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Second Branchial Arch Anomalies: A Single Center Experience

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ABSTRACT

Background: Second branchial arch anomalies represent the most common type of branchial arch anomalies, accounting for approximately 95% of all cases.

Patients and methods: This retrospective study evaluates the clinical presentation, management strategies, and outcomes of 86 patients with second branchial arch anomalies treated at Sohag University Hospital between January 2011 and March 2023.

Results: The majority of patients presented with a discharging neck fistula or sinus (70.97%), while non-discharging lesions were observed in 25.81%, and cystic neck masses accounted for 3.23%. Preoperative imaging included neck ultrasound (86.02%), CT scans (30.11%), and fistulography (10.75%). Fine-needle aspiration cytology was used in three cystic cases. Surgical excision was performed using a single incision in 91.4% of lesions, while 8.6% required two incisions. Methylene blue dye was utilized intraoperatively in 58.06% of cases to delineate the tract, with visualization of pharyngeal openings in 9.68% of lesions, confirming complete fistulas. Postoperative complications included wound hematoma/seroma (2.15%) and infection (1.07%). No recurrences were recorded during a mean follow-up period of 5.3 months.

Conclusions: This study highlights the importance of accurate diagnosis, careful surgical planning, and meticulous dissection to ensure successful outcomes with minimal morbidity

Key Words: *Branchial anomalies; Cervical cysts; Cervical sinuses; Cervical fistulae Infants; Children; Pediatric*

1. INTRODUCTION

Congenital cervical anomalies represent a significant proportion of head and neck pathologies encountered in pediatric surgical practice, with branchial arch anomalies accounting for a large percentage [(1)]. Among these, second branchial arch anomalies are the most common, representing up to 95% of all branchial anomalies[(2-3)]. These anomalies arise from incomplete involution of the branchial apparatus during embryonic development and may present clinically as cysts, sinuses, or fistulas along the lateral neck region[(2)].

Typically, second branchial cleft anomalies manifest as discharging or non-discharging sinus tracts anterior to the sternocleidomastoid muscle or as asymptomatic or infected cystic masses[(4)]. Due to their variable clinical presentation, diagnosis often requires a high index of suspicion. While clinical examination is usually sufficient for diagnosis, imaging modalities such as ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI) can be valuable tools in delineating the anatomical course of the lesion, particularly when the diagnosis is uncertain or preoperative planning is complex[(5)].

The definitive treatment for these lesions is complete surgical excision, which is essential to prevent recurrent infections and minimize the risk of recurrence[(6)]. Various surgical techniques have been described, including single or multiple incisions, with some surgeons utilizing methylene blue dye intraoperatively to trace the tract[(7)]. Despite advancements in diagnostic and surgical techniques, challenges remain in achieving complete resection without injury to adjacent neurovascular structures[(8)].

PATIENTS AND METHODS

The present study included a retrospective chart review of 86 patients with second branchial arch anomalies who were

managed at our center from January 2011 to March 2023. Institutional review board approval was obtained for this retrospective study based on hospital records only. Medical records of patients with a diagnosis of second branchial arch anomalies were examined and only records with complete data were included in the review.

Data gathered included age and sex, presenting symptoms, history of infection, associated anomalies, family history, the side of the lesion, preoperative investigations, any medical treatments as well as operative findings (including the number of surgical incisions, the intraoperative use of methylene blue dye to identify the tract and the extent of the sinus or fistula tract), follow up period, postoperative morbidity and outcome

2. RESULTS

Table (1) shows the summary of the results. This study retrospectively examined 86 patients with second branchial arch anomalies. Among them, 52/86 (60.46%) were male. The mean age at presentation was 2.6±0.71 years (range: 0-13); while the mean age at operation was 3.9±0.32 years (range: 0.7 - 15). Family history was positive in 2/86 patients (2.33%) who were siblings, and both had bilateral fistula. Associated anomalies were found in 2/86 patients (2.33%).

The lesion was right sided in 56/86 patients (65.12%), left sided in 23/86 (26.74%) and bilateral in 7/86 patients (8.14%) giving a total of 93 lesions.

A discharging neck fistula/sinus was the most common presentation (66/93 lesions; 70.97%). Among them, 8/93 lesions (8.60%) had purulent discharge indicating infection that resolved by a 5-days course of antibiotic therapy. The fistula/sinus was non-discharging in 24/93 lesions (25.81%). The presentation was that of a cystic neck mass in 3/93 lesions (3.23%). Neck abscess developed in 2/93 lesions (2.15%), both were treated with incision and drainage followed by a 5-days course of antibiotic therapy.

Preoperative investigations included neck ultrasound in 80/93 lesions (86.02%), CT scan in 28/93 lesions (30.11%), fistulogram in 10/93 lesions (10.75%) and FNA of the cyst in 3/93 lesions (03.23%).

A single neck incision was required to complete the excision of 85/93 lesions (91.40%), while 2 incisions were required to excise 8/93 lesions (8.60%). The track extended to the carotid vessels with no opening in the pharynx (sinus) in 66/93 lesions (70.97%). Methylene blue dye was used intraoperatively in 54/93 lesions (58.06%), and the dye was visible in the pharynx in 9/93 lesions (9.68%) consistent with a branchial fistula.

The mean follow-up period is 5.3±1.1 months (0–9). Postoperative wound hematoma/seroma occurred in 2/93 lesions (02.15%), while postoperative wound infection occurred in 1/93 lesion (1.07%). There were no recurrences during the follow-up period.

Table (1): Summary of the results

Patients' demographics	
Total number of patients	**86 patients**
Males; n (%)	52/86 (60.46%)
Age at presentation in years; Mean ± SD (Range)	2.6±0.71 (0-13)
Age at operation in years; Mean ± SD (Range)	3.9±0.32 (0.7-15)
Associated anomalies; n (%)	2/86 (2.33%)
Positive family history; n (%)	2/86 (2.33%)
Side	
Right; n (%)	56/86 (65.12%)
Left; n (%)	23/86 (26.74%)
Bilateral; n (%)	7/86 (8.14%) = 14 lesions
Total number of lesions	**93 lesions**
Presentation	

Discharging lateral neck sinus/fistula; n (%)	66/93 (70.97%)
Non-discharging lateral neck sinus/fistula; n (%)	24/93 (25.81%)
Cystic lateral neck mass; n (%)	3/93 (3.23%)
Previous Infection	
Purulent discharge; n (%)	8/93 (8.60%)
Abscess; n (%)	2/93 (2.15%)
Previous Treatment	
Antibiotics	10/93 (10.75%)
Incision and drainage	2/93 (2.15%)
Preoperative investigations:	
Neck ultrasound; n (%)	80/93 (86.02%)
Neck computed tomography; n (%)	28/93 (30.11%)
Fistulogram; n (%)	10/93 (10.75%)
FNA; n (%)	3/93 (3.23%)
Operative details	
Single incision; n (%)	85/93 (91.40%)
Two incisions; n (%)	8/93 (8.60%)
Track extended to carotid vessels; n (%)	66/93 (70.97%)
Methylene blue used; n (%)	54/93 (58.06%)
Fistulous opening in the pharynx; n (%)	9/93 (9.68%)
Postoperative	
Follow up in months; Mean ± SD (Range)	5.3±1.1 (0 – 9)
Postoperative seroma/hematoma; n (%)	2/93 (2.15%)
Postoperative wound Infection; n (%)	1/93 (1.07%)
Recurrence; n (%)	0/93 (0%)

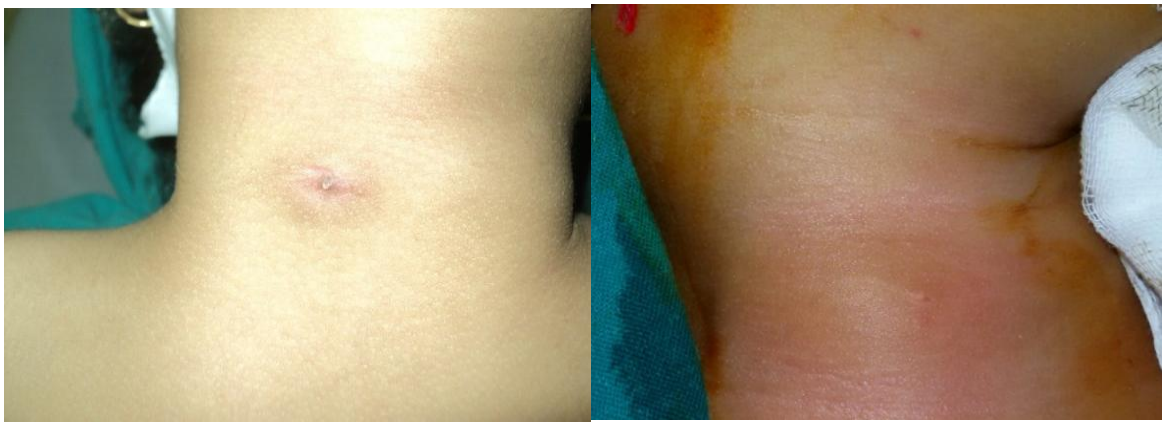

Figure (1): A discharging or non-discharging second branchial sinus/fistula.

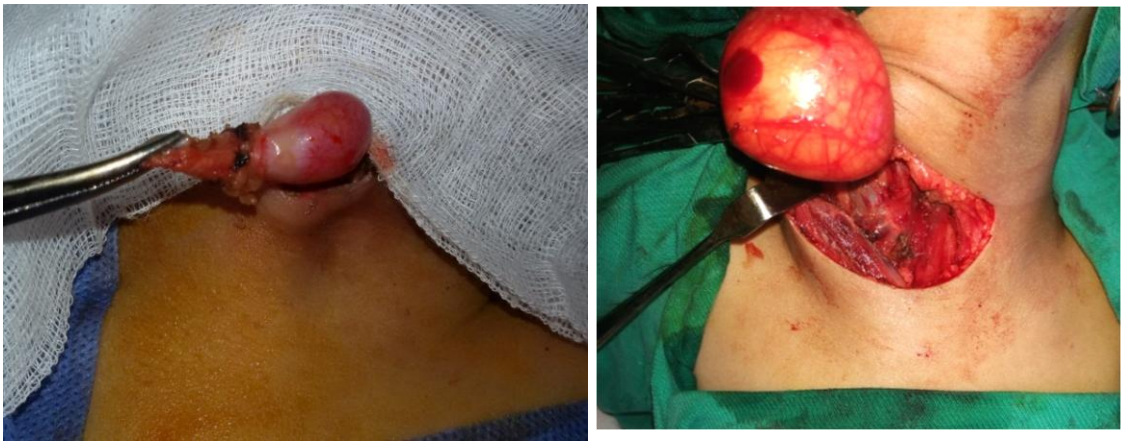

Figure (2): Operative images of second branchial cleft cysts

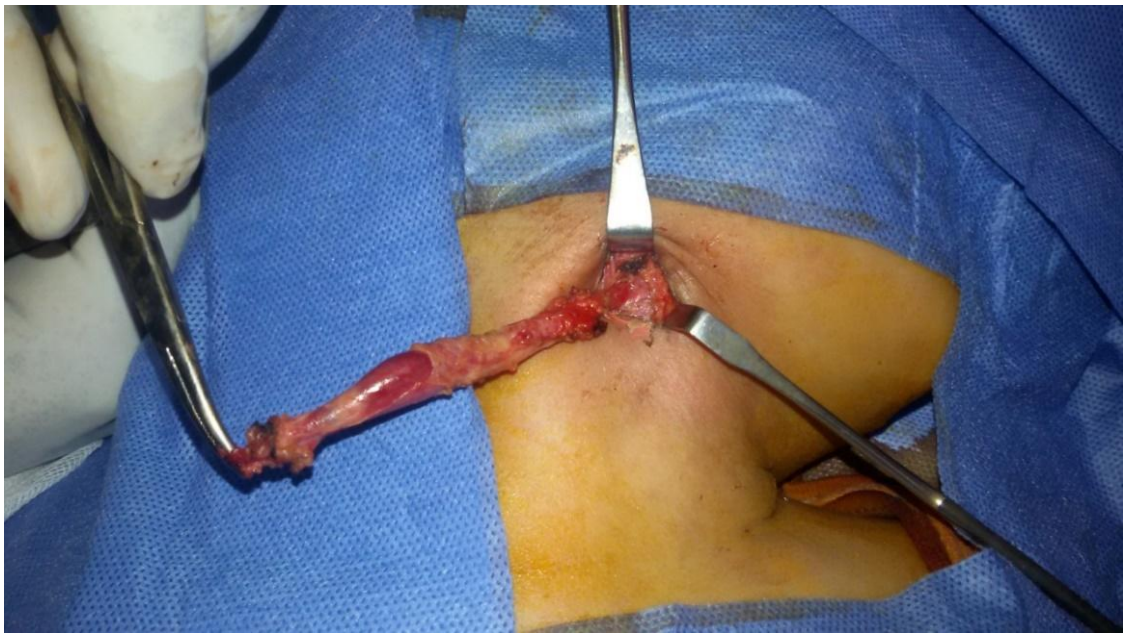

Figure (3): Operative image of a second branchial cleft fistula

3. DISCUSSION

Congenital neck malformations account for more than half of cervical masses treated on children. Of them, around 20% are branchial abnormalities, with second cleft anomalies being the most common[1].

During the fourth week of embryonic life, six pairs of branchial arches (the branchial apparatus) develop on each side of the foregut; each arch is made up of a cleft and a pouch. These arches serve as the embryonic foundation for the development of the face, neck, and pharynx. Incomplete involution of the branchial apparatus can result in branchial arch anomalies. Among these, anomalies of the second branchial arch account for up to 95% of all cases[2-3].

While most branchial anomalies are identified in infancy or childhood, certain lesions may remain unnoticed until later in life. **Karabulut et al.** [9] reported a mean age at presentation of 5.30 years, **Kalra et al.** [4] reported a mean age at presentation of 5.56 years. While, **Kajosaari et al.** [6] reported a mean age at presentation of 3.2 years. In our study, the mean age at presentation was 2.6±0.71 years (range: 0 to 13). Probably this difference in the age at presentation is related to differences in the nature of referral systems and health accessibility among different centers. The mean age at operation in our study was 3.9±0.32 years (range: 0.7 -15), which is very close to an age of 4.1 years reported by **Kajosaari et al.** [6].

Different reports gave varying male to female ratios in patients with second branchial arch anomalies. Sixty percent of patients in our study were males giving a male to female ratio of 1:0.67. **Simpson** [10] reported a male predominance with a ratio of 1:0.33. However, **Reddy et al.** [11] reported a ratio of 1:1.39.

Branchial anomalies are more common on the right side of the neck. In our study unilateral right-sided lesions were found in 65.12% and bilateral lesions in 8.14% of the patients. This came in accordance with **Patigaroo et al.** [12] who reported right-sided lesions in 78% and bilateral lesions in 10% of their patients and **Reddy et al.** [11] who reported 81% right sided and 15% bilateral lesions.

A branchial cyst often presents as a lateral cervical cystic mass, while branchial fistulae and sinuses commonly present with a discharging or non-discharging openings related to the anterior border of the sternocleidomastoid muscle [4]. In our study a discharging neck fistula/sinus was the most common presentation, occurring in 66/93 lesions (70.97%) (Figure 1). The fistula/sinus was non-discharging in 24/93 lesions (25.81%). Only 3 out of 93 lesions (3.23%) in our study presented as cysts. Similarly, **Kalra et al.** [4] reported branchial fistulae/sinus in the majority of their patients (90.7%) and branchial cysts in only 9.3%. However, **Lee et al.** [5] reported their experience with 25 patients with second branchial cleft anomalies; 23 of them (92.0%) presented as cysts and only 2 (8.00%) presented as fistulas. Sinuses and fistulas are usually detected earlier while cysts may remain asymptomatic until they grow large enough to cause noticeable symptoms [2, 13].

In the present study preoperative infection occurred in 10/93 lesions (10.75%) and manifested by purulent discharge from a lateral neck opening in 8/93 lesions (8.60%), and neck abscess in 2/93 lesions (2.15%). Preoperative infection rates of 24% and 23.88% were reported by **Kajosaari et al.** [6] and **Reddy et al.** [11] respectively. On the other hand, **Maddalozzo et al.** [14] reported a preoperative infection rate of 7.1%.

Clinical identification of a second branchial cleft sinus or fistula is typically possible. The degree of diagnostic certainty must be taken into consideration when deciding whether to conduct radiological investigations. The procedure can be successfully carried out without any prior imaging if the diagnosis of second branchial cleft sinus/fistula is certain. Nonetheless, preoperative localization of the lesion and determination of its relationship to surrounding tissues are made possible by ultrasonography, CT, and MRI, which can also be used to identify abnormalities in the branchial cleft [5]. The most commonly requested investigation in our series was a neck ultrasound (86.02%), while a neck CT was obtained in 30.11%. **Kajosaari et al.** [6] reported zero use of preoperative imaging in their patients, while **Maddalozzo et al.** [14] reported 96.4% use of preoperative imaging; the majority of them (82%) was in the form of neck CT. Similarly, **Lee et al.** [5] reported the use of CT in 100% of their patients. Fine-needle aspiration cytology (FNAC) had a diagnostic sensitivity of 100%, a positive-predictive value of 100%, and accuracy of 100% for diagnosing second branchial cleft cyst [5]. In our study, FNAC was obtained from all three cystic lesions.

A preoperative fistulogram was obtained in 10.75% of the lesions in the present study, while intraoperative injection of methylene blue dye was performed in 58.06%. **Rattan et al.** [7] investigated the use of fistulograms and methylene blue dye injections in delineation and full surgical excision of the tract and discovered that they provided no additional benefit.

The treatment of branchial fistula is complete surgical excision of the fistulous tract. Incisions for this excision could be a single or multiple (stepladder) incision [12]. Most of the lesions in our study were successfully excised via a single neck incision (91.40%), and only 8.6% of lesions required 2 incisions for complete excision. (Figures 2 and 3)

Bailey [15] categorized second branchial cleft fistulas/sinuses into four types. Type I is characterized by a blind-ending fistula that extends as far as the fascia of the sternocleidomastoid muscle. Type II reaches the surface of the carotid artery. Type III traverses the carotid bifurcation and extends to the lateral pharyngeal wall. Type IV, referred to as a complete branchial fistula, connects externally and internally, with an opening in the tonsillar fossa. As concluded in the review of literature by

Ang et al. [8] complete branchial fistulae (type IV) are rarely seen in clinical practice. This is consistent with our study data showing 9.68% of all lesions were complete fistulae. **Maddalozzo et al.** [14] reported a similar rate of 13.5% for complete fistulae in their series. On the other hand, sinus tracts extending to the carotid vessels (Type II & III) were very common representing 70.97% of the lesions in the present study.

Complications associated with the surgical treatment of branchial anomalies may include wound hematoma/seroma, secondary infections and injury to adjacent anatomical structures, such as the facial nerve, recurrent laryngeal nerve, hypoglossal nerve, spinal accessory nerve, and glossopharyngeal nerve. Careful dissection along the tract, as performed in our study, can help prevent such nerve damage [7]. In our study there was no cases of nerve injury, wound hematoma occurred in 2.15% of lesions while wound infection occurred in 1.07%. In the study of **Kalra et al.** [4] wound infection was reported in 4.25% of cases. Reported recurrence rates after surgery for second branchial cleft fistulae vary from 3 to 22% [11]. In our series, a recurrence rate of 0% was observed. This is similar to rates reported by **Rattan et al.** [7] and **Kalra et al.** [4].

4. CONCLUSIONS

Second branchial arch anomalies can present as a discharging or non-discharging neck fistula, although less commonly they may present as a cystic neck mass. The most important complication is infection, and complete surgical excision is the most effective treatment. The use of preoperative fistulogram or intraoperative methylene blue dye to delineate the lesion doesn't seem to add a significant value.

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