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# The utilization of selective neck dissection in the treatment of recurrent branchial cleft anomalies

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**Abstract**

To investigate the characteristics of recurrent branchial cleft anomalies (BCAs) and to evaluate the surgical technique and outcomes of patients undergoing reoperation.

From January 2005 to August 2018, the clinical data of 216 patients with recurrent second, third, and fourth BCAs were retrospectively analyzed. According to the embryological and anatomical features of the cleft palate and recurrence site, selective neck dissection techniques were used for surgical treatment.

Among all 216 patients, 203 healed by primary healing. Twelve patients with local infections and 1 patient with a pharyngeal fistula healed after dressing changes. Eleven patients experienced transient hoarseness and recovered after a few months. Three patients developed permanent hoarseness, and 5 patients developed coughing after eating and drinking. Three patients underwent internal jugular vein ligation. Only 4 recurrences occurred during a follow-up period of more than 1 year. The total cure rate was 98.15%.

Selective neck dissection is an effective and safe surgical treatment for recurrent second, third, and fourth branchial cleft anomalies.

**Abbreviations:** BCA = branchial cleft anomalies, SND = selective neck dissection.

**Keywords:** branchial cleft anomalies, complication, recurrence, selective neck dissection, surgical treatment

## 1. Introduction

Branchial cleft anomalies (BCAs) are the second most common congenital head and neck lesions in children, accounting for approximately 20% to 30% of pediatric congenital masses after thyroglossal duct cysts and sinuses.[1,2] The persistent anomalies have been described as incomplete obliteration of the branchial apparatus during embryogenesis, which can be categorized into four types.[3,4] Each type of BCA has a different incidence rate. Second BCAs are the most common, accounting for 90% to 95% of all BCAs. Third and fourth BCAs are very uncommon, with prevalence rates of 2% to 8% and 1% to 4%, respectively.[5,6] Commonly, the internal opening of second, third fourth BCAs is located in the low pole of palatine tonsillar fossa, the base of pyriform sinus and the apex of pyriform sinus, respectively. Besides, these developmental anomalies have been categorized anatomically as cysts, sinuses and fistulas by Olsen.[7]

Clinically, patients affected by BCAs commonly develop concurrent infections due to poor skin integrity and continual suppurative exudation. The most common presentation is a recurrent neck abscess in the lower third of the leading edge of the sternocleidomastoid muscle. Surgical removal is the primary treatment, and improper surgery can lead to recurrence.[8] Recurrence may occur due to poor illness identification, incorrect choice of surgery, or incomprehensive anatomical knowledge. Surgical treatment during infection is strictly prohibited and may result in a significant increase in the probability of recurrence. According to reports in the literature, the recurrence rate of BCA following surgery is 3% to 14%.[9] In addition, the recurrence rate is influenced by repeated infections, suppuration, incisions and drainage. Notably, a previous infection history and attempts at removal increase the recurrence rate to 14% to 22%.[10,11] However, recurrence causes many scars and inflamed tissues that are very difficult to distinguish from normal tissues. Conventional surgical excision has little to no curative effect. Therefore, repeated recurrence and surgical intervention can undoubtedly cause considerable physical and mental suffering to patients.

For this purpose, we performed selective neck dissection (SND) to treat patients with relapsed disease. Neck dissection is a surgical method to manage neck lymph node metastasis from a malignant tumor. The advantage of this technique is the preservation of crucial neck tissues, muscles, vessels and nerves.[12–14] Based on retaining the original structures, we performed en bloc removal to achieve complete curative resection. This article summarizes the outcomes of 216 patients

*Editor: Bernhard Schaller.*

*LL and JL contributed equally to this work.*

*This work was partly supported by the Science and Technology department of Sichuan Province (No. 2018SZ0133; Fei Chen) and (No. 2018JY0600; Jun Liu), the Science and Technology department of Chengdu (No. 2016-HM01-00167-SF; Fei Chen) and (No. 2016-HM01-00210-SF; Jun Liu), and the Science and Technology project of the health and family planning commission of Sichuan Province (No. 18PJ073; Jun Liu).*

*The authors have no conflicts of interest to disclose.*

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*Medicine (2019) 98:33(e16799)*

*Received: 17 March 2019 / Received in final form: 23 June 2019 / Accepted: 19 July 2019*

*http://dx.doi.org/10.1097/MD.0000000000016799*

**Figure 1.** Recurrent fourth branchial cleft anomalies on the right side of the neck.

with recurrent BCAs who underwent SND and further evaluates the use of this surgical procedure and its outcomes.

## 2. Methods

Approval from the Institutional Review Board of the West China Hospital of Sichuan University was obtained before this study was performed. Between January 2005 and August 2018, a total of 216 patients who were admitted to Department of Otorhinolaryngology, Head and Neck Surgery were diagnosed with recurrent second, third, and fourth BCAs. Informed consent was obtained from all patients. The hospitalization data of the patients were well documented at West China Hospital.

### 2.1. Patient characteristics

This group comprised 63 males and 153 females. The median age at diagnosis was 19 years (ranging from 4 years to 56 years). All of the patients' chief complaints were neck swelling, pain and

**Table 1**

**Patient details.**

| | No. of patients (%) |
|---|---|
| Age (yr) | |
| Range | 4–56 |
| Median | 19 |
| Gender | |
| Male | 63 (29.16%) |
| Female | 153 (70.84%) |
| M/F ratio | 1:2.4 |
| Type | |
| Second | 69 (31.94%) |
| Third | 140 (64.81%) |
| Fourth | 7 (3.25%) |
| Lesions Location | |
| Left | 145 (67.13%) |
| Second | 35 (16.20%) |
| Third | 103 (47.68%) |
| Fourth | 6 (2.78%) |
| Right | 71 (32.87%) |
| Second | 34 (15.74%) |
| Third | 37 (17.13%) |
| Fourth | 1 (0.45%) |

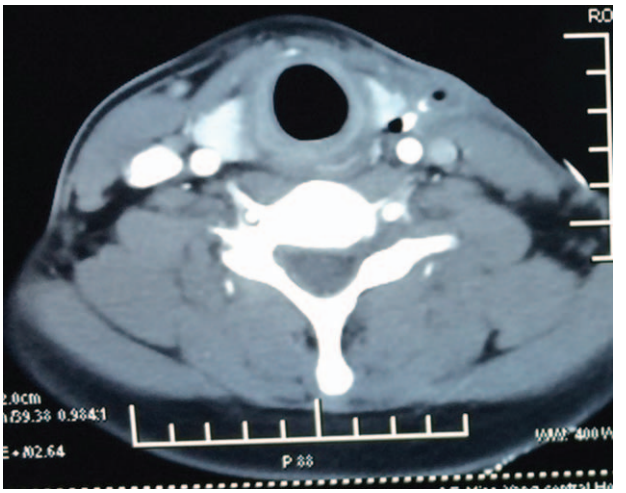

**Figure 2.** Preoperative CT showing a fistula opening at the esophagus.

fistulas. The course of recurrence ranged from 6 months to 46 years. Before this admission, each patient underwent inappropriate surgical procedures more than once. One of the patients even underwent 11 operations. According to the clinical manifestations and medical histories of the patients, 69 presented with recurrent second BCAs, 140 presented with recurrent third BCAs, and 7 presented with recurrent left rare fourth anomalies. Of the second BCAs, there were 35 on the left and 34 on the right. Of the third BCAs, there were 103 lesions on the left and 37 on the right. All fourth BCAs were located on the left side of the neck except for one (Fig. 1). The demographic characteristics and disease classifications are also summarized in Table 1. The pathway of the fistulas, the locations of cysts and the location relative to significant carotid vessels were preliminarily estimated by computed tomography (CT) and esophagography with lipiodol (Figs. 2 and 3).

**Figure 3.** Preoperative esophagography with lipiodol showing a fistula opening at the esophagus.

**Figure 4.** An arc incision was made along the previous surgical incision.

### 2.2. Surgical procedures

All patients underwent surgery during the dormant stage of the inflammatory reaction. An arc incision was made in the neck along the previous surgical incision scar, and the incision was extended appropriately (Fig. 4). When the superficial layer of the neck fascia and platysma were separated, the lesions and surrounding normal tissues were fully exposed. The margin of the exposed sternocleidomastoid muscle was also separated. Then, the sternocleidomastoid muscle was pulled away, and the carotid sheath was exposed. The accessory nerve, vagal nerve, hypoglossal nerve, superior laryngeal nerve, common carotid artery, and internal jugular vein were sequentially dissected and protected. Lesions adjacent to the carotid sheath and nerves were meticulously separated to ensure that important tissues and structures were not damaged. Along with the prevertebral fascia, the lesions and adjacent inflammatory tissues and scars were resected en bloc. If a streak tract was found between the digastric muscle and hypoglossal nerve plane, the tract was separated and ligated along the lateral wall of the pharynx. Then, the end of the lesion was sutured using a purse-string approach. If accompanied by suppurative thyroiditis or if the lesions passed through the thyroidal tissue, partial thyroid lobectomy was essential, and a small portion of the lateral thyroid cartilage was also dissected. The recurrent laryngeal nerve was dissected to avoid damage when removing lesions. The ends of third and fourth BCAs were located in the piriform sinus. To expose the lesions as much as possible, part of thyroid cartilage was excised. Dissection in the direction of the piriform sinus and the ends of the BCAs were also sutured with a purse-string approach. The surgical field after removing lesion is shown in Figure 5.

### 2.3. Postoperative treatment

Patients with second BCAs began oral feeding the day after surgery. Patients affected with third and fourth BCAs had nasogastric tubes placed and received nutritional care provided by professional nutritionists. All patients received conventional wound care and airway management.

## 3. Results

Postoperative pathological examinations confirmed that 216 lesions were BCAs, and the preoperative diagnosis was confirmed to be correct. A drainage tube was placed for 24 hours after surgery. The complication and recurrence rates are shown in Table 2. Of the 216 patients, the wounds of 203 (93.98%) patients healed primarily. Twelve (5.56%) patients had local infections, and 1 (0.46%) patient developed a pharyngeal fistula. Their wounds were treated with antibiotics and daily dressing changes for some time in the hospital before discharge. Eleven (5.09%) patients developed transient hoarseness due to the intraoperative stretching of the nerves. Unfortunately, due to scarring or tight focal adhesions that involved the recurrent laryngeal nerve, 3 (1.39%) patients developed permanent hoarseness. Two of them agreed to undergo anastomotic reconstruction of the recurrent laryngeal nerve and vocal cord adduction. The partial function of the nerve recovered, and hoarseness improved significantly. Five (2.31%) patients developed postoperative coughing when eating and drinking; however, their discomfort was improved through swallowing training.

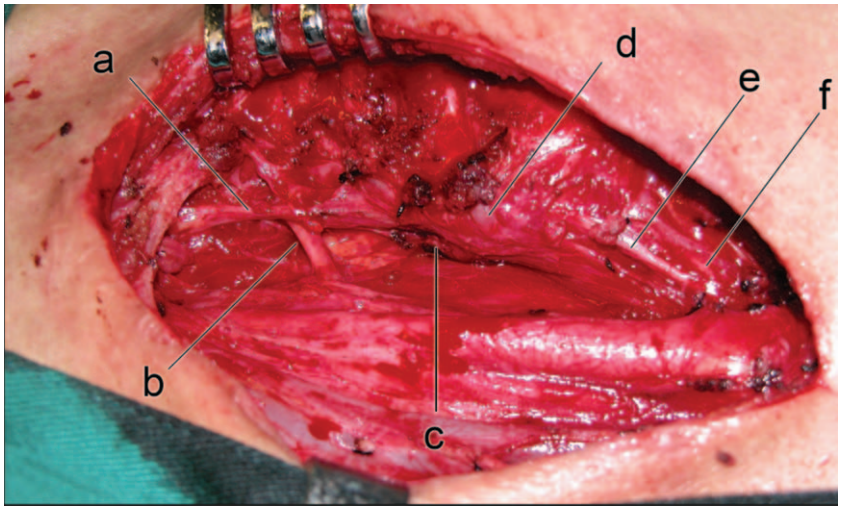


**Figure 5.** The surgical field after removing lesions from patients with fourth BCAs (A: the recurrent laryngeal nerve; B: the inferior thyroid artery; C: a fistula opening in the piriform sinus; D: the thyroid; E: the superior thyroid artery; F: the superior laryngeal nerve).

**Table 2**

**Complications.**

| | No. of patients (%) |
|---|---|
| Local infection | 12 (5.56%) |
| Pharyngeal fistula | 1 (0.46%) |
| Nervous injury | |
| Temporary vocal cord palsy | 11 (5.09%) |
| Permanent hoarseness | 3 (1.39%) |
| Reconstruction of RLN | 2 (0.92%) |
| Bucking | 5 (2.31%) |
| Vessel injury | |
| Rupture of internal jugular vein | 3 (1.39%) |
| Recurrence | 4 (1.85%) |
| Second | |
| Recurrent rate | 1/69 (1.45%) |
| Third | |
| Recurrent rate | 3/140 (2.14%) |
| Fourth | |
| Recurrent rate | 0% |
| Total cure rate | 212/216 (98.15%) |

RLN = recurrent laryngeal nerve.

Additionally, 3 (1.39%) patients underwent internal jugular vein ligation because of jugular rupture and bleeding.

All patients were followed up for 1 to 9 years. Of the patients with recurrence, 212 were cured by a one-time operation, and the cure rate was 98.15%. A total of 4 (1.85%) patients had recurrence once more, including 1 (0.46%) patient with a second BCA, 3 (1.39%) patients with third BCAs and none with fourth BCAs. The remaining patients with recurrence were cured by reoperation.

## 4. Discussion

Developmental anomalies related to the fusion and differentiation of the branchial apparatus are well described in the literature.[15,16] BCAs can present a cyst, sinus, or fistula. Fistulae present as internal and external openings. Cysts are described as secretion retention in tissue spaces without openings. In addition, one opening in the neck skin or pharynx is called a sinus. The 3 categories are interchangeable rather than fixed. Frequent inflammation can cause 1 or 2 fistula openings to become sealed and turn into a sinus or cyst. Due to the complex structures of recurrent BCAs, preoperative ultrasound is not better than CT or MRI. CT and MRI can more clearly display the shape, location, origin and insertion, course and adjacent relationships of recurrent lesions to reduce recurrence.[17]

Surgical removal is the primary treatment, and improper operation can lead to recurrence. The complexity of BCA anatomy is the primary cause of recurrence. Some doctors lack the clinical experience necessary for proper treatment and have insufficient comprehension of this disease. In addition, the conventional surgical decision for BCAs is a trapezoidal incision.[18] A surgeon may omit repair of an anomaly in a limited operative field due to a small primary surgical incision. If there are many cystic lobulations or fistula branches, a surgeon may also omit repair in a limited operative field. Due to repeated inflammatory stimulation and surgical intervention, the lesions that are adhered to peripheral structures are impossible to identify to the full extent and may lead to relapse again. Meanwhile, inappropriate surgical timing has implications for patient prognosis. Waldhausen found that infection can increase the recurrence rate.[19] During the acute inflammatory period, there is a breakdown in the integrity of the epithelium and an unclear field of view, which leads to an increased recurrence rate. In addition, the operator's surgical skills contribute to a certain proportion of the curative effect.

For the past few years, neck dissection has been applied in the treatment of recurrent congenital neck disease. Blackwell et al creatively used functional neck dissection to treat recurrent BCAs, and no patient relapsed.[20] The most distinctive feature of neck dissection is the ability to treat recurrent second, third and fourth BCAs via overall removal. The anatomical plane of the neck is not affected by postoperative adhesion or scarring, allowing complete removal of recurrent lesions, indistinguishable scarring and inflammatory tissue to prevent residual epithelial formation and tearing of tracts due to separation of adhesions and infected tissues. Because of the integrity of epithelial tissue resection, neck dissection is currently considered the best procedure to treat and prevent recurrence.[21] It is worth mentioned that the re-recurrence rate was only 1.85% among the 216 patients in our study. This outcome is related to our successful neck dissection techniques.

However, combined with evaluation of our patients and the reported literature, very few third and fourth anomalies have been identified to completely follow the classical pathways and parts of the lesions that can invade or penetrate the thyroid gland.[22] Therefore, for repeated episodes of acute suppurative thyroiditis, third and fourth BCAs must first be considered. If the branches and local anatomical structures are ignored, inappropriate surgical techniques can easily lead to relapse. As with the majority of surgeons' discoveries, the locations of third and fourth BCAs are predisposed to be on the left side. The emergence of this tendency may be explained by the asymmetrical development of the fourth branchial arch.[22] For second BCAs, the previous view was that ipsilateral tonsillectomy was effective for reducing recurrence.[23] However, in our study, none of the patients with recurrent second BCAs underwent ipsilateral tonsillectomy. In terms of the rate of recurrence, only 1 in 69 recurred again. Our study results from numerous surgeries indicate that ipsilateral tonsillectomy is not associated with recurrence.

For the sake of postoperative function and quality of life, organs must be kept intact while removing lesions. Management of adhesions between lesions and organs during exposure necessitates that surgeons be adept at various techniques. If the lesions adhere closely to or pass through the muscle, partial muscle resection is required to achieve en bloc resection of lesions. When removing muscles, careful dissection should be performed to reduce local tissue damage and avoid injury of important nerves. The pathways of third and fourth BCAs are closely related to the thyroid, superior laryngeal nerve and recurrent laryngeal nerve.[22,24] Vocal cord paralysis is the most common postoperative complication of BCAs. Nicoucar et al reported that the incidence of vocal cord paralysis is 3.71%.[25] Therefore, dissection of the thyroid and nerves is particularly important during surgery. When the lesions adhere tightly to or cross over the thyroid, lateral partial thyroidectomy is necessary to reduce recurrence effectively.[25,26] During resection of the lesion, surgeons should pay close attention to the upper pole of the thyroid and stay away from the lower pole. It is also feasible to perform nerve detection when necessary.

If a lesion is closely adherent to the important tissues of the carotid sheath (the most common adhesion is to the internal

jugular vein), the surgeon needs to be alert when removing the lesion because the consequences of the injury are difficult to bear. The complications of the patients in this study were significantly associated with tight adhesion lesions. Attempted separation of the lesions resulted in damage to the internal jugular vein, superior laryngeal nerve, and recurrent laryngeal nerve.

To completely remove the lesions, various methods were applied to visualize fistulas, sinuses and cysts. However, we think that the preoperative use of methylene blue or esophagography for recurrent lesions as a tracer is not necessary. Because of tissue adhesion and scarring, most dye tracers cannot reach the distal end of the fistula. Furthermore, for some branch lesions, the effect of methylene dye is insufficient, leading to incomplete resection and residual lesions. Furthermore, methylene spillover also influences surgical procedures. It is difficult for contrast medium to access the minute branch tracts because of surface tension. Rattan and Cai also hold the same views as ours, which they obtained through analyzing a large number of clinical records.[8,27]

In recent years, endoscopic cautery of the internal fistula has become a promising therapeutic approach for BCAs (only for fistulae and sinus tracts).[17,28] Compared to traditional open surgery, this method is minimally invasive and may reduce complications. Leboulanger et al achieved a fine effect for patients with BCAs by endoscopy.[29] However, the cauterization method has not been reported to be effective in patients with postoperative recurrence.

According to the various anatomical characteristics, we applied SND to treat recurrent second, third, and fourth BCAs, and we achieved our goal of good radical cure. Our experience has revealed that, through neck dissection, postoperative recurrent lesions along with inflammatory tissue and severe scar tissue can be completely en bloc resected while contouring the sternocleidomastoid muscle and carotid sheath. In addition, the cervical blood vessels and recurrent laryngeal nerve can be well preserved. In other words, the incidence of complications was also significantly lower than that with other procedures.

## 5. Conclusion

SND provides the virtues of minimal trauma, few complications and a low recurrence rate. Compared to other surgical methods, it is an effective method to treat recurrent second, third, and fourth BCAs.

## Author contributions

**Conceptualization:** Linke Li, Fei Chen.
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**Funding acquisition:** Linke Li, Fei Chen.
**Investigation:** Di Deng, Fei Chen.
**Methodology:** Jun Liu, Dan Lv, Di Deng.
**Project administration:** Tian Shen.
**Resources:** Jun Liu, Ji Wang, Fei Chen.
**Software:** Linke Li, Tian Shen.
**Supervision:** Jun Liu.
**Validation:** Jun Liu, Dan Lv.
**Visualization:** Jun Liu, Dan Lv, Ji Wang.
**Writing – original draft:** Linke Li, Jun Liu, Fei Chen.
**Writing – review & editing:** Linke Li, Jun Liu, Fei Chen.

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