy, complications and the pathologic findings were reviewed.

**Results**

Fifty patients presented with 60 branchial remnants were identified between 1986 and 2001. Of these, 23 on the right side, 22 on the left side and 5 bilaterally. The clinicopathological findings of all cases are summarized in Table (1). The youngest patient was a six month old female and the oldest one was 57 years old male. The most presenting symptoms were cervical swelling, cervical sinus opening or repeated infection. The size of the cysts was variable ranging from 3 to 8 cm in their largest diameter. All patients with branchial cyst were cured by cyst excision from the first time. Two patients with branchial sinus and two patients with branchial fistula had 2-3 times recurrence after the first operation. Eight patients developed minor complications following excision consisting of local wound infection <sup>6</sup> and seroma/hematoma formation. <sup>2</sup> None of the patients in this series developed major nerve injuries, 3 patients however, developed cutaneous parasthesia which resolved over several weeks. Hypertrophic scars were seen in 3 patients.

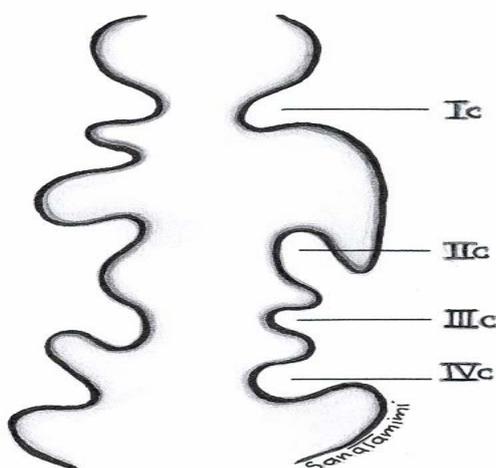


Figure 1: Schematic representation of the branchial apparatus during the weeks 4-6 of intrauterine life showing 6 pairs of arches and the branchial pouches (subscript P) and clefts (subscript C).



Figure 2: First branchial cleft cyst, t Contrast-enhanced axial CT at the level of the hyoid bone reveals an ill-defined, nonenhancing mass (M) posterior to the right submandibular gland (G).

Table (1)

<u>Demographics</u>	
<u>Age</u>	range 1-57year
<10 years	19 patients
11-20 years	19 patients
21-30 years	8 patients
> 31 years	4 patient
<u>Sex</u>	
Male	23
Female	27
<u>Presentation</u>	
Branchial cyst	31
Branchial fistula	10
Branchial sinus	12
Bilateral fistulae	3
Cartilage remnant	1

## Discussion

Remnants of the second branchial arch are the most frequent branchial anomalies with figures as high as 95% being reported.<sup>4, 5</sup> First branchial remnants account for 1-8% with third and fourth branchial remnants being rarely reported.<sup>6, 7</sup> Their incidence however, is rising and several cases described in the recent literature.<sup>8- 11</sup> In this study, branchial cysts of the second branchial arch were prevalent among woman (68%) in their second decade (52%). The junction of the upper and middle third of sternocleidomastoid muscle was the most common site for branchial cyst (96%) and in one case the cyst was found inside the parotid gland. Twelve patients in this series presented with branchial sinuses and 10 patients with branchial fistula, 19 patients with BR (38%) were below the age of 10 years.

In a clinical review of 52 branchial anomalies in pediatric patients by Choi and Zalzal from US, second BR was seen in 40%, first in 25%, third in 8% and fourth in 2%. In this series, sinuses and fistulas were seen at an earlier age than cysts.<sup>3</sup> These results supported the previous findings, which were carried out by Doi et al from Australia who described 58 cases of branchial remnants, 67% of whom were suspected to be from the second branchial cleft, 43/66 branchial fistulas 87% of them were <5 years of age at the time of operation.<sup>12</sup> The incidence of first BR in our report is 12% (7cases) similar to what is reported by Doi et al.<sup>12</sup> The presence of thyroid tissue in the wall of the cyst or an internal opening communicating with the pharynx at the level of thyrohyoid membrane suggests a third or a fourth BR. None of our patients was suggestive of 3<sup>rd</sup> or 4<sup>th</sup> branchial remnants. These third or fourth BR will be distinguished by the precise relationship of the tract and the carotid artery, nerves, thymus and thyroid. The association of these remnants with thyroid or thymus tissue will suggest the origin of these remnants.<sup>13</sup>

A history of intermittent swelling and tenderness of the lesion during upper respiratory tract infection is the usual presentation of branchial remnants. Discharge may be reported if the lesion is associated with a sinus tract.<sup>14- 16</sup>

Most patients in this study presented with the classic signs and symptoms of branchial remnants, i.e., soft or mobile mass or draining tract located around the middle third of SCM (30) branchial cyst, (6) fistula and (3) branchial sinus.

History and physical examination are the most important elements in the diagnosis of branchial remnants. Radiographic studies can be helpful in the evaluation of suspicious cases to confirm the diagnosis or to suggest alternative diagnosis.<sup>16- 17</sup> Fistulography and CT scan were used in this study to confirm the diagnosis in some patients with typical sites. A CT scan is a noninvasive and an accurate tool to confirm the diagnosis of BR, define the location and the extent of the lesion and suggests the presence of infection.<sup>17, 18</sup> Sinogram or barium contrast studies are helpful in delineating the course of branchial sinuses or fistula especially in third and fourth BR.<sup>5, 18- 20</sup> Accurate initial diagnosis is crucial because recurrence is uncommon, with a risk estimated at 3%, unless previous surgery or recurrent infection has occurred, in which case, it may be as high as 20%.<sup>2, 3</sup>

Complete surgical excision is recommended for all types of branchial remnants because untreated lesions are prone to recurrent infection and abscess formation with resultant scar formation and possible compromise to local structures.<sup>3, 4, 21</sup> The lesion should be excised completely without injury to normal structures. First branchial remnants are removed through an incision similar to that used for parotidectomy (Figure-3). It is essential to delineate the branches of the facial nerve before any further surgery is done. The parotid gland must be reflected forward. Dissection of the lesion continues up to its attachment at the external auditory canal, where it is ligated and divided. A transverse cervical incision near the anterior border of the sternocleidomastoid muscle is recommended for the second branchial cleft cyst (Figure -4).<sup>16, 22, 23</sup> The cyst must be removed completely, and any adjoined fistula with it. Great care must be taken to avoid injury to nearby nerves, particularly, the vagus, the spinal accessory nerve and the hypoglossal nerve.

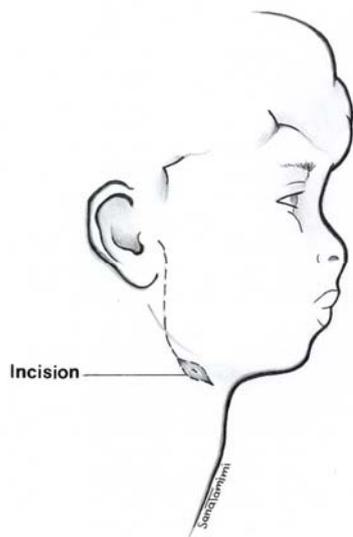


Figure 3: Incision used for removal of first branchial remnants derivatives.

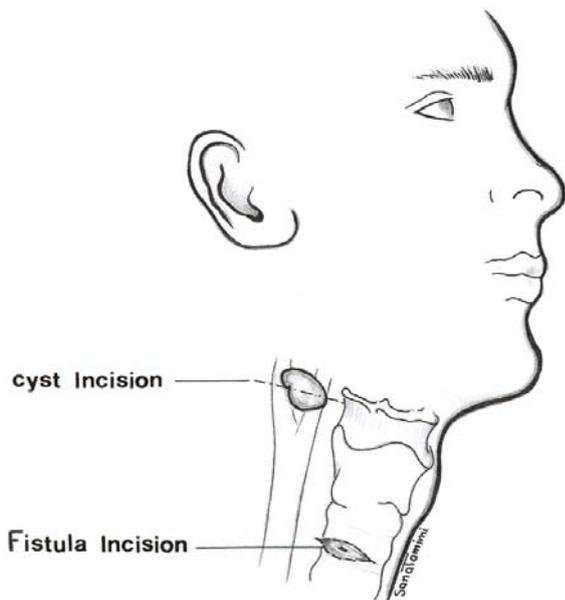


Figure 4: Skin incision used for the removal of 2<sup>nd</sup> branchial cyst or fistula in adults.

Antibiotics, incision and drainage are required to control acute inflammation before definitive surgery is undertaken. Eleven patients in this study developed minor wound complications which has resolved over the following weeks and 3 patients developed hypertrophic scars. Postoperative complications, however tend to increase in older patients, patients presenting with infections and first branchial cleft anomalies.<sup>12, 24</sup>

In conclusion, branchial remnants should be considered in the differential diagnosis of lateral neck masses, especially in children and young adults. The presence of a mass or draining sinus, with or without infection usually suggests the diagnosis. Lesions with a typical location or infectious presentation may confound the diagnosis and radiological evaluation with a CT scan or fistulography is recommended in these cases. The objectives of therapy should be a complete surgical excision from the first attempt.

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## البقايا الخيشومية: دراسة راجعة لخمسین مريض مع مراجعة للأدبيات الطبية

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### ملخص:

### الأهداف:

دراسة الأظياف السريرية للبقايا الخيشومية وتحليل الطرق التشخيصية والعلاجية لها.

### الطريقة:

دراسة راجعة لخمسین مريض فيهم ستين حالة من البقايا الخيشومية عولجت في مستشفى الجامعة الأردنية في الفترة الواقعة بين 1986 - 2001.

### النتائج:

50 مريض ( 23 ذكر، 27 أنثى ) فيهم 60 حالة بقايا خيشومية. منها 23 حالة كانت موجودة في الجهة اليمنى، 22 حالة في الجهة اليسرى، 5 حالات في الجهتين معاً، و 4 شذوذات خيشومية موجودة في مريض واحد. البقايا الخيشومية كانت على النحو التالي: 31 كيسة خيشومية، 12 جيب عنقي، 10 نواسير خيشومية، 3 مرضى يعانون من ناسور و كيس خيشومي معاً، و مريض واحد يعاني من غضروف خيشومي.

68% من الأكياس الخيشومية حدثت في الأعمار الأقل من 20 سنة، في حين كانت هناك 76% من الأكياس والجيوب الخيشومية في الأعمار الأقل من 10 سنوات.

المكان الأكثر شيوعاً للأكياس الخيشومية ( 96% ) هو مكان التقاء ( أو إتصال ) الثلث العلوي بالثلث الأوسط من العضلة القصية الترقوية الخشائية. كيسة خيشومية واحدة كانت موجودة في الغدة النكفية. حالتان من الجيوب الخيشومية وحالتان من النواسير الخيشومية رجعت لحالاتهم المرضية 2-3 مرات بعد العملية الأولى.

الخاتمة: تظهر البقايا الخيشومية سريرياً على شكل كيسة، جيب، أو ناسور. التشخيص الصائب قبل العملية هام جداً لمنع رجوع البقايا الخيشومية بعد استئصالها جراحياً. السيرة المرضية و الفحص السريري الجيدان كافيان لتشخيص معظم حالات البقايا الخيشومية. يساعد التصوير الشعاعي قبل العملية والذي يفضل أن يكون تصويراً طبقياً في تحديد موقع ونوع هذه الشذوذات والعواقب المحتملة منها. ننصح بالاستئصال الكامل جراحياً لجميع أنواع البقايا الخيشومية.

### مفتاح الكلمات:

الشذوذات الخيشومية، البقايا الخيشومية، كيسة، ناسور.