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Management of congenital fourth branchial arch anomalies: a review and analysis of published cases

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Abstract

Background/Purpose: Congenital fourth branchial arch anomalies are uncommon entities, heretofore described only in case reports, affecting primarily children, and typically presenting as a cervical inflammatory process. The aim of the study was to collect appropriate data on the diagnosis, treatment, and outcome of this condition and to suggest guidelines for its management.

Methods: We conducted a structured review of the literature for cases explicitly identified as congenital fourth branchial arch anomalies or meeting anatomical criteria for this condition. We computed descriptive statistics and performed several post hoc 2-way comparisons of subgroups of cases.

Results: We located and critically evaluated 526 cases. Fourth arch anomalies were usually located on the left (94%) and generally presented as acute suppurative thyroiditis (45%) or recurrent neck abscess (42%). Barium swallow and direct laryngoscopy were the most useful diagnostic tools. Treatment options differed mainly in recurrence rates: incision and drainage, 89%; open neck surgery and tract excision, 15%; endoscopic cauterization of the sinus tract opening, 15%; and open neck surgery with partial thyroidectomy, 8%. Complications after surgery occurred primarily in children 8 years or younger.

Conclusion: Fourth arch anomalies are more common than once thought. Treatment of these disorders with repeated incision and drainage yields high rates of recurrence; thus, complete excision of the entire fistula tract during a quiescent period appears preferable. Combining this surgery with partial thyroidectomy may further decrease recurrence rates. Complications can likely be minimized by using antibiotic treatment of acute infections or endoscopic cauterization in children 8 years or younger, and delaying open neck surgery.

The branchial arches give rise to specific derivatives, which for the fourth arch include the laryngeal cartilages, the laryngeal and pharyngeal constrictor muscles, the superior laryngeal nerve, the left thoracic aorta, the right proximal subclavian artery, the ultimobranchial body from which the

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calcitonin-secreting interfollicular cells of the thyroid arise, and the superior parathyroid glands [1]. Most congenital lateral cervical cysts, fistulae, and sinuses are considered to originate from the branchial apparatus. Branchial cysts have no external or visceral opening and thus retain secretions, whereas branchial sinuses communicate with either the skin or the pharyngeal lumen, and fistulas are tracts connecting the pharynx to the skin. The first published description of a branchial cleft lesion was in 1832 by Ascherson [2]. One hundred years later, Raven [3] reported a case of a pyriform sinus fistula, and Sandborn and Shafer [4] more recently described a branchial cyst of fourth pouch origin. Recurrent neck abscess and acute suppurative thyroiditis are described as the most common clinical presentations [5-7].

The reported prevalence of fourth arch anomalies is very low. According to most authors, the number of published cases is less than 100 [8], representing 1% to 4% of all branchial anomalies [9]. Therefore, consistent recommendations on diagnosis and management are lacking, and reports on recurrence and complication rates vary widely. To provide more comprehensive information on this condition, we undertook a review of the literature to identify all reported cases, summarize their clinical features, and identify the best options for diagnosis and treatment.

1. Method

1.1. Literature search and selection of cases

We searched PubMed, Medline, and Embase using the Scopus database with the following boolean combination: ("fourth" AND "branchial" AND "arch") OR ("pyriform" AND "sinus" AND "fistula") OR ("piriform" AND "sinus" AND "fistula"). We restricted the search to "human" studies published between 1968 and 2006. Of the 235 articles retrieved, 92 were excluded because they were reviews (4), dealt with fourth arch vascular malformations (6), described complications of laryngeal surgery for tumors (19), reported other branchial arch anomalies (17), or were embryologic/basic science studies (46). A total of 143 publications reporting 535 cases remained. From the references of the selected articles, we found an additional 34 publications describing another 55 cases. Unpublished reports were not considered.

We reviewed case descriptions in the 177 selected publications and included only those cases that were either (1) explicitly identified by the authors as fourth branchial arch anomalies or (2) characterized by a clearly described tract starting at the apex of the pyriform sinus and exiting through the cricothyroid membrane. Based on these criteria, we excluded 64 cases, leaving 526 for analysis. A list of the 177 publications, together with a number of qualifying cases presented in each publication, is available online as a supplement to this article.

1.2. Data extraction

Two authors (KN and RG) independently reviewed all cases and extracted the following variables: sex, side of presentation, age of onset, age of diagnosis, mode of presentation, diagnostic investigations, bacteriologic and histologic findings, thyroid function, treatment, complications, and recurrence rate. Discordant findings between the 2 reviewers were discussed and resolved to achieve agreement.

1.3. Data analysis

Data were scored as numeric or categorical variables. We first calculated summary statistics and then performed several post hoc 2-way comparisons of subgroups of cases. We compared these subgroups using t tests to compare means and χ^2 to compare proportions, with α set at .05, 2-tailed.

2. Results

2.1. Clinical features

Most fourth branchial arch anomalies were diagnosed in childhood (Table 1), although often many years after the onset of symptoms. The great majority of cases occurred on the left side, and most commonly presented either as acute suppurative thyroiditis or neck abscess (Table 1). Individuals presenting with acute suppurative thyroiditis were significantly older at time of onset and time of diagnosis than other cases (Table 2). Of the 518 cases in which the patient's age was reported, 45 occurred in neonates, and 29 (64%) of these presented with respiratory distress. Of the 47 case patients

Table 1 Features of 526 cases of branchial arch anomalies

Male (n [%])	258 (49%)
Age of onset (mean [range])	9 y (0-68 y)
Age of diagnosis (mean [range])	14 y (0-79 y)
Time from onset of symptoms to diagnosis (mean [range])	5 y (0-69 y)
Site of presentation (521 cases) (n [%])	
Left	487 (93.5%)
Right	32 (6%)
Bilateral	2 (0.5%)
Initial presentation (n [%])	
Acute suppurrative thyroiditis	240 (46%)
Neck abscess	223 (42%)
Neck mass	26 (5%)
Neck mass with respiratory distress	14 (3%)
Cutaneous discharging fistula	13 (2%)
Cutaneous discharging fistula with neck infection	10 (2%)

Table 2 Comparison of patients presenting with acute suppurative thyroiditis vs other clinical presentations

Clinical presentation	AST	Others	*P*
Total cases (n [%])	240 (46%)	286 (54%)	
Male (n [%])	108 (45%)	149 (52%)	NS
Age of onset (mean years [SD])	12.2 (1.0)	7.5 (0.7)	<.001
Age of diagnosis (mean years [SD])	15.3 (0.8)	12.7 (0.8)	<.001
Diagnostic investigations performed (n [%])			
Barium swallow	199 (83%)	191 (67%)	<.001
Laryngoscopy	76 (32%)	161 (56%)	<.001
CT scan	73 (30%)	101 (35%)	NS
Ultrasound	80 (33%)	52 (18%)	<.001
Thyroid scan	89 (37%)	40 (14%)	<.001
Fine-needle aspiration	36 (15%)	21 (7%)	.005
MRI	10 (4%)	17 (6%)	NS
Plain radiographs of the neck	53 (22%)	30 (10%)	<.001
Treatment (n [%])			
Incision and drainage	99 (41%)	149 (52%)	.05
No. of attempts (mean [SD])	2.5 (0.3)	3 (0.3)	NS
Failure after initial attempt	82 (83%)	139 (93%)	.03
Open neck surgery initial procedure	80 (52%)	103 (46%)	NS
Open neck surgery procedure after failure	74 (48%)	120 (54%)	NS
No. of procedures (mean [SD])	1.1 (0.1)	1.6 (0.1)	<.001
Failure after initial procedure	15 (16%)	43 (48%)	.008
Partial thyroid surgery	73 (41%)	65 (26%)	.001
Failure after procedure	5 (7%)	6 (9%)	NS
EC initial attempt	13 (5%)	22 (8%)	NS
EC after failure	18 (7%)	9 (3%)	NS
No. of attempts (mean [SD])	1	1.2 (0.1)	NS
Failure after initial attempt	2 (15%)	7 (32%)	NS
Complications after open neck surgery [a]	6 (3%)	14 (6%)	NS

AST indicates acute suppurative thyroiditis; EC, endoscopic cauterization; NS, not significant.

[a] See Table 4 for listing of complications.

age 1 to 3 years, 24 (51%) presented with acute suppurative thyroiditis and 21 (45%) with neck abscess. Among 126 case patients age 4 to 8 years, 69 (55%) presented with neck abscess and 49 (39%) with acute suppurative thyroiditis; and of the 300 case patients older than age 8, 165 (55%) presented with acute suppurative thyroiditis and 121 (40%) with neck abscess.

2.2. Findings

In 100 cases (19%), specimens were sent for bacteriologic cultures, which yielded a variety of organisms (Table 3). Penicillin was administered alone or in combination in most cases where the specific antibiotic treatment was reported (70%). Pure aerobes, such as streptococci or staphylococci, were typically treated with β-lactam antibiotics (penicillin G or V) or with β-lactamase–resistant antibiotics (oxacillin, dicloxacillin, meticillin, nafcillin, and piperacillin) or β-lactamase inhibitors (sulbatam and clavulanic acid). *Klebsiella*, *Escherichia coli*, *Pseudomonas aeruginosa*, or *Haemophilus* species were typically treated with second- or third-generation cephalosporins (cefoxitin , cefotaxim, or ceftriazone). *Eikonella corrodens* was treated with penicillin or cephalosporins. *Citrobacter* and *Proteus* were treated with penicillin in combination with an aminoglycoside (gentamicin). Mixed cultures were treated with penicillin in combination with clindamycin or metronidazole. Bacteroides species were treated with metronidazole. Culture cases with enterococci (formerly classed as group D streptococci) or with *Streptococcus viridans*, especially when growing alone, were typically treated with a combination of penicillin and an aminoglycoside (gentamicin or tobramycin) to avoid endocarditis.

Thyroid function tests were reported in 73 cases (14%); of these, 59 (81%) displayed normal thyroid function, 10 (14%) showed transient hyperthyroidism, and 4 (5%) showed hypothyroidism; one of the latter required temporary thyroxine treatment. Histopathologic examination was reported in 213 cases (40%); of these, 161 (76%) demonstrated a stratified squamous epithelium, 23 (11%) respiratory ciliated epithelium, 24 (11%) chronic inflammatory cells along the sinus tract, and 5 (2%) subacute inflammatory cells. Thyroid, thymic, and parathyroid tissues were reported in 24%, 19%, and 1% of the 213 cases, respectively. A cystic lesion was described in 8% of cases.

Table 3 Results of specimens sent for culture (100 cases)

Species	No. of cases
Pure aerobes	57
Streptococcus sp	33
Staphylococcus sp	10
Haemophilus sp	1
Klebsiella	5
E coli	5
P aeruginosa	1
Proteus	1
Citrobacter	1
Pure anaerobes	6
E corrodens	3
Bacteroides sp	2
Actinomyces odontolyticus	1
Mixed	25
No growth	12

2.3. Diagnostic investigations

Investigations used (positive predictive values [ppv's]) were barium swallow in 74% of cases (ppv, 88%), direct laryngoscopy in 45% (ppv, 90%), magnetic resonance imaging (MRI) in 5% (ppv, 63%), computed tomography (CT) in 33% (ppv, 46%), and thyroid scan in 25% (ppv, 2%). Thirty-four cases (7%) were diagnosed during the surgical procedure. Plain radiographs of the neck and fine-needle aspiration did not contribute to the diagnosis in any case. The choice of investigations varied depending on whether the patient presented with acute suppurative thyroiditis or another clinical picture (see Table 2). Several reports documented incidental discoveries of fourth branchial arch anomalies during laryngoscopy, laryngectomy, and MRI. Misdiagnoses of esophageal perforation [8], thyroid cancer [10], thyroid cyst, or ruptured diverticulum [11] were also documented.

2.4. Treatment and outcome

Initial treatments performed were incision with drainage in 248 (47%) of cases, open neck surgical procedures in 183 (35%), endoscopic procedures in 35 (7%), and conservative treatment in 53 (10%). Seven patients (1%) were waiting for treatment at the time of the report. Looking at individual age categories among the 518 patients where age was reported, and tabulating all treatments administered, rather than just initial treatments, we found that 14 (31%) of the 45 neonates received incision and drainage, 34 (76%) open neck surgery (with partial thyroidectomy in 6 [13%]), 2 (4%) endoscopic cauterization, and 3 (7%) antibiotic treatment. Of the 47 infants age 1 to 3, 21 (45%) received incision and drainage, 25 (53%) open neck surgery (with partial thyroidectomy in 9 [19%]), 4 (9%) endoscopic cauterization, and 2 (4%) antibiotic treatment; 7 (15%) were not operated on. Of the 126 children age 4 to 8, 60 (48%) received incision and drainage, 61 (48%) open neck surgery (with partial thyroidectomy in 26 [21%]), 12 (10%) endoscopic cauterization, and 7 (5%) antibiotic treatment; 20 (16%) were not operated on. Finally, among the 300 cases older than 8, 153 (51%) received incision and drainage, 119 (40%) open neck surgery (with partial thyroidectomy in 97 [32%]), 44 (15%) endoscopic cauterization, and 11 (3%) antibiotic treatment; 29 (10%) were not operated on.

Looking again at the overall group, the failure rate after initial incision with drainage was 221 (89%). Repeated attempts at incision and drainage were performed in 132 (60%) of initially failed cases; in 14 (6%), no further treatment was reported despite recurrences. Among individuals initially treated with incision and drainage, the mean number of incision and drainage attempts was 2.7 (range, 1-28). Only 2 cases (1%) were successfully treated exclusively by incision and drainage; almost all cases required another subsequent treatment (open neck surgery, with or without partial thyroidectomy, or endoscopic tract cauterization). Altogether, 377 patients received open neck surgery, either initially or after the failure of incision and drainage, and 62 received endoscopic procedures, either initially or after the failure of incision and drainage.

Of the 377 patients receiving open neck surgical procedures, 58 exhibited recurrence after the initial procedure—a failure rate of 15%. However, among the 138 patients who received surgery that included a partial thyroidectomy, only 11 (8%) showed recurrence. Among all patients initially treated with open neck surgery, the mean number of procedures per person was 1.3 (range, 1-11). Twenty (5%) of the 377 patients developed complications (summarized in Table 4). Most of these complications occurred in children who received surgery at age 8 or younger (see Table 4); indeed, of a total of 161 children who received surgery at age 8 or younger, 16 (10%) developed complications, as compared with only 4 (2%) of the 216 individuals older than age 8 who received surgery ($P < .001$). Among cases with complications, one involved removal of parathyroid tissue with the specimen, but the patient possessed parathyroid glands on the contralateral side, and hence did not develop hypoparathyroidism. Among the 62 patients receiving endoscopic procedures, the failure rate after the initial procedure was 9 (15%), and the mean number of procedures per person was 1.1 (range, 1-3). No complications were reported after this treatment option.

Among the 56 cases treated conservatively, 23 received antibiotics, 14 were observed, and 7 refused surgery. In 12 cases, no reasons were given for the lack of surgery. One case presented recurrent episodes of thyroiditis, corresponding to a failure rate of 2% among those treated conservatively.

3. Discussion

Anatomically, fourth branchial arch anomalies represent vestiges of a tract that originates as a sinus tract from the apex of the pyriform sinus (Fig. 1). It travels inferiorly in the tracheoesophageal groove, posterior to the thyroid gland, and into the thorax, where it loops below the aorta on the left and subclavian artery on the right. The descending part of this tract before the first loop is the most common location of clinical infection [12]. The tract then changes direction, coursing superiorly, passing posterior to the common carotid artery, and ascending in the neck to reach the hypoglossal nerve. It then makes a second loop around the hypoglossal nerve and finishes its course at the medial border of the sternocleidomastoid muscle [12].

Some authors group third and fourth arch fistulas generically as pyriform sinus tracts or pyriform fossa lesions [13-15]. However, these 2 types of anomalies are anatomically distinct: third branchial arch anomalies originate from the base (cranial end) of the pyriform sinus and pass above the superior laryngeal nerve. In

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

Study	Case no.	Sex	Side	Age at onset	Age at the time of treatment	Type of presentation	No. of open neck surgeries	Partial thyroid surgery	Complication
Andrieu-Guitrancourt (1979)	1	F	L	7 mo	1 y 6 mo	Neck abscess	3	No	Salivary fistula
Andrieu-Guitrancourt (1979)	4	F	L	6 y	6 y	Neck abscess	2	Yes	Infected wound
Ford (1992)	1	M	L	Birth	2 y	Respiratory distress	1	No	Vocal cord paralysis
Nonomura (1993)	1	F	L	5 y	7 y	Neck abscess	2	No	Arytenoids edema
Schneider (1995)	1	F	L	4 y	17 y	Neck mass	2	No	Salivary fistula
Nicollas (1998)	2	M	L	5 y	5 y	Neck abscess	1	Yes	Vocal cord paresis
Nicollas (1998)	5	F	L	5 y	5 y	Acute suppurative thyroiditis	1	Yes	Vocal cord paralysis
Minhas (2001)	1	F	L	32 y	32 y	Acute suppurative thyroiditis	1	Yes	Vocal cord paralysis
Schrime (2003)	1	F	L	Birth	Birth	Neck mass	1	No	Vocal cord paralysis
Roediger (1977)	1	Not reported	L	Birth	Birth	Neck abscess	1	No	Vocal cord paresis
Miyauchi (1981)	5	F	L	8 y	8 y	Acute suppurative thyroiditis	1	Yes	Vocal cord paralysis
Miyauchi (1981)	21	M	L	Not reported	45 y	Acute suppurative thyroiditis	1	Yes	Vocal cord paresis
Contencin (1990)	12	F	L	7 y	7 y	Neck abscess	3	No	Vocal cord paresis
Contencin (1990)	13	F	L	6 y	6 y	Acute suppurative thyroiditis	3	No	Vocal cord paralysis
Narcy (1988)	2	M	L	Birth	Birth	Respiratory distress	1	No	Salivary fistula[a]
Narcy (1988)	3	F	L	1 mo	1 mo	Respiratory distress	1	No	Vocal cord paresis
Narcy (1988)	4	F	L	6 y	6 y	Neck abscess	3	Yes	Vocal cord paralysis
Hashizume (1993)	5	F	L	1 y	6 y	Neck abscess	4	No	Salivary fistula
Garrel (2006)	4	F	R	13 y	15 y	Acute suppurative thyroiditis	1	Yes	Vocal cord paresis
Garrel (2006)	7	F	L	Birth	6 mo	Neck abscess	1	No	Vocal cord paresis

[a] A cyst was removed, and parathyroid gland tissue was found in t he external wall, but hypoparathyroidism did not occur presumably because the contralateral parathyroid glands were preserved.

contrast, 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 [12]. In our review process, in instances where the author had not explicitly diagnosed a fourth branchial arch anomaly, we used these criteria to identify cases of fourth arch origin. It should be noted that thymic tissue is not specific for a third pouch derivative, as accessory thymic tissue derived from the fourth branchial pouch has been described [16]. Parathyroid tissue was not used as a selection criterion because of its rarity (3 cases) and its occasional presence in third branchial arch anomalies.

Our review yielded 526 reported cases of fourth branchial arch anomalies meeting the above criteria—a much larger number than most authors have previously suspected [8]. Many of these cases appear in individual reports or small case series, varying in their degree of quantitative detail. Thus, the data do not permit detailed statistical analyses. One must also allow for variations in the quality of the reports; in some cases, for example, instances of recurrence might have been missed because of inadequate follow-up. In addition, any interpretation must consider the possibility of publication bias; cases with particular clinical attributes may have been disproportionately represented in the literature. Despite these limitations, however, several impressions seem clear.

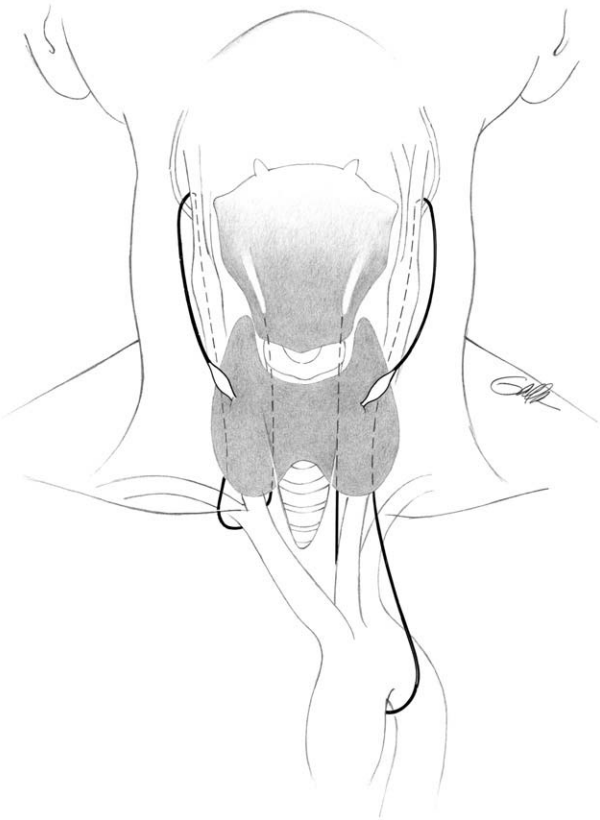

Fig. 1 Theoretical pathway of a fourth branchial arch fistula.

The clinical presentation of fourth branchial arch anomalies varies with age. In the neonate, the presentation is usually one of respiratory distress; by contrast, cervical cutaneous fistulas develop only in late childhood. Cases of mediastinal abscess of fourth branchial arch origin have been reported and must be considered [17,18]. Acute suppurative thyroiditis is a late clinical presentation, suggesting that the sinus tract may progress with age, beginning close to the pyriform sinus at a younger age and migrating lower in the neck in older children. In a child presenting with suppurative thyroiditis, various features may suggest a pyriform sinus fistula; these include a previous upper respiratory tract infection, fever, painful thyroid swelling, elevated erythrocyte sedimentation rate, a minor variation of thyroid hormone levels, and left side predominance [19]. Attempts at visual identification of the sinus tract may sometimes fail because the tract may be difficult to see, especially in the acute phase [20,21].

The frequent delay in diagnosis of fourth arch anomalies demonstrates not only the benign nature of this condition but also the difficulty in diagnosing it accurately. The left side predominance might be because of the more complex and extended pathway of the fourth branchial tract on the left side (Fig. 1) [9]. It might also be because of the absence or involution of the ultimobranchial body on the right side [22]. No associations with anomalies of the aortic arch or the right subclavian artery were found in the cases reviewed.

Most of the organisms cultured are oral flora susceptible to penicillin or related β-lactamase–resistant antibiotics as noted above. Treatment of other bacteria is best guided by sensitivity findings on culture. Other organisms, such as fungi, parasites, and mycobacteria, occur less frequently. Reports of several cases of acute suppurative thyroiditis because of *Eikenella corrodens* in pure or mixed culture are probably explained by the adaptation of this organism to thyroid tissue and its reduced susceptibility to empirical antibiotic treatment [19,23-25].

Turning to the issue of diagnostic studies, the barium esophagogram may fail to demonstrate a sinus tract during the acute inflammatory phase, and therefore the study should be repeated 6 to 8 weeks later [9]. In contrast, direct laryngoscopy can be performed during an acute episode and can achieve several goals: (1) identification of a possible pyriform sinus tract opening; (2) identification of the sinus tract during neck exploration by insertion of various probes, catheters, contrast fluids, or by transillumination of the tract; and (3) definitive treatment with cauterization of the opening of the sinus tract. Therefore, some authors recommend direct laryngoscopy before excision of any suspected fourth branchial arch anomaly [26]. Magnetic resonance imaging is especially useful for detecting cystic anomalies of the sinus tract. The use of Vasalva or trumpet maneuvers during CT scan or barium swallow may help to open the sinus tract and make it more readily visible [27,28].

A range of treatment options can be considered. Incision and drainage are frequently performed, but the failure rate is high. By contrast, open neck surgery, especially when combined with partial thyroidectomy, yields far lower recurrence rates. However, it appears that open neck surgery may yield higher rates of complications in children 8 years or younger (see Table 4). Endoscopic treatment, reported in 62 cases [8,14,29-33], represents a minimally invasive technique using cauterization to obliterate the internal opening of a pyriform sinus tract during a quiescent period [33]. Possible advantages of this technique over open neck surgery include a lower complication rate and lower cost as an outpatient procedure. We found similar rates of recurrence after open neck surgery and endoscopic cauterization.

Overall, the above findings would suggest a tentative treatment algorithm for individuals presenting with acute suppurative thyroiditis or recurrent neck abscess attributable to fourth branchial arch anomalies (Fig. 2). To begin with, it should be recognized that there is a substantial risk of recurrent nerve injury, and this is in large measure because of ongoing local inflammation of the tissues surrounding the infected fistula tract. Therefore, when the presentation is an abscess, it seems prudent to delay treatment until this inflammatory process is maximally resolved by adequate and prolonged incision and drainage, plus antibiotics. Thereafter, up to about the age of 8, it seems preferable to emphasize medical treatment and delay surgical neck exploration. After age 8, complete excision of the entire fistula tract with a partial thyroidectomy (excision of a small portion of the

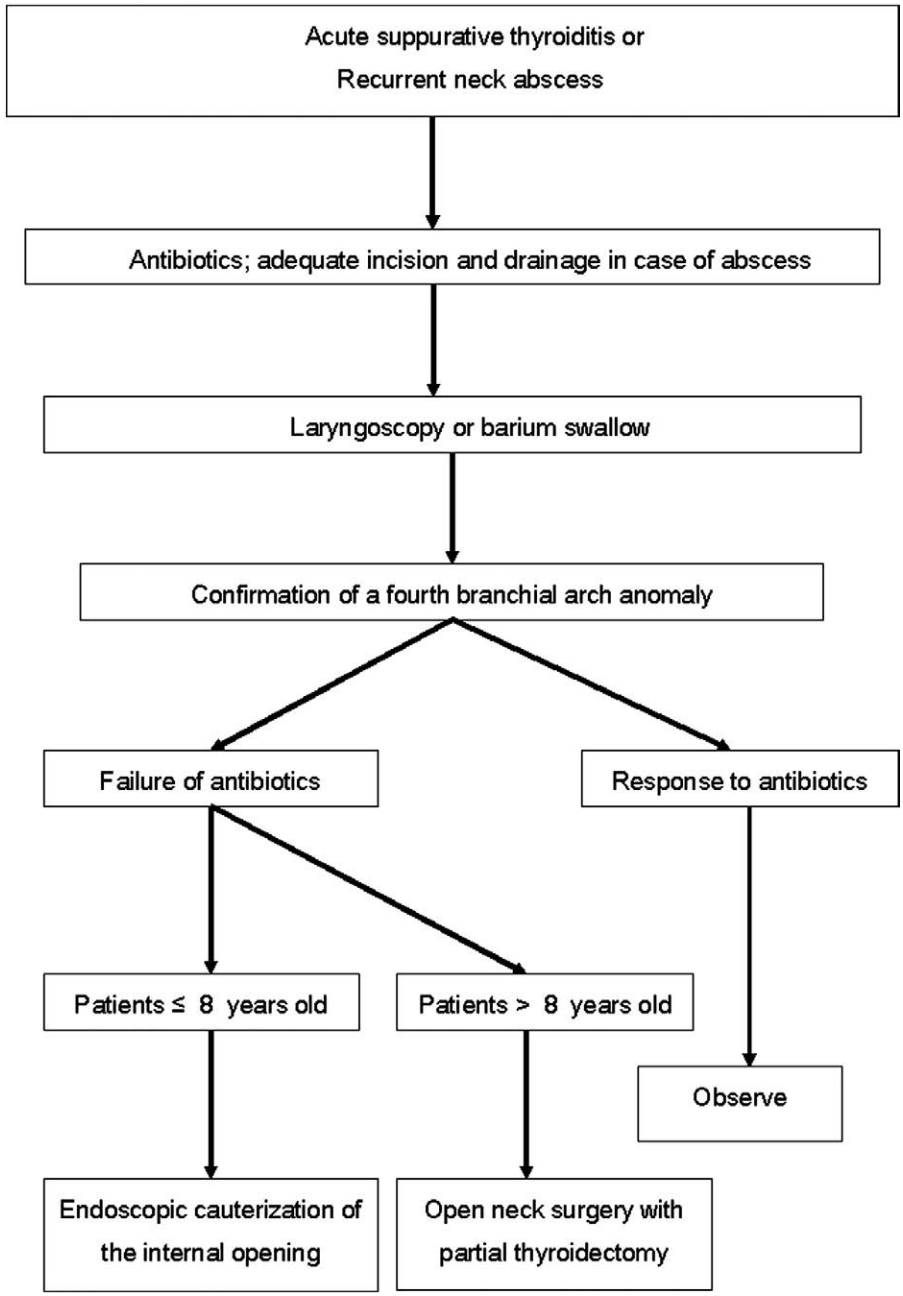

Fig. 2 Suggested algorithm for the management of congenital fourth branchial arch anomalies.

upper pole of the thyroid gland) during a quiescent period appears to be the treatment of choice. The resection starts at the distal portion of the fistula, which typically reaches the abscess in the vicinity of the thyroid gland by 1 of 3 courses: (1) lateral to the left lobe, (2) medial to it, or (3) through the lobe [34,35]. Identification of the recurrent laryngeal nerve and removal of the posterior portion of the thyroid cartilage during the surgical dissection are sometimes necessary [36-39]. A complete excision is essential, as retention of any squamous or columnar epithelium may lead to recurrence [40]. A conservative approach without surgery is possible in selected cases, but the apparently low rates of recurrences in such cases may be partially attributable to publication bias. Therefore, in cases of recurrence after conservative treatment, we recommend surgery.

In summary, our review suggests that fourth branchial arch anomalies are more common than previously suspected. These anomalies typically present as recurrent neck abscess, acute suppurative thyroiditis, or, in the neonatal period, respiratory distress. The barium swallow and direct laryngoscopy are the diagnostic tools with the highest ppv. Complete excision of the entire fistula tract during a quiescent period, together with a partial thyroidectomy, appears to be the treatment of choice. To minimize complications, however, we suggest that surgery be reserved for patients older than age 8; in children 8 years or younger, we recommend

medical treatment. Repeated incision and drainage or incomplete surgery must be avoided because of the increased risk of recurrence. Endoscopic cauterization can be an effective alternative strategy.

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