Second branchial cleft cyst causing internal jugular vein thrombosis – A case report

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Abstract

The branchial cleft cyst also known as lateral cervical cyst is usually present in the lateral part of neck deep to sternocleidomastoid muscle at the junction of its upper third and lower two thirds. Branchial cysts are known for repeated infection with sudden increase in size and pain and for its recurrence. Thrombosis of major vessels of neck secondary to recurrent infection and inflammation of branchial cyst is extremely rare. Here we present the first case of internal jugular vein thrombosis; a rare complication seen with branchial cyst in a 54-year-old male. The patient was treated initially with antibiotics followed by excision of cyst and ligation of internal jugular vein without any complication.

Keywords Branchial · Cyst · Thrombosis · Infection

Introduction

Branchial anomalies may present as a cyst, a sinus or a fistula. Second branchial cleft cysts are the most common type of branchial anomalies and usually found high in the neck. The majority present as asymptomatic neck swelling but around one third of patients present as rapidly enlarging mass due to infection [2]. The peak age for branchial cyst is the third decade of life. Branchial cysts are lined by stratified squamous epithelium and 80% have lymphoid tissue in the wall. The treatment of choice is surgical removal because of the natural history of increase in size and liability for repeated infections [3]. The unusual complications of infected second branchial arch cleft cyst involving neurovascular structures of neck are very rare. On review of world literature only three such cases were found: one case with branchial arch cleft cyst to jugular vein fistula [4] and 2 cases with hypoglossal nerve palsy [5]. Here we present the first case of internal jugular vein (IJV) thrombosis associated with a noninfected branchial cyst.

Case Report

A 54-year-old male presented with a history of swelling in right upper cervical region of 2 years duration which had increased in size with pain since last two months. He had similar history of sudden increase in size of swelling with pain twice in past which was treated with antibiotics and repeated aspiration of swelling with resolution of symptoms but this time the swelling did not respond to antibiotics and aspiration. So the patient was referred to our institution for further management. Local examination revealed an ill defined 5x3cm size; firm, tender swelling present along the upper third of sternocleidomastoid muscle and from the lower part of swelling a cord like structure was extending downward. The patient received intravenous antibiotics for 7 days with little improvement in symptoms. Seven ml of straw colored fluid was aspirated from the swelling and the cytology showed squamous epithelium and lymphoid cells.

Key Message

Thrombosis of internal jugular vein secondary to recurrent infection and inflammation of branchial cyst is extremely rare. Here we present the first case of internal jugular vein thrombosis in a 54-year-old male. The patient was treated initially with antibiotics followed by excision of cyst and ligation of internal jugular vein without any complication.

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Computerized tomography (CT) scan of neck revealed a homogenous cyst with hyperdense capsule in right upper neck posterior to submandibular gland and infiltrating into upper third of sternomastoid muscle anteromedially (Fig. 1). The cyst was reaching up to the carotid vessels with thrombosis of IJV. Based on history, clinical examination and radiology, a diagnosis of branchial cyst was made. Surgical exploration was done via transcervical approach which revealed a ill defined cystic mass with a very thick wall firmly adherent to sternomastoid muscle, capsule of submandibular gland and underlying carotid sheath. There was loss of natural tissue planes with signs of inflammation. The cyst was dissected by sharp dissection. The carotid sheath was opened and the cyst was found firmly adherent to the IJV, which found to be thrombosed. The upper and lower limits of thrombosed segment of IJV were dissected out. Superiorly the thrombosis extending superiorly up to

Fig. 1 Contrast enhanced computerized tomography axial section showing hypodense lesion infiltrating into sternomastoid muscle and submandibular gland with thrombosis of internal jugular vein on right side.

Fig. 2 Cyst wall lined by stratified squamous epithelium with lymphoid tissue in wall

the posterior belly of diaphragm and inferiorly to the level of inferior belly of omohyoid muscle. The cyst was dissected out completely along with thrombosed part of wall of IJV. The patient made an uneventful post-operative recovery. Histopathology of excised specimen confirmed the diagnosis of branchial cyst (Fig. 2). The patient is asymptomatic 2 months after surgery.

Discussion

Cervical fistulas resulting from anomalous development of branchial apparatus were first reported in 1832 [6]. It is crucial to understand the embryology of the cervical area in order to understand the potential anatomic location of branchial cysts, sinuses and fistula. The main complications of branchial cysts are recurrent infections, recurrence after surgery and injury to adjacent neurovascular structures those become encased as a result of the inflammatory process. Deafness is also linked to branchial anomalies, but this probably represents another manifestation of anomalous development rather than a manifestation of fistula or sinus [7]. Rarely, carcinoma may develop in a branchial cleft cyst [8]. Gatot A. et al [5] have reported hypoglossal nerve palsy in two patients with infected second branchial arch cyst. Rosemn JM et al [4] have described one case of branchial cleft cyst to jugular vein fistula as a complication of recurrent episode of infection in branchial cleft cyst. A review of world literature indicates that ours is the first reported case of internal jugular vein thrombosis as a complication of infected branchial cyst. There are few important factors to discuss the cause of IJV thrombosis in this particular case. First, the patient's reluctant to undergo an excision of the cyst before multiple recurrences of infection and aspiration of swelling may explain the eventual thrombophlebitis and subsequent thrombosis of IJV. Second, although aspiration of a cyst is seldom adequate for complete disappearance of swelling, repeated aspirations of cyst should be avoided. Repeated aspiration may cause IJV thrombosis in various ways; (1) spillage of cyst content into surrounding tissue may evoke an inflammatory reaction, (2) spread of infection to the surrounding tissue, (3) direct injury to the IJV. An aspiration should be done prior to formal incision and drainage of a presumed infected cyst to define the process. Third, if the infection in cyst not responding to antibiotics and a cord like structure felt extending downwards from the swelling, thrombosis of IJV should be suspected as seen in our case. CT scan confirmed the thrombosis of IJV in our case. Once diagnosis of thrombosis of IJV is confirmed, wide surgical exposure is required to get access to the thrombosed segment of IJV. Fourth, in case of IJV thrombosis, we recommend ligation of upper and lower end of IJV and evacuation of clots along with excision of cyst to prevent further propagation of thrombosis and thrombo-embolic phenomenon and also to avoid post-operative bleeding if the thrombus is displaced.

Conclusion

Potentially serious, unusual complication like IJV thrombosis can occur in a branchial cyst, particularly with recurrent episodes of inflammation and aspiration. Treating clinician must be aware of this very unusual complication. We recommend early excision of a cyst to prevent such unusual complications.

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