CASE REPORT

Bilateral isolated submandibular gland mumps

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SUMMARY

Isolated submandibular swellings pose a diagnostic challenge to the practising otolaryngologist. We report an unusual case of mumps isolated to bilateral submandibular glands. We discuss the case and the literature surrounding this condition and remind clinicians that mumps should be considered as a diagnosis in the presence of submandibular gland swelling in the absence of typical parotid swelling associated with mumps. Early consideration of this differential diagnosis, serological testing and a multidisciplinary approach may help to clinch the diagnosis earlier and prevent spread of the virus.

BACKGROUND

Mumps is an infection caused by a paramyxovirus via droplet spread.¹ Since the widespread uptake of the vaccine for mumps in the UK from 1988, the incidence of mumps infections has greatly decreased.² The typical presentation of mumps includes enlargement of the parotid salivary glands, but may also lead onto complications such as meningitis and orchitis.¹ We present an unusual presentation of mumps isolated to the submandibular salivary glands and review the literature associated with this case.

CASE PRESENTATION

A 19-year-old female student otherwise previously fit and well, presented acutely with a <24 hour history of right-sided neck swelling in the submandibular region. She quickly developed further

symptoms including fever, sore throat, trismus and the inability to eat and drink. There was no history of preceding prodromal symptoms of coryza. On clinical assessment, she demonstrated a tender diffuse swelling in the right submandibular region with associated trismus. There was no obvious abnormality of the palate or oral cavity and flexible nasolaryngoscopy was unremarkable aside from a mild degree of oedema of the right aryepiglottic fold and lateral pharyngeal wall. She was feverish with a temperature measured at 38.9°C and tachycardic at 122 beats per minute but other clinical observations were within normal limits. Serology showed a raised C reactive protein, measuring 41 mg/L (reference range <5 mg/L), but urea and electrolytes, full blood count and liver function tests were otherwise unremarkable. A blood culture yielded no growth.

A CT scan with contrast was requested to investigate her unilateral neck swelling further and to exclude a deep space neck abscess. This confirmed the presence of a swollen right submandibular salivary gland and lymphadenopathy. Note was made of fluid on the right side of the neck tracking along the subcutaneous areas although there was no abscess (figure 1).

TREATMENT

She initially responded to treatment with intravenous penicillin and metronidazole with dexamethasone for a presumptive diagnosis of a parapharyngeal infection. She was discharged on oral antibiotics having made some improvement after 3 days of receiving in-patient therapy. Prior to discharge, the pharyngeal swelling had subsided.

OUTCOME AND FOLLOW-UP

Two days later she represented with new onset left-sided submandibular swelling associated with difficulty swallowing. The parotid glands were not swollen, and there was no palpable cervical lymphadenopathy. The pharyngeal swelling had not recurred. An ultrasound scan confirmed bilateral submandibular gland enlargement and inflammation; the parotid glands were noted to be normal. Advice was sought from physician colleagues for further appropriate investigations. Serology confirmed mumps positive but other autoimmune markers were negative (table 1). Her antibiotic treatment was stopped and she made an uneventful recovery after discharge from hospital.

DISCUSSION

Mumps paramyxovirus causes inflammation of the parotid salivary glands in between 40% and 85%

Figure 1 Axial CT scan undertaken with contrast showing unilateral submandibular gland swelling, with associated parapharyngeal oedema but no deep space neck abscess.

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Table 1 Serological and viral throat swab results during second admission, now with isolated bilateral submandibular swelling

Test	Result	Reference ranges
Total white cell count	7.0×10 ⁹ /L	4–10×10 ⁹ /L
Haemoglobin	15.3 g/dL	11.5–16.5 g/dL
Sodium	134 mmol/L	135–145 mmol/L
Potassium	4.41 mmol/L	3.5–5 mmol/L
Urea	4.3 mmol/L	2.5–6.6 mmol/L
Creatinine	78 µmol/L	62–124 µmol/L
Serum albumin	38 g/L	35–50 g/L
Alkaline phosphatase	43 U/L	30–105 U/L
Aspartate transaminase	46 U/L	7–35 U/L
Serum bilirubin	16 µmol/L	1–21 µmol/L
C reactive protein	4.6 mg/L	0–6 mg/L
Blood culture	No growth after 5 days incubation	
Connective tissue antinuclear antibody screen	Negative	
Antineutrophil cytoplasmic antibodies (perinuclear ANCA IgG and cytoplasmic ANCA IgG)	Negative	
Toxoplasma IgG	Negative	
Mumps IgM	Positive	
Mumps IgG	Positive	
Viral throat swab	Mumps viral RNA beta-2 microglobulin detected	

of cases.^{1 3} Complications of the infection include meningitis, encephalitis, orchitis in postpubertal males, pancreatitis and pericarditis.^{1 4}

The differential diagnosis of an adult patient presenting with unilateral submandibular swelling and trismus does include viral infections, but acute sialadenitis and parapharyngeal collections are more commonly seen since widespread uptake of the mumps vaccine.⁵ Outbreaks of the virus can be found even in vaccinated populations, particularly among young adults in close contacts, such as in our case, but ensuring boosters are given to such groups helps to limit the spread.⁶ Unfortunately, despite the vaccine being very effective, it fails to confer full immunity in 100% of individual cases.⁶

Bilateral gland involvement is common following a seemingly unilateral infection. This has been observed in others with affected parotid glands.³ The second gland is affected within 2–3 days of the initial gland swelling. The virus has a 12-day to 24-day incubation period, and salivary gland swelling is usually the marked symptom of the infection, maximal within 48 hours, but resolves within 3–7 days.^{1 4} The mumps infection is usually self-limiting, but affected patients may require supportive or more specific therapy if the symptoms are particularly troublesome, or in event of secondary complications.¹

There are currently no other reported cases of mumps affecting isolated bilateral submandibular glands from the UK, however, simultaneous parotid and submandibular salivary gland involvement has been reported in up to 11% cases and isolated submandibular gland involvement in 1.3% in other countries.³ Raised IgM or IgG antibodies are required to reliably diagnose the infection.⁷ One Korean case reported isolated bilateral submandibular gland involvement with secondary orchitis and meningitis in a young adult male,³ and a further case reporting mumps with both parotid and submandibular adenopathy was eventually diagnosed as lymphoma.⁴ Together, these cases serve as a useful reminder to clinicians to remain wary of atypical presentations of submandibular swellings.

CONCLUSION

On review of the case and the literature surrounding this condition, we remind clinicians that mumps should be considered as a diagnosis in the presence of submandibular gland swelling in the absence of typical parotid swelling associated with mumps.

Learning points

- ▶ Although mumps most commonly affects the parotid salivary glands, isolated infection of the submandibular salivary glands can occur.
- ▶ Bilateral submandibular mumps is rare with few reported cases in the medical literature.
- ▶ Serological testing and a multidisciplinary team approach may help to clinch the diagnosis earlier.

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