

Fourth branchial arch fistula and suppurative thyroiditis: a life-threatening infection

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Abstract

A potentially life-threatening case of recurrent left-sided thyroid abscess formation secondary to a fourth branchial arch sinus fistula is presented. The patient developed a reversible left vocal fold palsy during an acute episode of suppurative thyroiditis requiring a temporary tracheostomy due to a compromised airway. Investigations commonly used to demonstrate this anomaly may fail to confirm the diagnosis as in the case presented and exploratory surgery with excision of the fistulous tract should still be considered. We describe a method of repairing the pharyngeal opening to reduce the risk of recurrence or pharyngeal leak.

Key words: Abscess; Branchial Arches; Thyroiditis; Suppurative; Vocal Cord Paralysis

Case report

A 32-year-old lady presented in 1990 with a 10-day history of sore throat and a tender left thyroid swelling. On examination she had a hard diffuse goitre and fine needle aspiration cytology (FNAC) was performed. Subacute thyroiditis was suspected and the patient commenced on aspirin for symptomatic relief. At this time thyroid function tests were normal, ESR was 71 mm/Hr, thyroglobulin autoantibodies (1:80 000) were positive. At review one week later she was clinically much better and her goitre smaller. At this time she was clinically euthyroid and biochemically hyperthyroid (Free T₄ 26.9 [9–24 pmol/l], Free T₃ 10.5 [2.5–8.5 pmol/l], TSH <0.1 [0.4–4.5 mu/l]). The diagnosis of subacute thyroiditis was confirmed and supported by the findings from FNAC which showed necrotic debris, polymorphs, lymphocytes and degenerative cells.

Two weeks later she presented again to the endocrinology team with a swollen painful neck and a several day history of purulent discharge from sites in the anterior neck. This had failed to respond to oral erythromycin. A diagnosis of suppurative thyroiditis with a possible underlying neoplasm was made and a referral made to the general surgical team. Thyroid function tests at this time had improved (FT₄ 16.2 pmol/l, FT₃ 5.4 pmol/l, TSH 0.11 mu/l). The patient was commenced on i.v. cefuroxime and underwent emergency incision and drainage of the anterior neck space abscess. The abscess was noted to be lying superficial to the strap muscles and the thyroid was mildly indurated. Subsequent pus culture grew *Streptococcus milleri*. Histology of curettings from the abscess cavity showed acutely inflamed granulation tissue on a background of chronic inflammation. No thyroid epithelium or recognizable colloid was found. It was thought at this time the abscess was due to bacterial contamination at the time

of the FNAC. The patient made a good recovery and after several follow-up visits was discharged six months later.

In 1994 the patient developed post-partum hypothyroidism requiring thyroxine replacement therapy but otherwise remained well until 1998. Eight years on she developed a sore throat which failed to respond to amoxicillin or ciprofloxacin by her general practitioner. On admission to the ENT department she had a marked pyrexia, an enlarging painful thyroid and associated dysphagia. There was no response to i.v. benzylpenicillin and she complained of increasing pain and dysphagia. A flexible nasendoscopy showed oedema of the left aryepiglottic fold and also a left vocal fold palsy. A neck ultrasound scan (USS) showed a left thyroid abscess. The patient underwent an urgent incision and drainage procedure and laryngoscopy. Thirty to 40 ml of pus was drained and a possible communication posteriorly into the hypopharynx was suspected. At direct laryngoscopy, there was considerable oedema of the arytenoids and epiglottis. Pus was noted in the pharynx.

The presence of significant laryngeal oedema, coupled with a paralysed vocal fold, indicated the need for a tracheostomy which was performed after dividing the thyroid isthmus. A nasogastric tube was inserted for feeding purposes. The patient made an excellent recovery on i.v. Augmentin. A contrast swallow showed no obvious fistulous tract. A repeat flexible nasendoscopy showed a healthy larynx and recovery of the vocal fold palsy. The patient was successfully decannulated and discharged home on oral antibiotics (Augmentin). Post-operatively the possibility of a third/fourth branchial arch fistula was considered the likely route of infection resulting in repeated left anterior neck space infection. A repeat barium swallow three months later was normal and failed to demonstrate a fistula. Whilst awaiting a computed tomography (CT) scan, the patient presented yet again as an acute emergency with a sore throat, dysphagia and an

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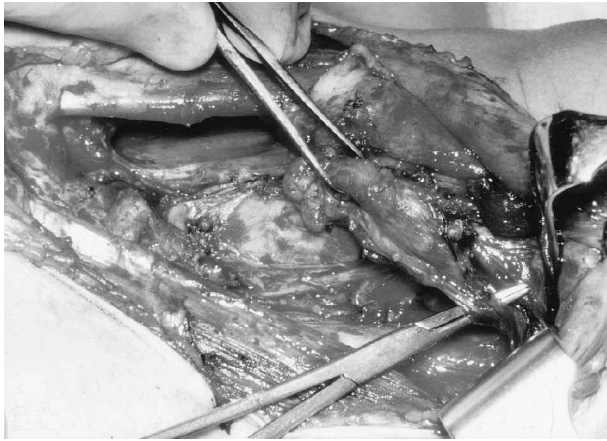


FIG. 1

Fistulous tract identified at left upper thyroid pole.

increasingly tender left-sided neck swelling. This time, she failed to respond to oral antibiotics (azithromycin, cefuroxime). Flexible nasendoscopy showed only mild laryngitis and the patient was admitted for i.v. cefuroxime. An USS showed a homogeneous thyroid with no nodules, an ill-defined $1.5 \times 2 \times 2$ cm low echo area collection consistent with an abscess in the left anterior triangle superior to the thyroid. Three ml of pus was aspirated under USS guidance which subsequently grew *Streptococcus viridans* and alpha haemolytic streptococcus. The patient then made a good recovery and was discharged home. The postponed CT scan with i.v. contrast showed abnormality of the soft tissues of the left side of the neck below the level of the thyroid cartilage but no focal collection or fistula was demonstrable.

At subsequent exploration, direct laryngopharyngoscopy appeared normal and despite a meticulous search of the left pyriform fossa, no fistulous opening was visible. A left thyroid lobectomy was performed and a fistulous tract was identified at the upper thyroid pole (Figure 1). This was traced superiorly above the cricopharyngeus and found to end in the left pyriform fossa. The left thyroid lobe and fistula were excised completely. The opening in the pharynx was closed and the repair covered with a transposed right sternohyoid muscle pedicled flap (Figure 2). The patient made an uneventful recovery and has had no problems to date.



FIG. 2

Pharyngeal repair covered with a transposed right sternohyoid muscle pedicled flap.

Discussion

The resistance of the thyroid gland to infection is well recognized.¹ The differential diagnosis of a painful inflamed anterior neck space mass includes acute, subacute or chronic thyroiditis, a thyroid abscess, infected thyroglossal cyst, thyrotoxicosis, neoplasm, bleeding into a thyroid cyst, cellulitis and cervical lymphadenitis.² Routine initial investigations of any suspected thyroid pathology should include thyroid function tests, thyroid antibodies and FNAC. If an inflammatory condition is likely, a full blood count, ESR and/or C-reactive protein are usually requested. Suppuration following fine needle aspiration is recognized but uncommon.^{3,4}

This case demonstrates that suppurative thyroiditis may mimic subacute thyroiditis especially in the early phase. Patients may initially show transient hyperthyroidism.^{5,6} With abscess formation patients become unwell and toxic with increasing pain and swelling of the thyroid that is not relieved by simple analgesia. Features of dysphagia, odynophagia, hoarseness, choking sensation and airway compromise may occur if there is inflammation of the laryngopharynx.

A branchial arch fistula is now recognized as a possible cause for recurrent left anterior neck space infection with, or without, abscess formation. Whether this arises from the third or fourth branchial apparatus is debatable but the most likely is a fourth branchial arch fistula.⁷ The fourth arch fistula is described as running from the apex of the left pyriform fossa to open in the skin anterior to the sternocleidomastoid passing close to the upper pole of the left thyroid lobe.^{8,9} These abnormalities seem to be exclusively left-sided although no definite reason for this can be provided.^{7,10-12} The fistula is thought to act as a conduit for infecting organisms from the pharynx and a preceding upper respiratory tract infection is often elicited as in this case. This would explain the repeated episodes of suppurative thyroiditis in these congenital anomalies given that the thyroid gland otherwise is fairly resilient to bacterial insult. Abscess formation may occur in relation to the left thyroid lobe,^{12,13} superficial tissues of the anterior neck,¹¹ retropharyngeal space¹⁴ and has been reported tracking into the thorax.¹⁵ We suspect that reports in the early literature of recurrent left anterior neck space infection requiring repeated incision and drainage would most likely be attributable to fourth arch fistula. Alternative presentations of a fourth arch abnormality include lateral neck fistula, a cyst, mass or oesophageal stricture.¹⁶

A congenital anomaly was not suspected in this case until after a second potentially life-threatening episode of suppuration despite the finding of pus in the pharynx and left-sided laryngeal inflammation. The patient required incision and drainage with a covering tracheostomy due to a compromised airway.

A barium swallow may demonstrate a fistula arising from the apex of the left pyriform fossa in the absence of acute infection, or a direct laryngoscopy may demonstrate the fistula opening.^{11-13,17} More recently a CT scan with a forced aeration manoeuvre has been reported to demonstrate the fistula as an air-filled sinus contrasting with the surrounding soft tissues of the neck.¹⁸ However, these investigations are not infallible and may fail to show any anomaly especially after repeated severe infections. This is especially true if there is fibrosis and inflammation at the fistula opening in the pyriform fossa as in this case. Given a strong history of recurrent neck space infection, a fistula should be suspected even if the above investigations are normal. Excision of the complete tract is recommended.^{11,14,17,19,20} The use of a muscle interposition flap has not been previously described. We feel this adds

negligible time and morbidity to the operation whilst rendering the likelihood of recurrent fistula formation extremely remote.

In conclusion, this case demonstrates that the presence of recurrent left-sided anterior neck space infection should raise the possibility of a fourth arch fistula²¹ despite negative radiological investigation or endoscopy. Recurrence is common if not adequately treated by total excision of the tract once the acute inflammatory episode has subsided.

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