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LITERATURE REVIEW

Management of congenital third branchial arch anomalies: A systematic review

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ABSTRACT

OBJECTIVE: To systematically review the existing literature on third branchial arch anomalies and suggest guidelines for their management.

DATA SOURCES: We searched PubMed, Medline, and Embase using Scopus, and collected additional publications cited in bibliographies. We included all English-language articles and all foreign-language articles with an English abstract.

REVIEW METHODS: Two investigators reviewed all cases explicitly identified as third arch anomalies or meeting anatomical criteria for third arch anomalies; they assessed presentation, diagnostic methods, intervention, and outcome.

RESULTS: We found 202 cases of third arch anomalies; they presented primarily on the left side (89%), usually as neck abscess (39%) or acute suppurative thyroiditis (33%). Barium swallow, direct laryngoscopy, and magnetic resonance imaging were the most useful diagnostic tools. The recurrence rate varied among the treatment options: incision and drainage, 94 percent; endoscopic cauterization of the sinus tract opening, 18 percent; open-neck surgery and tract excision, 15 percent; and partial thyroidectomy during open-neck surgery, 14 percent. Complications after surgery appeared somewhat more frequently in children eight years of age or younger.

CONCLUSION: Third arch anomalies are more common than previously reported. They appear to be best treated by complete excision of the cyst, sinus, or fistula during a quiescent period. Repeated incision and drainage yields high rates of recurrence and should be avoided. Complications might be minimized by first initiating antibiotic treatment, delaying surgical treatment until the inflammatory process is maximally resolved, and by using endoscopic cauterization.

The branchial arches give rise to specific derivates, which for the third arch include the superior constrictor muscles, internal carotid artery, ninth cranial nerve, greater horn, and body of hyoid bone. The third branchial pouch gives rise to the inferior parathyroid gland and, by migrating caudally along the thymopharyngeal duct, the thymus. Branchial arch anomalies may occur as fistulas, sinus tracts, or cysts, and are responsible for approximately 17 percent of all pediatric neck masses.[1] Branchial cysts have no external or visceral opening and thus retain secretions, while branchial sinuses communicate with either the skin or the pharyngeal lumen, and fistulas are tracts connecting the pharynx to the skin.

Most branchial anomalies arise from the second branchial apparatus; third branchial anomalies represent only two to eight percent of all branchial anomalies.[2,3] Because of their rare occurrence, consistent recommendations on diagnosis and management are lacking, and reports on recurrence and complication rates vary widely. To augment this limited information, we sought to review all published cases of third branchial anomalies in order to summarize their clinical features and to identify the best options for diagnosis and treatment. Our review follows the model of a previous review that we recently published, examining the features of all published cases of fourth branchial arch anomalies.[4]

Materials and Methods

Literature Search and Selection of Cases

We searched the PubMed, Medline, and Embase databases using Scopus with the following word combinations: (“third” AND “branchial” AND “cyst”) or (“pyriform” AND “sinus” AND “fistula”) or (“thymopharyngeal” AND “duct”).We considered all English-language articles plus all foreign-language articles that provided an abstract in English. We restricted the search to human studies published between 1968 and 2006. Of the 216 articles retrieved, 163 were excluded because they represented review articles on the subject (12), dealt with other congenital malformations or arch anomalies (119), described complications of surgery for tumors (18), or were laboratory studies on animals (14). A total of 53 publications remained. From the references of

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the selected articles, an additional 37 publications were identified and included. Unpublished reports were not considered.

Data Extraction

The first 90 selected publications, which included case reports, case series, and chart reviews, often included cases with other congenital anomalies in addition to cases of third branchial arch anomalies (Table 1, available online only, www.otojournal.org). Thus, we employed specific inclusion criteria to identify and segregate third arch cases, requiring either that the case be 1) explicitly identified as a third branchial arch anomaly by the authors, or 2) characterized by a clear description of a tract starting in the base of the pyriform sinus and exiting through the thyrohyoid membrane. These criteria were especially important for distinguishing between third and fourth branchial arch anomalies because both can present with suppurative thyroiditis or recurrent deep neck infection and both have a left-sided predominance. However, third and fourth arch anomalies are distinguished by anatomically different tracts. In the present review, we encountered eight reports, collectively describing 28 cases, where the distinction between third and fourth arch origin was equivocal. For example, in one report, cases were described as "remnant[s] of either the third or fourth branchial pouch."[5] For the purposes of this review, we decided to exclude these eight reports and their 28 equivocal cases from our analysis.

A flowsheet summarizing our selection of publications and of cases is shown in Figure 1. Among the 604 cases presented in the selected publications, 402 were excluded from analysis, mostly because they represented anomalies of first, second, or fourth branch origin. Thus, we were left with 202 cases meeting our inclusion criteria.

Two investigators (K.N. and R.G.) independently reviewed all cases and extracted the following information from each article: sex, side of presentation, age of onset, age of patient at diagnosis, mode of presentation, diagnostic investigations, histological findings, treatment, complications, and recurrence rate. With regard to mode of presentation, we did not score acute suppurative thyroiditis unless there was radiographic confirmation. With regard to recurrence rate, we did not require a specific minimum period of follow-up because the reports varied in detail of presentation, and we did not want to limit our information only to the subset of reports with explicit follow-up criteria. Discordant findings between the two reviewers were discussed and reexamined in order to reach agreement.

Data Analysis

Data were scored as numeric or categorical variables. We first calculated summary statistics and then performed several post hoc two-way comparisons of subgroups of cases. We compared these subgroups using the Wilcoxon rank sum test to compare continuous variables and Fisher's exact test to compare proportions, with alpha set at 0.05, two-tailed. Most of these statistical tests involved comparisons of individuals presenting with acute suppurative thyroiditis versus individuals with other presentations (Table 2). Although we quote crude odds ratios and 95% confidence intervals in Table 2, it should be noted that we repeated all of the comparisons in Table 2 using Mantel-Haenszel tests with stratification for study site. These latter tests were very limited in power, however, because many of the reports involved only one or two patients, and thus only a small number of the strata formed in these analyses actually contributed information about the effect of a given explanatory variable. Nevertheless, these analyses produced results generally congruent with the crude odds ratios, as detailed in the footnote to Table 2.

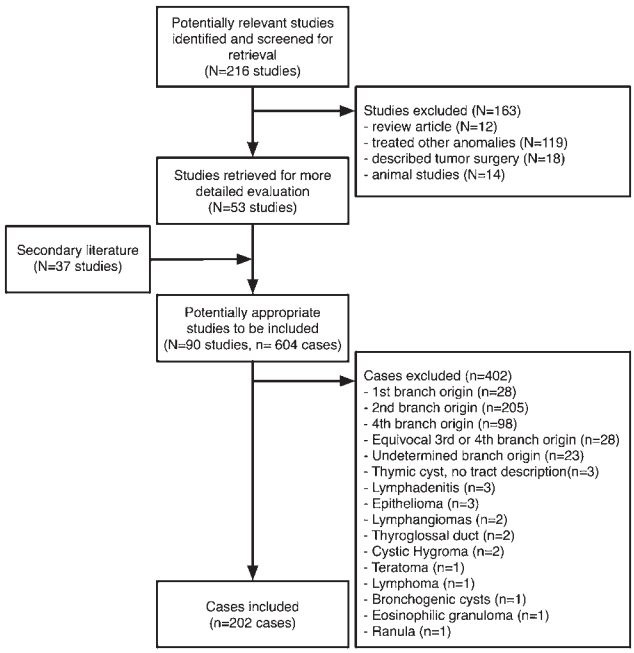

Figure 1 Flow diagram of publication and case selection.

Results

Age and Clinical Presentation

The 82 selected publications,[2,3,6-85] presenting 202 cases of third branchial arch anomalies, are summarized in Table 1. Most third branchial arch anomalies were diagnosed in childhood (Table 3). The great majority of cases occurred on the left side (89%) and presented either as neck abscess or acute suppurative thyroiditis (Table 3). Patients presenting with acute suppurative thyroiditis were older at the time of diagnosis than other cases (Table 2). Of the 202 identified cases, 24 occurred in neonates, and 10 (42%) of these presented with respiratory distress. Of the 16 cases aged one to three years, six (38%) presented with acute suppurative thyroiditis and five (31%) with neck abscess. Among 43 cases aged four to eight years, 13 (30%) presented with acute suppurative thyroiditis and 22 (51%) with neck abscess. Of the 119 cases older than eight years of age, 50

Table 2
Features of patients presenting with acute suppurative thyroiditis versus patients with other presentations

Clinical presentation	AST	Others	Odds ratio [95% CI]	*P* value*
Total cases, n (%)	69 (34)	133 (66)		
Male, n (%)	23 (33)	76 (57)	0.4 [0.2, 0.7]	0.002
Age at diagnosis (yrs), median yrs (range)	12 (0-44)	9 (0-67)	†	0.08
Diagnostic methods, n (%)				
Barium swallow	60 (87)	72 (54)	5.6 [2.5, 13.9]	<0.001
Laryngoscopy	31 (45)	59 (44)	1 [0.5, 1.9]	1.0
CT scan	17 (25)	64 (48)	0.4 [0.2, 0.7]	0.001
Ultrasound	24 (35)	30 (23)	1.8 [0.9, 3.6]	0.07
Thyroid scan	23 (33)	15 (11)	3.9 [1.8, 8.8]	<0.001
Fine needle aspiration	15 (22)	13 (10)	2.6 [1.1, 6.3]	0.03
MRI	8 (12)	17 (13)	0.9 [0.3, 2.3]	1.0
Radiographs	1 (1)	19 (14)	0.1 [0.0, 0.6]	0.002
Treatment, n (%)				
Incision and drainage	18 (26)	50 (38)	0.6 [0.3, 1.2]	0.1
Number of attempts, median (range)	2 (1-5)	2 (1-28)	†	0.7
Failure after initial attempt	15 (83)	49 (98)	0.1 [0.0, 1.4]	0.05
Open-neck surgery initial procedure	30 (43)	70 (53)	0.7 [0.4, 1.3]	0.2
Open-neck surgery procedure after failure	9 (13)	49 (37)	0.3 [0.1, 0.6]	<0.001
Number of procedures, median (range)	1 (1-5)	1 (1-8)	†	0.3
Failure after open-neck surgery	4 (10)	20 (17)	0.1 [0.1, 1.9]	0.4
Partial thyroid surgery	9 (13)	13 (10)	1.4 [0.5, 3.7]	0.5
Failure after initial procedure	0	3 (2)	‡	0.2
Endoscopic cauterization initial attempt	19 (28)	9 (7)	5.2 [2.1, 14.0]	<0.001
Endoscopic cauterization after failure	4 (6)	1 (1)	8.1 [0.98, 403]	0.05
Number of attempts, median (range)	1	1 (1-2)	†	0.007
Failure after endoscopic cauterization	2 (9)	4 (40)	0.1 [0.0, 1.0]	0.05
Complications after open-neck surgery¶	0	10 (8)	‡	0.12

AST, acute suppurative thyroiditis; *MRI*, magnetic resonance imaging.
*By Fisher's exact test for differences between proportions and the Wilcoxon rank sum for differences between continuous variables. Note that the significance of these differences was also calculated using Mantel-Haenszel tests with stratification for study site. The findings from the Mantel-Haenszel tests were generally congruent with the findings computed without stratification. However, given that many of the reports involved only one or two patients, only a small number of the strata formed in these latter analyses actually contributed information about the effect of a given explanatory variable. Thus, the Mantel-Haenszel tests were of very limited power.
†Odds ratio does not apply because the difference is between median values.
‡Odds ratio cannot be calculated because of a zero value.
¶See text for definition of complications.

(42%) presented with acute suppurative thyroiditis and 56 (47%) with neck abscess.

Pathology Findings

Histopathological examination was reported in 72 of the 202 identified cases (36%); of these, 42 (58%) demonstrated a stratified squamous epithelium and 25 (35%) a respiratory ciliated epithelium. Thymic, thyroid, and parathyroid tissues were reported in 40 percent, 19 percent, and 10 percent of these cases, respectively. A cystic lesion was described in 54 percent of cases.

Diagnostic Investigations

Investigations used and positive predictive values (ppv) were as follows: barium swallow in 65 percent of cases (ppv 81%), direct laryngoscopy in 45 percent (ppv 82%), magnetic resonance imaging (MRI) in 12 percent (ppv 84%), computed tomography in 40 percent (ppv 49%), ultrasound in 27 percent (ppv 7%), and thyroid scan in 19 percent (ppv 3%). Sixty-five cases (32%) were diagnosed during the surgical procedure. Radiographs and fine needle aspiration did not contribute to the diagnosis in any of the reported cases. The choice of investigations varied depending on whether the patient presented with acute suppurative thyroiditis or another clinical picture (Table 2).

Treatment and Outcome

Initial treatments performed in reported cases were as follows: incision with drainage in 68 (34%) of the cases, open-neck surgical procedures in 100 (50%), endoscopic procedures in 28 (14%), antibiotic treatment alone in five (2%), and no treatment in one case. Considering all treatments administered, as opposed to just the initial treatments, we found that two (8%) of the 24 neonates received incision and drainage, and all 24 (100%) ultimately received open-neck surgery (with partial thyroidectomy in 3 cases [13%]).

Table 3
Features of 202 cases of branchial arch anomalies

Male, n (%)	99 (49)
Age at onset (yrs), median (range)	5 yrs (0-60)
Age at diagnosis (yrs), median (range)	10 yrs (0-67)
Site of presentation, n (%)	
Left	180 (89)
Right	22 (11)
Initial presentation, n (%)	
Neck abscess (NA)	79 (39)
Acute suppurative thyroiditis (AST)	66 (33)
Neck mass (NM)	28 (14)
Cutaneous discharging fistula with NA or AST	11 (5)
Cutaneous discharging fistula	8 (4)
Respiratory distress with NA, AST, or NM	6 (3)
Respiratory distress	4 (2)

NA, neck abscess; *AST*, acute suppurative thyroiditis; *NM*, neck mass.

Of the 16 patients younger than three years of age, four (25%) received incision and drainage, 13 (81%) open-neck surgery (with partial thyroidectomy in 1 case [6%]), one (6%) endoscopic cauterization, and one (6%) received antibiotic treatment alone. Of the 43 children between four to eight years of age, 18 (42%) received incision and drainage, 41 (95%) open-neck surgery (with partial thyroidectomy in 5 cases [12%]), and two (5%) received antibiotic treatment alone. Finally, among the 119 cases older than 8 years of age, 44 (37%) received incision and drainage, 80 (67%) open-neck surgery (with partial thyroidectomy in 13 cases [11%]), 32 (27%) endoscopic cauterization, and four (3%) antibiotic treatment alone; one (1%) received no treatment.

Returning to the overall sample, and looking at the 68 individuals who initially received incision with drainage, we found that the failure rate after the first attempt was 64 (94%). Repeated attempts at incision and drainage were performed in 37 (58%) of the initially failed cases. Only four cases (6%) were successfully treated by incision and drainage alone; almost all cases ultimately needed another treatment (open-neck surgery with or without partial thyroidectomy or endoscopic tract cauterization). Among patients initially treated with incision and drainage, the median number of incision and drainage attempts was two (range 1-28). A total of 158 patients received an open-neck surgical procedure, either initially (n = 100) or following the failures of other treatment options (n = 58). Of these 158 cases, 24 exhibited recurrence; there was no significant difference in the rates of recurrence among the 22 patients who underwent surgery that included a partial thyroidectomy (3 [14%] relapsed) and the 136 patients who did not receive a partial thyroidectomy (21 [15%] relapsed; odds ratio [OR] [95% confidence interval], 0.9 [0.2, 3.3]; $P = 1.0$). Comparing patients presenting with acute suppurative thyroiditis with those displaying other presentations, we found that the former group demonstrated somewhat fewer recurrences after endoscopic cauterization (2 [9%] of 23 cases vs 4 [40%] of 10 cases; OR, 0.1 [0.0, 1.4]; $P = 0.053$). Among the 100 patients initially treated with open-neck surgery, 95 (95%) required only one procedure, four (4%) required two procedures, and one (1%) required five procedures. Ten (6%) of the 158 patients who underwent open-neck surgery developed complications (summarized in Table 4). Children eight years of age or younger demonstrated a somewhat higher complication rate after open-neck surgery than older patients (8 [10%] of 78 cases vs 2 [3%] of 80 cases; OR, 4.5 [0.8, 44.0]; $P = 0.055$). Among the 33 patients who received endoscopic procedures (as either an

Table 4
Reported complications of open-neck surgery for third branchial arch anomalies

Study	Case (n)	Sex	Side	Age at onset/ diagnosis (yrs)	Presentation	Open-neck surgeries (n)	Partial thyroid surgery	Complication
Lee[40] (1991)	1	M	L	13/22	CDF	2	No	Vocal cord paralysis
Ostfeld[58] (1991)	1	M	L	21/22	NA	1	No	IX/X/XII Cranial nerve paralysis
Singh[74] (1979)	1	F	L	4/4.5	NM	1	No	Facial nerve paralysis
Vade[79] (1994)	1	F	L	Birth	NM	1	No	Wound infection
Lin[43] (1991)	8	M	L	NR/6	CDF	1	No	Salivary fistula
Lin[43] (1991)	9	F	L	NR/7	CDF	2	No	Salivary fistula
Lin[43] (1991)	10	F	L	NR/8	CDF	1	No	Salivary fistula
Ford[3] (1992)	2	F	R	Birth	RD	1	No	Vocal cord paralysis and Horner syndrome
Franciosi[22] (2002)	1	M	L	Birth	NM RD	2	No	Vocal cord paralysis
Lowry[45] (2002)	1	F	L	Birth	RD	1	No	Vocal cord paresis

NR, not reported; *CDF*, cutaneous discharging fistula; *RD*, respiratory distress; *NM*, neck mass; *NA*, neck abscess.

initial or subsequent treatment), a majority were female (73%). Six relapsed after the initial procedure (failure rate 18%), and three of these received a second endoscopic procedure. No complications were reported following this treatment option. Among the patients treated conservatively, five received only antibiotics and one received no treatment. No recurrences were reported in these six patients.

Discussion

Anatomically, third branchial arch anomalies represent vestiges of a tract that originates from the base of the pyriform sinus (Fig 2). It crosses the thyrohyoid membrane, then travels upward to loop around the hypoglossal nerve below the glossopharyngeal nerve before running downward posterior to the common or internal carotid artery and anterior to the vagus nerve. The external opening, if present, is typically found along the anterior border of the junction of the middle and lower third of the sternocleidomastoid muscle.[86] There is no difference between the left and right side, which contrasts with the anatomy of sinus tract remnants from the fourth branchial arch. However, third and fourth branchial arch anomalies have similar clinical presentations, and some authors have even suggested grouping them as the same entity. Nevertheless, these two types of anomalies are anatomically distinct: third branchial arch anomalies are proposed to originate from the base (cranial end) of the pyriform sinus and pass above the superior laryngeal nerve, whereas the tract of fourth branchial arch anomalies originates from the apex (caudal end) of the pyriform sinus and passes through the cricothyroid membrane beneath the superior laryngeal nerve.[22]

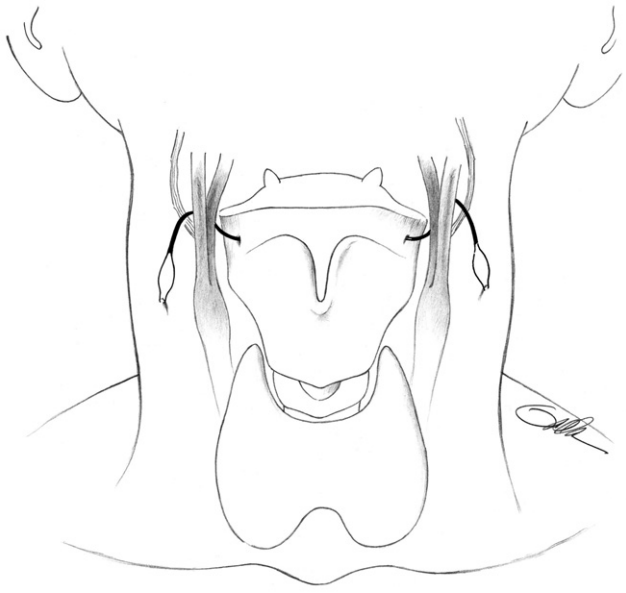

Figure 2 Theoretical pathway of a third branchial arch fistula.

The thymus and the inferior parathyroid glands develop from the third branchial pouch. A spectrum of anomalies is attributed to the embryologic descent of the thymic primordium into the mediastinum and the caudal migration of cells from the inferior parathyroid glands. For example, thymic cysts result from persistent remnants of the thymopharyngeal duct. The majority of these anomalies are found on the left side of the neck. Cervical thymic cysts should not be removed in toto before the age of five years, to avoid serious repercussions on the immune system.[87] Parathyroid cysts can be found anywhere around the thyroid gland, but they are most commonly located inferior to the gland. Although parathyroid cysts are usually not associated with biochemical anomalies, there have been reports of hyperparathyroidism secondary to functioning cysts.[88] In our review, no hormone imbalances were described in the two cases with parathyroid cysts.[12]

Our review yielded 202 reported cases of third branchial arch anomalies that met our criteria. This number is much larger than previously reported.[2,3] However, we identified fewer cases than in our review of fourth branchial arch anomalies.[4] We believe that this smaller number of cases is unlikely to be due to a failure to identify relevant articles, since we used the same comprehensive search methodology used in our previous review of fourth branch anomalies and screened every reference cited in every identified publication to locate additional articles. Our smaller number of cases might be partially explained by our selection criteria, since we rejected a number of articles (reporting on a total of 402 cases), some of which may have included third arch cases but where the third arch origin was not clearly established or documented. The smaller number of cases might also be attributable to a lower incidence of infection in third arch anomalies: due to the ascending trajectory of the tract compared to the descending trajectory of fourth branchial arch, third arch anomalies are much more likely to remain clinically silent. Also, it is likely that for all types of branchial arch anomalies, only a minority of cases are published in the literature. The true prevalence of third arch anomalies is therefore likely to be much higher than the number of cases we have identified in this review.

We found a strong left-sided predominance of third arch anomalies (89% vs 11% on the right side); this phenomenon may be related to the trajectory of the neighboring fourth branchial arch, which also yields predominantly left-sided anomalies.[4] We found that direct laryngoscopy and barium swallow were the primary methods of diagnosis of third branchial arch anomalies, with MRI used primarily in the presence of cystic anomalies.

A range of treatment options were reported in the cases reviewed. Incision with drainage was frequently performed, but the initial treatment failed in a majority of cases. In neck abscesses without thyroid involvement, open-neck surgery with excision of the fistula tract was performed most commonly. Partial thyroidectomy was perfomed in most cases of acute suppurative thyroiditis. The failure rate with or

without partial thyroidectomy was similar in these cases. Our data suggest that open-neck surgery may yield a somewhat higher rate of complications in children aged eight years or younger (Table 4). The accidental removal of important immune organs, with potentially serious long-term consequences, must also be considered with this treatment option in patients younger than five years of age. Endoscopic cauterization of the sinus tract represents a minimally invasive therapeutic alternative to open-neck surgery. This technique has become increasingly popular, and indeed might now deserve consideration as initial therapy for third arch anomalies.[37,38,62] Endoscopic cauterization of the opening tract has been reported since the late 1990s. With the patient under general anesthesia, the opening tract is exposed by suspension laryngoscopy, and the internal opening is cauterized in a circumferential fashion with a trichloroacetic acid solution at two different concentrations (20%-40%).[37,62] Cauterization is performed at the same time as the drainage of the neck abscess, and oral feeding is started on the second postoperative day. The failure rate with this technique was comparable to that with traditional open-neck surgery (18% vs 15%), indicating that it might sometimes need to be repeated, especially in cases presenting with neck abscess. However, reports of cases with third branchial arch anomalies treated with this technique are few, so these impressions must be considered preliminary.

Based on the findings of the present review of published cases, we suggest the following treatment algorithm for patients presenting with neck abscess or acute suppurative thyroiditis attributable to third branchial arch anomalies (Fig 3). In patients up to about the age of eight years, it seems preferable to favor medical treatment and delay surgical neck exploration. After the age of eight years, a complete excision of the entire fistula tract during a quiescent period appears to be the treatment of choice. We found no advantage to the inclusion of a partial thyroidectomy in cases of third branchial arch anomalies. A conservative approach without surgery is possible in selected cases, but the low recurrence rate among the conservative cases found in the present literature review may be partially attributable to publication bias. Therefore, in cases of recurrence after conservative treatment, a surgical approach would probably be indicated.

In conclusion, it should be recognized that our review is limited by the fact that it is based on a large and heterogeneous collection of reports, many of them describing only one or two cases, and varying in their methodology and details of presentation. Thus, this literature does not permit sophisticated statistical analysis of the type normally performed in a meta-analysis of studies with larger samples and more quantitative information. Nevertheless, our review suggests several general conclusions. Specifically, it appears that third branchial anomalies have a varied clinical presentation and are more frequent than previously reported. During the last decade, several advances in the recognition and management of this anomaly have been made. Incision and drainage yields a high recurrence rate. Complete excision of the entire fistula tract during a quiescent period remains the treatment of choice for the management of third branchial anomalies. Complications of this surgical approach appear to be somewhat more frequently reported in children aged eight years and younger; thus, even though the number of reported cases treated with endoscopic cauterization is limited, our review tentatively suggests that it may be preferable to use endoscopic cauterization treatment in this younger-age group.

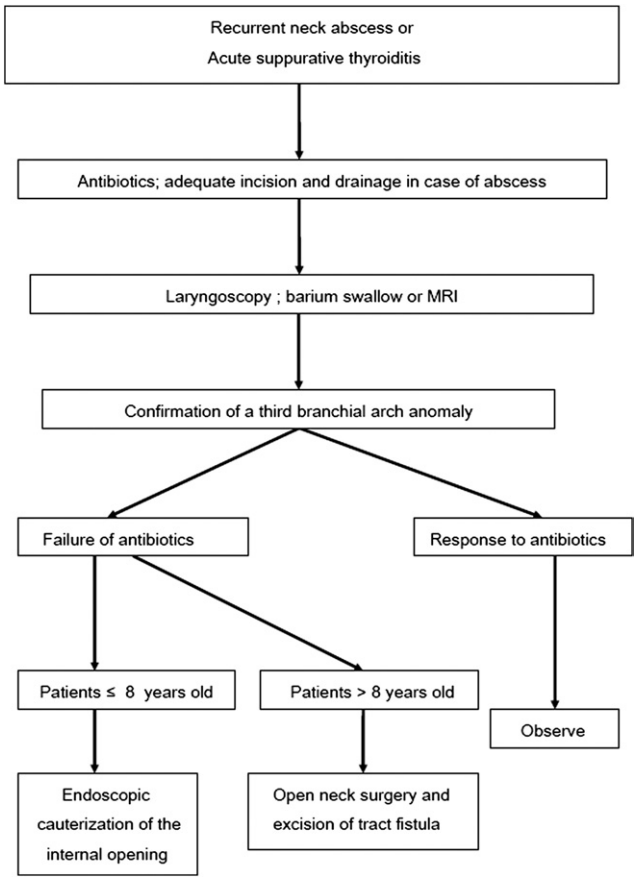

Figure 3 Suggested algorithm for the management of congenital third branchial anomalies.

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Disclosures

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Table 1
Articles included in the review and relevant descriptive details

Author	Year	Geographic origin	Sample size,* n = 202/576	Study design	Diagnostic methods	Final therapy used	Previous treatment
Abe et al[6]	1981	Japan	3/3	Case report	BS	Surgery	—
Al-Akeely et al[7]	2005	Saudi Arabia	1/1	Case report	CT	Surgery	—
Ballesteros et al[8]	1998	Spain	1/1	Case report	BS	Surgery	—
Batuecas Caletrio et al[9]	2006	Spain	1/1	Case report	MRI	Surgery	—
Boyd et al[10]	1993	USA	1/1	Case report	CT Surgery	Surgery	—
Boysen et al[11]	1979	Norway	2/4	Case series	BS, laryngoscopy, surgery	Surgery	—
Braccini et al[12]	2000	France	2/2	Case report	Surgery	Surgery	—
Burton et al[13]	1995	USA	2/2	Case report	CT	Surgery	—
Chang et al[14]	2002	Taiwan	8/11	Case series	BS	Surgery	—
Chi et al[15]	2002	Taiwan	5/15	Case series	BS	I&D (3), antibiotics (2)	Antibiotics
Chin et al[16]	2000	USA	2/2	Case report	CT, surgery	Surgery	—
Choi et al[2]	1995	USA	4/52	Chart review	CT	Surgery	I&D
Cigliano et al[17]	2004	Italy	1/1	Case report	BS, laryngoscopy	EC	—
Doi et al[18]	1988	Australia	1/58	Chart review	NR	Surgery	NR
Dutt et al[19]	1994	India	1/1	Case report	Laryngoscopy	Surgery	I&D
Edmonds et al[20]	1997	USA	3/3	Case report	Laryngoscopy, CT	Surgery	I&D
Elahi et al[21]	1997	Canada	3/4	Case report	BS, laryngoscopy	Surgery	I&D
Ford et al[3]	1992	UK	2/106	Chart review	BS, surgery	Surgery	I&D
Franciosi et al[22]	2002	USA	2/3	Case report	BS, laryngoscopy, surgery	Surgery	I&D
Gan et al[23]	2004	Singapore	5/5	Case series	BS, CT, US	Surgery	—
Gibbs et al[24]	2004	USA	1/1	Case report	CT	Surgery	—
Goudreau et al[25]	1985	Canada	1/1	Case report	BS, laryngoscopy	Surgery	I&D
Guerrero Fernandez et al[26]	2002	Spain	1/1	Case report	BS	Surgery	I&D
Hamoir et al[27]	1998	Belgium	1/43	Chart review	BS, laryngoscopy	EC	I&D, surgery
Harding et al[28]	2005	Australia	2/6	Case series	Surgery	Surgery	I&D
Hermann et al[29]	1992	USA	1/1	Case report	BS, CT	Surgery	—
Hewel et al[30]	1996	USA	1/1	Case report	Surgery	Surgery	I&D
Hsin et al[31]	1998	UK	1/1	Case report	BS	Surgery	I&D
Hsu et al[32]	1998	Taiwan	1/1	Case report	BS	Surgery	I&D
Huang et al[33]	2000	USA	1/1	Case report	Laryngoscopy, surgery	Surgery	I&D
Johnsen et al[34]	1976	Denmark	1/1	Case report	Surgery	Surgery	—
Kaufmann et al[35]	2001	USA	2/2	Case series	Surgery	Surgery	—
Kartheuser et al[36]	1986	France	1/1	Case report	BS	Surgery	I&D
Kim et al[37]	2004	Korea	14/30	Chart review	BS, CT, laryngoscopy	EC	I&D
Kim et al[38]	2000	Korea	2/18	Case series	Laryngoscopy	EC	—
Kubota et al[39]	1997	Japan	1/4	Case series	Laryngoscopy	Surgery	I&D
Lee et al[40]	1999	Taiwan	7/7	Case series	BS, laryngoscopy	Surgery	I&D
Lee et al[41]	1991	Malaysia	1/3	Case series	BS, laryngoscopy	Surgery	I&D
Lin et al[42]	2003	Taiwan	1/1	Case report	BS, laryngoscopy	None	—
Lin et al[43]	1991	Taiwan	16/16	Case series	BS, surgery	Surgery	—
Liberman et al[44]	2002	Canada	3/8	Case series	BS, CT, surgery	Surgery	—
Lowry et al[45]	2002	USA	1/1	Case report	Surgery	Surgery	—
Lyall et al[46]	1956	USA	1/1	Case report	Surgery	Surgery	I&D
Lucaya et al[47]	1990	USA	4/4	Case series	BS	Surgery	I&D

Table 1
(*continued*)

Author	Year	Geographic origin	Sample size,* n = 202/576	Study design	Diagnostic methods	Final therapy used	Previous treatment
Mahomed et al[48]	1998	Scotland	2/2	Case report	MRI, surgery	Surgery	—
Makino et al[49]	1986	Japan	5/9	Case series	BS, laryngoscopy	Surgery	I&D
Martin del Rey et al[50]	2002	Spain	2/2	Case report	BS	Surgery	I&D
Mizuno et al[51]	1998	Japan	1/1	Case report	BS, laryngoscopy	Surgery	—
Moreno Hurtado et al[52]	1996	Spain	1/9	Case series	Surgery	Surgery	—
Morrish et al[53]	1991	USA	1/1	Case report	Surgery	Surgery	—
Mortelmans et al[54]	2005	Belgium	1/1	Case report	MRI, CT, US	Surgery	—
Mouri et al[55]	1998	Japan	4/25	Chart review	CT, laryngoscopy, surgery	Surgery	—
Nonomura et al[56]	1993	Japan	3/4	Case series	BS, laryngoscopy	Surgery	I&D
Nusbaum et al[57]	1999	USA	1/12	Case series	CT	Surgery	—
Ostfeld et al[58]	1991	Israel	1/1	Case report	Surgery	Surgery	I&D
Orti et al[59]	2003	Spain	3/3	Case series	BS	Surgery	I&D
Ozaki et al[60]	1990	Japan	1/1	Case report	US	Surgery	I&D
Pantaleoni et al[61]	1992	Italy	1/1	Case report	Laryngoscopy	Antibiotics	—
Park et al[62]	2000	Korea	17/17	Case series	BS, CT, MRI, laryngoscopy	EC (15), antibiotics (2)	Antibiotics
Park et al[63]	1993	Korea	4/4	Case series	BS, CT, surgery	Surgery	I&D
Patra et al[64]	2002	India	1/1	Case report	BS, laryngoscopy	Surgery	I&D
Pereira et al[65]	2003	USA	1/5	Case series	MRI, surgery	Surgery	—
Perez Candela et al[66]	2000	Spain	3/3	Case series	CT, MRI	Surgery	—
Rea et al[67]	2004	UK	5/8	Case series	BS, laryngoscopy, surgery	Surgery	I&D
Regas Bech de Careda et al[68]	1992	Spain	3/3	Case series	BS, laryngoscopy	Surgery	I&D
Reiter et al[69]	1982	USA	1/1	Case report	Surgery	Surgery	I&D
Rochibaud et al[70]	2000	Canada	1/1	Case report	BS	Surgery	—
Sanusi et al[71]	1982	USA	1/1	Case report	Surgery	Surgery	—
Schloss et al[72]	1986	Canada	2/2	Case report	BS, laryngoscopy	Surgery	I&D
Skuza et al[73]	1991	USA	1/1	Case report	BS	Surgery	—
Singh et al[74]	1979	USA	1/1	Case report	Surgery	Surgery	—
Stone et al[75]	2000	USA	1/1	Case report	BS, CT, laryngoscopy	Surgery	I&D
Taylor et al[76]	1982	USA	1/2	Case report	CT	I&D	—
Tyler et al[77]	1992	USA	1/1	Case report	BS, CT, laryngoscopy	Surgery	—
Ueda et al[78]	1986	Japan	1/4	Case series	BS, laryngoscopy	Surgery	I&D
Vade et al[79]	1994	USA	1/1	Case report	MRI, laryngoscopy	Surgery	—
Vermeire et al[80]	1993	Belgium	1/1	Case report	BS, CT, laryngoscopy	EC	—
Wagner et al[81]	1988	USA	4/8	Case series	Surgery	Surgery	—
Wang et al[82]	2003	Taiwan	5/5	Case series	BS	Surgery	—
Yanai et al[83]	2004	Japan	1/1	Case report	Laryngoscopy	Surgery	—
Yamashita et al[84]	1994	Japan	1/1	Case report	BS	Surgery	—
Zarbo et al[85]	1983	USA	1/1	Case report	Surgery	Surgery	—

BS, barium swallow; *CT*, computed tomography; *MRI*, magnetic resonance imaging; *US*, ultrasound; *I&D*, incision and drainage; *EC*, endoscopic cauterization; *NR*, not reported.

*The numerator represents the number of cases meeting criteria for a third branchial arch anomaly (see text); the denominator represents the total number of cases presented in the report.