Case Report

Hypoglossal Nerve Palsy in the Presence of a Branchial Cyst

David J. Black, Andrew W. McCombe, and Nnaemeka C. Okpala

Frimley Park Hospital, Portsmouth Rd, Frimley, Surrey, GU16 7UJ, UK
Address correspondence to David J. Black, davidblack@doctors.org.uk

Received 27 March 2013; Accepted 6 May 2013

Copyright © 2013 David J. Black et al. This is an open access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

Abstract We present an unusual case of a 20-year-old woman with a left-sided infected branchial cyst causing an ipsilateral 12th nerve palsy. This resolved upon excision of the cyst. Branchial cysts, infected or otherwise, should be high in the differential of a lateral cystic neck mass. This and other cases discussed demonstrate that the presence of cranial nerve palsies does not necessarily indicate a neoplastic cause.

Keywords branchial cyst; branchial cleft; hypoglossal nerve palsy

1. Introduction

Branchial cysts are congenital abnormalities resulting from the incomplete maturation of the branchial apparatus. They commonly present in the second and third decade, usually anterior to the sternocleidomastoid muscle. Branchial cysts are generally asymptomatic unless infected. Here we present an unusual case of an infected branchial cyst presenting with an ipsilateral hypoglossal nerve palsy.

2. Case report

A 20-year-old woman was referred to the Otolaryngology outpatient department with a rapidly increasing left upper neck swelling of 5 weeks duration. This was associated with left otalgia and reduced left tongue movements. She was otherwise well, with no significant past medical history other than a previous episode of glandular fever. She was a non-smoker and had only mild alcohol consumption.

Clinical examination found a large, firm non-pulsatile left level 2 neck mass. It was 7 cm in diameter, mildly tender, and compressible. The tongue showed left-sided fasciculation, reduced movement, and slight wasting. There was deviation to the left on tongue protrusion. All other cranial nerves were intact. Further clinical examination was unremarkable apart from a slightly prominent left tongue base, which did not look suspicious. Clinically, the lump appeared to be a branchial cyst (Figure 1).

Blood investigations were normal except for an ESR of 40. Monospot test was negative. An FNA of the lump, in clinic, produced 4 mL of serosanguinous fluid. This was reported as an inflamed squamous lined cyst, possibly branchial cyst in origin. An MRI showed a large cystic mass

3.9 cm × 3.1 cm × 3.3 cm in size, displacing the parotid superiorly and the submandibular gland anteriorly. There was enhancement around the cyst indicating active infection (Figure 2).

Figure 1: Pre-operative images demonstrating a left-sided branchial cyst and an ipsilateral hypoglossal nerve palsy.

Figure 2: An axial T2 weighted MRI image demonstrating a left-sided branchial cyst.

The cyst was excised intact under GA. The hypoglossal nerve was identified and preserved. Histology confirmed an infected branchial cyst. The patient was reviewed 2 months later in clinic, and tongue movements had returned to normal.

3. Discussion

Branchial cysts most commonly arise in the lateral part of the neck, along the anterior border of the upper third of sternocleidomastoid. Branchial cleft abnormalities may also occur anywhere along a tract extending from the border of sternocleidomastoid, passing between the internal and external carotid arteries and into the oropharyngeal tonsillar fossa. They most commonly present in the second to fourth decades, although their origins are thought to be embryological. During the fifth week of development, the second branchial arch grows over the third and fourth branchial clefts. This process forms the elongated cervical sinus of His. Normally, this is obliterated shortly after formation; however, incomplete closure leads to the formation of branchial cysts and fistulae [4,7]. Branchial cysts generally develop quickly, often preceded by an upper respiratory tract infection. Treatment is by surgical excision. Intra-operative identification of the internal and external carotids, vagus, hypoglossal, glossopharyngeal, and superior laryngeal nerves is suggested to avoid injury to these structures [9]. The hypoglossal nerve passes close to the typical position of a branchial cyst, and there is at least one case in the literature of a hypoglossal nerve injury following branchial cyst excision [6].

Injury to the hypoglossal nerve is uncommon but may happen at any point along its course. This can be divided into five segments: the medullary, cisternal, skull base, nasopharyngeal/oropharyngeal carotid space, and sublingual segments [10]. The fibers arise in the hypoglossal nuclei and extend through the medulla. In the cisternal segment, the nerve lies posterolaterally to the vertebral artery within the premedullary cistern before entering the skull base through the hypoglossal canal. It emerges from the skull base to enter the nasopharyngeal carotid space initially lying deep to the internal carotid artery, internal jugular vein, vagus, and glossopharyngeal nerves. It passes inferiorly, lying between the internal carotid artery and internal jugular vein and superficial to the vagus before looping anteriorly at the level of the angle of the jaw. Here it becomes superficial, lying inferior to the posterior belly of digastric and at risk during procedures such as submandibular gland and branchial cyst excision. At the level of the hyoid bone, it crosses the lingual artery to run along the surface of hyoglossus before penetrating genioglossus.

A cranial nerve palsy in the presence of an enlarging neck lump is generally considered to be suggestive of local malignancy or metastatic lymph nodes [8]. Hypoglossal

nerve palsies are uncommon and often present as signs rather than symptoms. Kean [5] published a series of 100 cases of 12th nerve palsy over 26 years at a single center. Tumors, mostly malignant, accounted for 49 of the cases; trauma (mainly gunshot wounds) was the second most common cause with 12 cases. Stroke (six cases), multiple sclerosis (six cases), surgery (five cases), Guillain-Barre neuropathy (four cases), and infection (four cases) were responsible for the remaining cases. The most common tumors were metastatic carcinoma, chordoma, lymphoma, and nasopharyngeal carcinoma. The infective causes were brainstem encephalitis (two cases), nasal sinus mucormycosis, viral meningitis, and sub-acute meningitis. Interestingly, he found only 15% of cases experienced complete or near complete recovery. There have been several smaller case series published in the literature [1,11] reporting a similar pattern, with roughly half of the cases due to malignancy. Branchial cysts were not reported.

There have been few reports of branchial cysts causing cranial nerve palsies. The authors have found three previous reports. In two cases, the branchial cysts were found to manifest in the parapharyngeal space, rather than the more common location at the anterior border of sternocleidomastoid [2,7]. Both cases reported multiple palsies, involving the 9th, 10th, and 12th cranial nerves. Two cases of isolated hypoglossal nerve palsies in the presence of a branchial cyst have been reported [3]. In both of these cases, the hypoglossal nerve was found to be congested intra-operatively and histology confirmed an infected branchial cyst. Our case was also shown to have an infective component on FNA, MRI, and histology. The hypoglossal did not appear congested. In all these cases, the nerve paralysis can be attributed to an acute irritation of the nerve following a rapid enlargement of the cyst, probably exacerbated by the infectious inflammatory process.

4. Conclusion

In summary, we present a highly unusual case of a branchial cyst causing an isolated hypoglossal nerve palsy. A cranial nerve palsy, although suspicious, is not pathognomonic of malignancy. A rapidly growing, infected branchial cyst may also present with lower cranial nerve palsies.

References

- [1] O. Combarros, A. Alvarez de Arcaya, and J. Berciano, *Isolated unilateral hypoglossal nerve palsy: nine cases*, J Neurol, 245 (1998), 98–100.
- [2] T. J. Durrant, R. J. Sevic, C. Laurysen, and M. E. MacRae, *Parapharyngeal branchial cleft cyst presenting with cranial nerve palsies*, Can Assoc Radiol J, 45 (1994), 134–136.
- [3] A. Gatot, F. Tovi, D. M. Fliss, and I. Yanai-Inbar, *Branchial cleft cyst manifesting as hypoglossal nerve palsy*, Head Neck, 13 (1991), 249–250.
- [4] M. Gleeson, ed., *Scott-Brown's Otorhinolaryngology, Head and Neck Surgery*, Hodder Arnold, London, 7th ed., 2008.

-
- [5] J. R. Keane, *Twelfth-nerve palsy: Analysis of 100 cases*, Arch Neurol, 53 (1996), 561–566.
 - [6] S. K. Mukherjee, C. B. Gowshami, A. Salam, R. Kuddus, M. A. Farazi, and J. Baksh, *A case with unilateral hypoglossal nerve injury in branchial cyst surgery*, J Brachial Plex Peripher Nerve Inj, 7 (2012), 2.
 - [7] J. H. Shin, H. K. Lee, S. Y. Kim, H. W. Park, S. K. Khang, C. G. Choi, et al., *Parapharyngeal second branchial cyst manifesting as cranial nerve palsies: MR findings*, AJNR Am J Neuroradiol, 22 (2001), 510–512.
 - [8] S. M. Sobol, *Benign tumours*, in Comprehensive Management of Head and Neck Tumors, S. E. Thawley and W. R. Panje, eds., WB Saunders, Philadelphia, 1987, 1353–1383.
 - [9] V. Thomaidis, K. Seretis, D. Tamiolakis, N. Papadopoulos, and I. Tsamis, *Branchial cysts. A report of 4 cases*, Acta Dermatovenereol Alp Panonica Adriat, 15 (2006), 85–89.
 - [10] E. O. Thompson and W. R. Smoker, *Hypoglossal nerve palsy: A segmental approach*, Radiographics, 14 (1994), 939–958.
 - [11] C. Tommasi-Davenas, A. Vighetto, C. Confavreux, and G. Aimard, *Causes of paralysis of the hypoglossal nerve. Apropos of 32 cases*, Presse Med, 19 (1990), 864–868.