

## Parapharyngeal (retrostyloid)—third branchial cleft cyst

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### Abstract

This is the first description of successful surgical excision of a parapharyngeal (retrostyloid compartment) cyst remnant of the third branchial arch.

### Introduction

An extremely rare parapharyngeal-retrostyloid compartment presentation of a deep upper neck abscess, proved on surgery to originate from an infected branchial cyst is presented.

### Case report

A 22-year-old male was admitted to the Otolaryngology Department with fever, sore throat and dysphagia of one week's duration. He had been hospitalized four months earlier with the same complaints at another hospital, where he had undergone repeated drainage of a left parapharyngeal space abscess. During the ensuing months recurrent bouts of fever and dysphagia temporarily improved following courses of amoxicillin.

On admission, the patient was febrile (39.7°C) and complained of tenderness in the left retromandibular area. Inward bulging of the left retrotonsillar pharyngeal wall was observed. The blood count showed moderate leukocytosis. From the

mucopurulent exudate obtained by transoral needle aspiration, *Streptococcus viridans* was cultured. The contrast enhanced Computed Tomographic (CT) scans of the neck were suggestive of a parapharyngeal space abscess (Fig. 1). The surgical exploration of the left side of the neck through a standard cervical approach and excision of the submandibular gland revealed no pathological findings in the anterior compartment of the parapharyngeal space. Deep to the great vessels a cystic formation was palpated. Following blunt dissection of the anterior aspect of the cystic formation from the internal carotid artery and to a lesser extent from the internal jugular vein, their lateral retraction and caudad dissection, the inferior pole of the cyst was found adjacent to the tonsillar region without communication with the pharynx. The cyst was lying on the prevertebral fascia (Fig. 2). The dissection of the cephalad aspect was hampered by the narrow operating field and the presence of the styloid process, but facilitated to some extent by the good plane of the cleavage. Fracture of the styloid process was useful and facilitated the dissection of the cephalad pole of the cyst adja-

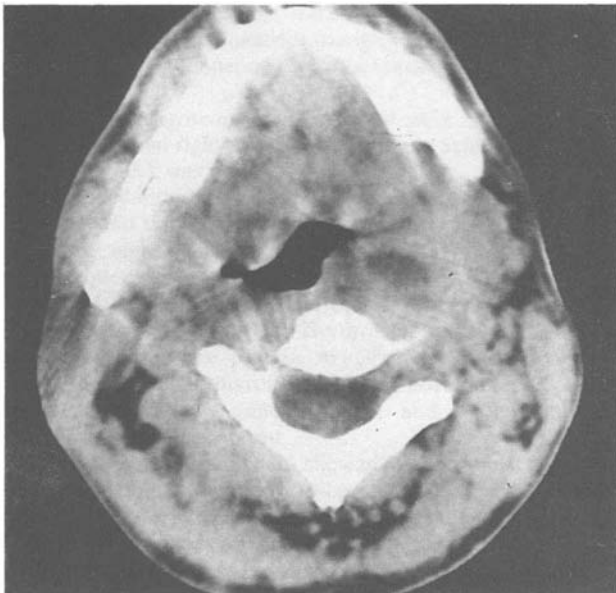


FIG. 1

Axial CT scan with contrast enhancement reveals a hypodense core of a poorly delimited formation in the left parapharyngeal space.

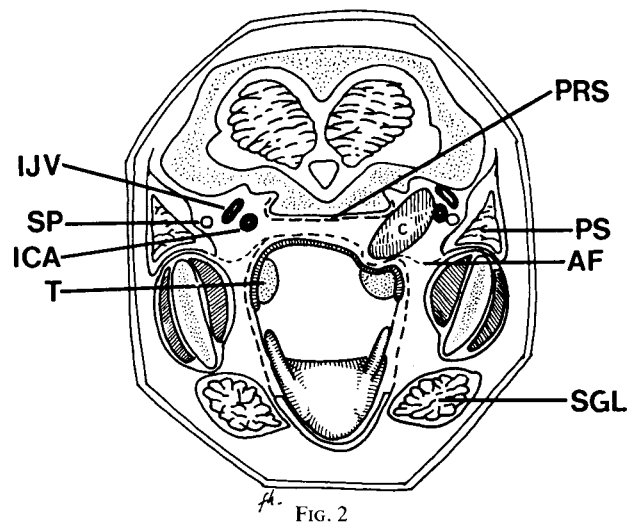


FIG. 2

The anatomic relationship of the parapharyngeal space—retrostyloid compartment 'branchial type' cyst. Oblique section through the neck at the mandibular angle. (Modified after Langenbrunner and Dajani, 1971). AF—Alar Fascia; C—Cyst; ICA—Internal carotid artery; IJV—Internal jugular vein; PRS—Prevertebral space; PS—Parotid space—SGL—Submandibular gland; SP—Styloid process; T—Tonsil.

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FIG. 3

Low power view of part of the large cyst lined by thickened squamous epithelium. The surrounding wall of the cyst is formed by dense fibrous inflammatory granulation tissue infiltrated by lymphocytes.  $\times 65$

cent to the base of the skull. A mini silver clip was applied to the superior pole and the cyst (7/2.5 cm) was excised. The glossopharyngeal and hypoglossal nerves were not identified but seem to have been located dorsal to the cyst. Hemovac drainage was provided. The post-operative course was complicated by dysphagia and hoarseness due to palsy of the IXth, Xth and XIIth cranial nerves. The operative wound healed primarily, resulting in an aesthetic scar. The neurological palsies and deficits subsided gradually during the next eight months; first the dysphagia (three weeks), then the hoarseness (six months) and last, the velopalatal palsy (eight months). Post-operative CT scans and MRI showed no residual pathology in the parapharyngeal space.

Histological examination of the excised specimen showed a thick-walled cyst lined by stratified squamous epithelium and subepithelial lymphoid tissue (Figs. 3–5).

### Discussion

Infections of the parapharyngeal space, known as 'deep neck infections', generally resolve with high doses of systemically administered potent antibiotic treatment (Shoss *et al.*, 1985; Batsakis *et al.*, 1989) while the non-responsive patients may require surgical drainage.

The clinical picture was typical of retrostyloid space infection: bulging of the retrotonsillar posterior pharyngeal wall, without trismus and cervical swelling. The patient's condition did not improve following antimicrobial therapy and repeated transoral drainage. Enhanced CT scans, considered the most

useful diagnostic tool of the parapharyngeal space lesions (Shoss *et al.*, 1985), during both an earlier and current hospitalization failed to demonstrate the cystic structure of the specimen eventually excised.

Som *et al.* (1984) found that enhanced CT scans are highly efficient in the diagnosis of cysts located in the parapharyngeal space. The CT failure to demonstrate clearly a cystic structure results from the massive pericystic lymphocytic infiltration triggered by a superimposed upper respiratory infection (Verbin and Barnes, 1985), impairing the quality of resolution and consequently preventing the cyst identification. An improved soft tissue resolution for such lesions was recently reported by use of magnetic resonance imaging (Matt and Lusk, 1987).

The teratogenesis supported by the cyst histologic structure indicates a branchial apparatus anomaly (Batsakis, 1979; Ostfeld *et al.*, 1985). The cervical duct resulting from obliteration of the cervical sinus of His is lined by ectoderm. This sinus mesoderm is the origin of the internal carotid artery—the third branchial arch artery (Donegan, 1986). The cyst topolocation, dorsal to the internal carotid artery (the third branchial arch artery), strongly supports a third branchial arch origin (ReMine, 1963; Gage *et al.*, 1976; Batsakis, 1979; Davies, 1980; Donegan, 1986). The anatomical space dorsal to the internal carotid artery is the retrostyloid compartment, thus, a third branchial arch anomaly would be located in this space.

The theory of branchial cleft origin has been challenged (Verbin and Barnes, 1985). Most recently Wild *et al.* (1987, 1988) found that biochemically (Keratin phenotype analysis) the branchial cysts inner lining is similar to upper digestive tract squamous epithelia and concluded that those cysts would be an acquired condition, most likely resulting from Waldeyer's ring crypt epithelium cells settled in lymph nodes to form finally a lymphatic-epithelial combination. This alternative theory is applicable also to our case, *e.g.* the movement of tonsillar epithelium to a retrostyloid compartment lymph node, resulting in an epithelial-lymphoid complex: the cyst.

Branchial cysts located in the parapharyngeal space are very rare, ranging from 0.7 per cent (two patients) (Barnes and Grupp, 1985), 2.1 per cent (ten patients) (Batsakis and Sneige, 1989), to three per cent (one patient) (Som *et al.*, 1981), among lesions of this space. The clinical presentation of a branchial cyst as an abscess of the retrostyloid compartment is exceptional. A review of the literature turned up only two similar cases. The first, dating back to as early as 1904 (Coakley) diagnosed as an abscess and drained transorally; the biopsy specimen taken from the abscess cavity wall indicated an epithelial lining suggesting an infected branchial cyst. The second, reported as a retrotonsillar infected cyst, dorsal to the carotid artery (Bass, 1982) could have been also a retrostyloid compartment third branchial arch anomaly.

The external cervical approach allows fair surgical exposure

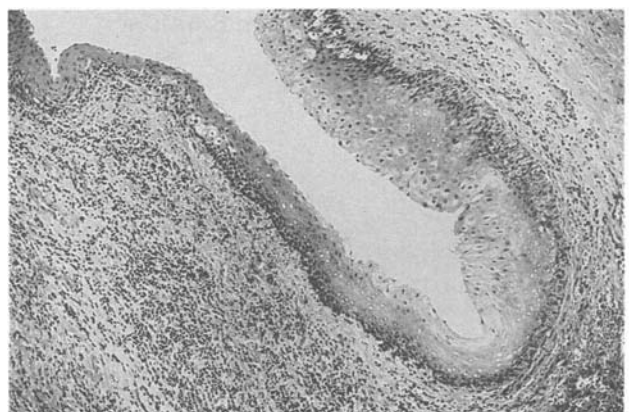
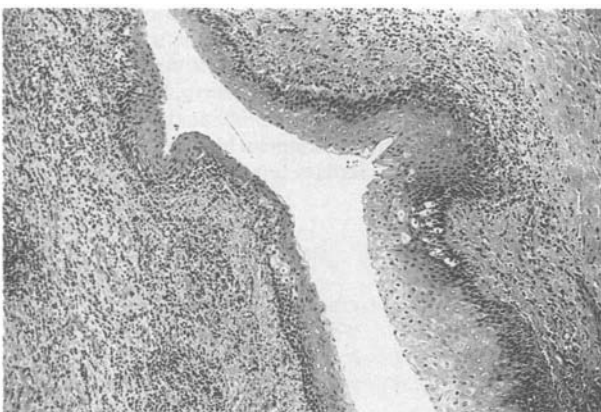


Fig. 4a,b

To show the epithelial lining of the cyst. The parakeratotic squamous epithelium covers the fibrous wall of the cyst densely infiltrated by lymphocytes.  $\times 160$ .





FIG. 5

A fairly large part of the cyst was ulcerated and covered by purulent granulation tissue. This has filled the lumen of the cyst.  $\times 65$ .

for the excision of benign lesions of the parapharyngeal-retrostyloid compartment, control of great vessels and neural structures adding to the operation's safety (Work, 1977; Shoss, 1985) and minimizing the risk of infection in comparison with the transoral or combined transoral-external approach (Bass, 1982) preferred by some surgeons (Goodwin and Chandler, 1988; Allison, 1989). The post-operative scar seems to be cosmetically superior to the appearance of the classic Mosher-type incision (Langenbrunner and Dajani, 1971).

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