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Use of botulinum toxin type A in a case of persistent parotid sialocele

用A型肉毒毒素治療持續性腮腺唾液腺腫大

Sialocele is an uncommon complication of parotidectomy. Most cases resolve after conservative therapy consisting of repeated aspiration and pressure dressing. The condition is, however, occasionally resistant to such therapy. We report on a 52-year-old Chinese man who had a 10-year history of right parotid swelling. Following fine-needle aspiration cytology, Warthin's tumour was diagnosed, but after elective parotidectomy, a swelling developed and parotid sialocele was diagnosed. Botulinum toxin type A was given after the sialocele had persisted for almost 3 weeks after surgery, and after conservative management had been tried; the sialocele disappeared after two doses of treatment. Botulinum toxin therapy was thus an effective method of treating persistent sialocele.

唾液腺腫大是腮腺切除術後一個比較罕見的併發症，患者在反覆接受抽吸術和壓迫貼敷的保守治療後，大多能痊癒，但是有個別情況，保守治療成效不大。本文報告的就是這樣的一個病例：患者為52歲的華裔男性，右腮腺腫脹已達10年。我們以細針吸引細胞術診斷病者患上沃爾信瘤。在進行非緊急的腮腺切除術後，腮腺出現一處腫脹，被證實為唾液腺腫大。我們初期以保守治療法處理，但術後三個星期，腫脹仍然未見消退，於是施用A型肉毒毒素，腫脹在用藥兩劑後消失。這個病例顯示，肉毒毒素療法能有效醫治持續性唾液腺腫大。

Introduction

Sialocele and salivary fistula are uncommon complications after parotidectomy: Laskawi et al[1] reported that 4% of patients who underwent parotid surgery for pleomorphic adenoma developed persistent fistula. Most cases resolve with conservative therapy consisting of repeated aspiration and pressure dressing. Occasionally, fistulae are refractory to conservative treatment. Oral anticholinergic drugs are seldom used to inhibit salivation because of the distressing side-effects. Rarely, creation of oral fistula, radiotherapy, and total parotidectomy are required for resistant cases,[2] but these are invasive methods. In this article, we report a difficult case of postparotidectomy sialocele that was successfully managed by the administration of botulinum toxin type A.

Case report

A 52-year-old Chinese man presented to the Department of Surgery, United Christian Hospital, in December 2000 with a 10-year history of right parotid swelling. The mass had enlarged gradually over the past year. Physical examination revealed a 3 cm x 3 cm soft parotid mass. There was no cervical lymphadenopathy. Fine-needle aspiration (FNA) cytology confirmed the diagnosis of Warthin's tumour. However, pain developed and the tumour enlarged further because of the formation of a haematoma after the FNA. The haematoma resolved with conservative treatment. Elective parotidectomy was performed in March 2001.

During the operation, a 2.5-cm diameter cystic tumour was found at the lower pole of the deep parotid lobe. Dense tissue adhesion was present around the tumour because of the previous haematoma. Subtotal parotidectomy was

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performed, thereby preserving the facial nerve. A suction drain was inserted at the conclusion of surgery.

The drain was removed 4 days after surgery and the patient was discharged home the following day. He was admitted again, however, on postoperative day 12 because a swelling had developed under the surgical wound. There was no discharge from the wound and the patient remained afebrile. Aspiration of the swelling yielded 6 mL serosanguinous fluid containing more than 20 000 units of amylase. The diagnosis of sialocele was thus made. Despite repeated aspiration and fasting for 5 days, the sialocele quickly relapsed. Because the sialocele had persisted for almost 3 weeks after surgery, continuation of conservative therapy was unlikely to be rewarding. Hence, the usage of botulinum toxin[2,3] was considered. Two doses of botulinum toxin type A (Botox; Allergan Botox Ltd, Westport, County Mayo, Ireland), of 50 and 70 units, were administered percutaneously in the parotid region around the sialocele 4 days apart. Almost immediately after the second injection, the sialocele disappeared, even though the patient had resumed oral nutrition after the first botulinum toxin treatment. The patient was discharged 8 days after the readmission. He has been followed up for more than 14 months. There has been no evidence of recurrent sialocele or facial nerve injury related to the botulinum toxin injection.

Discussion

Postparotidectomy or post-traumatic sialocele can sometimes be unresponsive to any therapy.[2,3] Significant discomfort is experienced by patients, and postoperative recovery is prolonged. If untreated, the sialocele may also lead to abscess formation or salivary fistula. The authenticity of sialocele in this case was proven by persistence of serous collection beyond 2 weeks postoperatively and a high amylase level. We attributed the cause of sialocele in this patient to the presence of peritumoural fibrosis as a result of the haematoma formation after the preoperative FNA. The healing of the parotid remnant was thus impaired.

A case of persistent parotid fistula that was successfully treated by using botulinum toxin type A has been reported.[4] The patient was a Chinese man who developed an intractable salivary fistula after parotidectomy in a private hospital in Hong Kong. This is the only case of parotid fistula successfully treated by botulinum toxin type A reported in Hong Kong. The drug acts by blocking acetylcholine release, thereby inhibiting neurotransmission at the secretomotor parasympathetic autonomic nerve ending responsible for salivation.[5] Likewise, as demonstrated in this case, botulinum toxin type A is also efficacious for parotid sialocele when conventional therapy fails. Marchese Ragona et al[2] and Vargas et al[3] have also reported the use of botulinum toxin in treating cases of parotid sialocele resistant to conventional modes of treatment. They asserted that botulinum toxin is a highly effective, safe, and non-invasive therapy for this condition. The clinical effect of botulinum toxin type A starts after 3 days,[5] as was illustrated in our case: the sialocele vanished 4 days after the administration of the first dose. In hindsight, the second dose of botulinum toxin type A was probably not needed.

Apart from its use to manage sialocele and salivary fistula, botulinum toxin has also been used to treat Frey's syndrome[6,7] and sialorrhoea,[8,9] with high efficacy and safety. The patient in this case is free of any adverse effect related to the use of this drug. However, temporary muscle weakness after therapy has been reported in the literature.[5] Given that botulinum toxin acts on the motor end-plate, complications can be avoided by giving the injection away from the vicinity of the mouth or eye. In conclusion, botulinum toxin type A should be considered as an alternative treatment for postparotidectomy sialocele when conservative treatment fails.

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