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Spontaneous retropharyngeal haematoma attributable to Epstein-Barr virus infection

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Abstract
Life-threatening sequelae of Epstein-Barr virus infection are uncommon but may present as: local pharyngeal manifestations, splenic rupture, neurological and haematological disorders and altered hepatic function. We present a case of retropharyngeal haematoma with posterior hypopharyngeal wall necrosis, thrombocytopenia and altered clotting function as a result of Epstein-Barr virus infection. A review of the literature on retropharyngeal haematoma reveals this to be the only recorded case which can be directly attributed to Epstein-Barr virus infection.

Key words: Epstein-Barr virus; Haematoma; Pharynx

Introduction

Life-threatening sequelae of Epstein-Barr virus infection are rare, as are retropharyngeal haematoma resulting from any cause. The following case is the first report of a retropharyngeal haematoma occurring as a direct consequence of Epstein-Barr virus infection.

Case report

A previously fit thirty-year-old man was admitted complaining of a severe sore throat, dysphagia and multiple cervical lymphadenopathy. On examination he was pyrexial (39.7 °C) with hyperplastic tonsils coated by fibrinous exudate, and with hepato-splenomegaly. Initial investigations included a positive monospot test with atypical lymphocytes in the peripheral blood film confirming a diagnosis of Epstein-Barr virus infection. He was found to be thrombocytopenic (platelets 56×10^9/l) with a prolonged activated partial thromboplastin time (41 sec). In addition there was evidence of hyponatraemia (Na 128 mmol/l) and abnormal liver cell function (bilirubin 33 micromol/l, alanine aminotransferase 90 U/l, gamma-glutamyltransferase 158 U/l). His initial management involved correction of fluid and electrolyte imbalance together with intravenous hydrocortisone.

Over subsequent days his clinical condition deteriorated and on the eighth day of admission he developed a severe hypopharyngeal bleed. Profuse haemorrhage from the throat occurred at an approximate rate of 500 ml each half hour. Resuscitation with packed red blood cells and fresh frozen plasma successfully arrested the bleeding before any surgical intervention became necessary. Post-haemorrhage the patient was unable to swallow and had a noticeably swollen neck. He was continuously expectorating small clots of blood and blood stained sputum. Nasendoscopic examination showed extensive hypopharyngeal swelling with visible erosion of the posterior hypopharyngeal wall which appeared to be the site of the bloody expectorate. A CT scan examination of the neck and thorax (Figures 1 and 2) showed several abnormalities. A large haematoma was seen in the retropharyngeal area displacing the larynx forwards and extending inferiorly to the level of the carina. There was compression of the oesophagus and trachea behind the sternum. Distension of the jugular veins in the neck was also evident as a result of compression of the superior vena cava in the mediastinum. Surgical emphysema was noted near the top of the mass behind the hyoid bone on the left and multiple small lymph nodes were seen in the superior mediastinum and the neck.

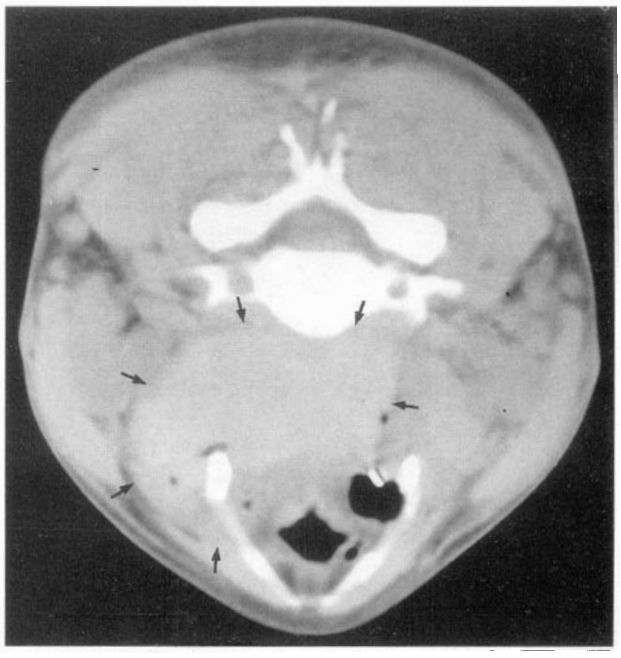

FIG. 1
CT Scan at level of C5. Anterior displacement and compression of the pharynx by a large retropharyngeal haematoma.

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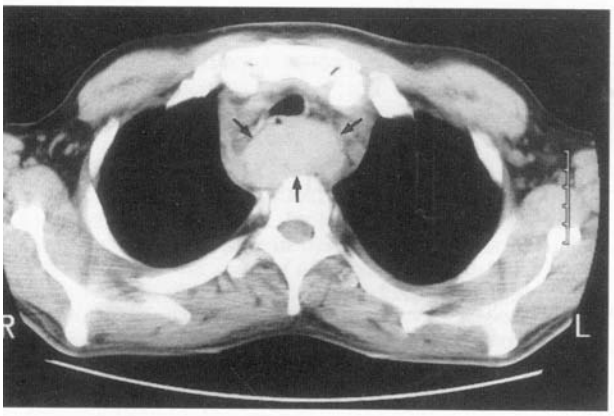

FIG. 2
CT Scan at level of T4. Superior mediastinal mass causing compression of the trachea. Multiple small mediastinal lymph nodes.

Conservative management consisted of prophylactic antibiotics and nasogastric tube feeding. However, one week after the initial bleed a swinging pyrexia was noted prompting a second CT scan to look for abscess formation. Resolution of the haematoma was seen but also the development of a large sinus directed towards the centre of the haematoma (Figure 3). A gastrograffin swallow (Figure 4) confirmed that the sinus extended deeply in to the superior mediastinum. Closure of this sinus was observed by sequential gastrograffin swallows. The volume of expectorated blood clot decreased and although

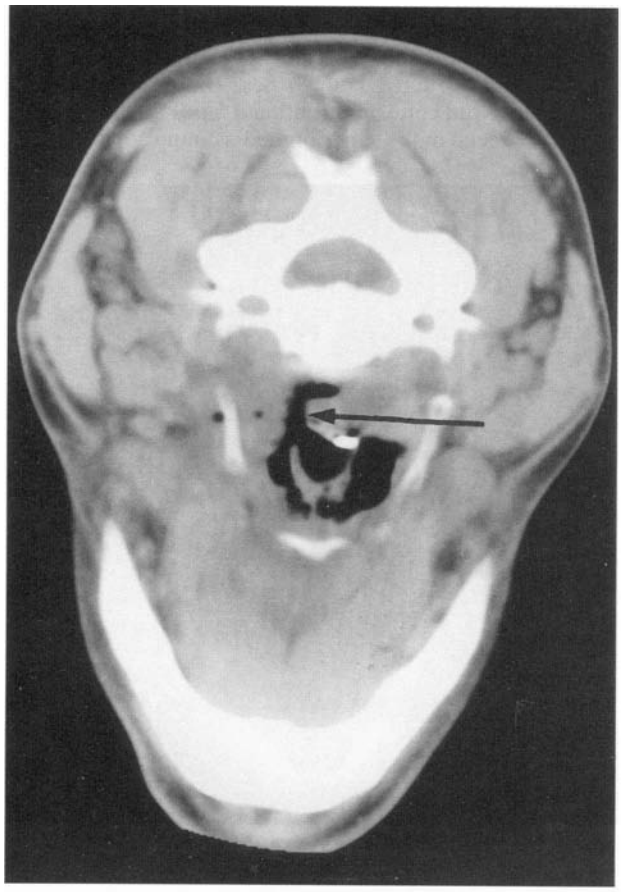

FIG. 3
Repeat CT Scan at level of C4. Significant resolution of haematoma. Posterior hypopharyngeal wall sinus extending into the haematoma.

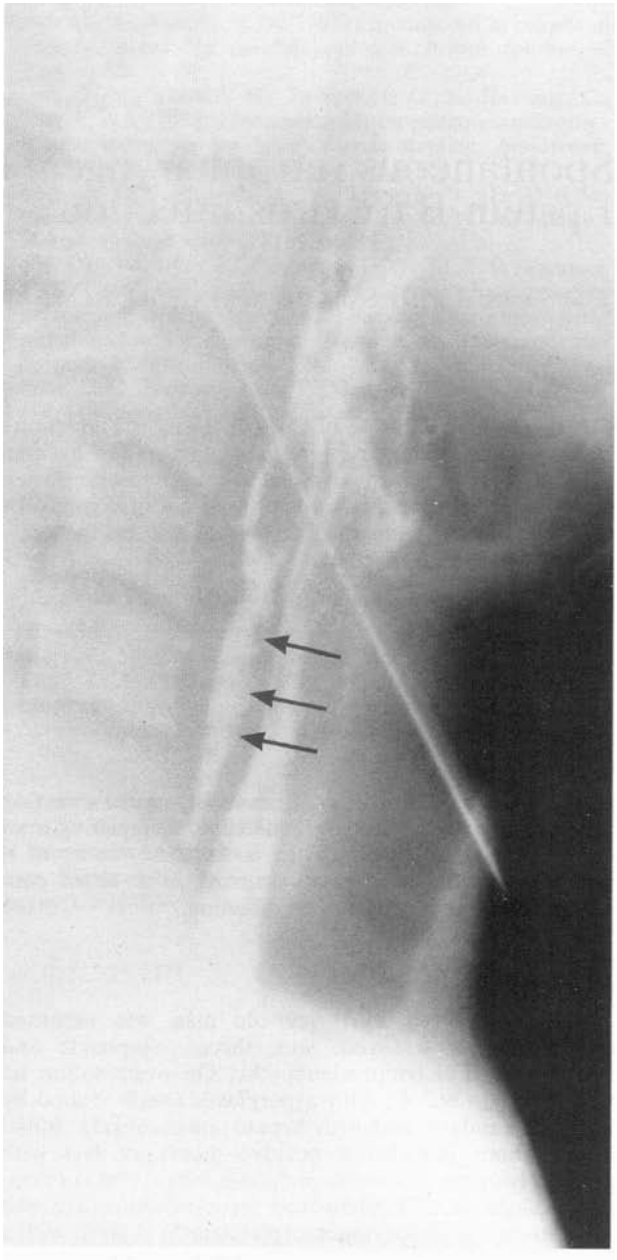

FIG. 4
Gastrograffin swallow demonstrating the caudal extension of the sinus tract into the superior mediastinum.

prolonged, recovery was largely unremarkable. Complete resolution of the haematoma and correction of all biochemical and haematological indices occurred over the following 10-week period.

Discussion

This case is another rare example of the potentially life-threatening sequelae of Epstein-Barr virus infection. Until recently, Epstein-Barr virus infection resulting in the clinical entity of infectious mononucleosis was considered a relatively benign condition (Westmore, 1990). It is often forgotten that it may be associated with serious complications, most notably: local pharyngeal manifestations; splenic rupture (Ali, 1993); neurological (Connelly and DeWitt, 1994) and haematological disorders (Levy *et al.*, 1993; Iishi *et al.*, 1991); altered hepatic function (Markin, 1994; Tazawa *et al.*, 1993) and death (Penman, 1970; Tazawa *et al.*, 1993).

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The most common local pharyngeal manifestations are those of pharyngeal and laryngeal obstruction. These may be severe enough to result in dysphagia or airway obstruction requiring surgical intervention (Stevenson *et al.*, 1992). The obstruction typically results from a combination of two processes. Hyperplasia of the pharyngeal lymphoid tissue as a consequence of viral infection and secondary laryngeal oedema resulting from a local acute inflammatory response. Rarer complications include cervical abscess formation (Westmore, 1990).

A recent review of retropharyngeal haematomas by Al-Fallouji *et al.* (1993) revealed only 23 reported cases appearing in the world literature. Of these case reports 20 had identifiable causes. Eight resulted from poorly controlled anticoagulant therapy. One occurred due to a bleeding diathesis in a patient with polycythaemia rubra vera. Three further cases were subsequently reported by Dingle *et al.* (1993). None, however, were associated with Epstein-Barr virus infection.

The commonest presentation of retropharyngeal haematoma reported to date (Al-Fallouji *et al.*, 1993) include the following triad of clinical findings: superior mediastinal obstruction, anterior displacement of the trachea and subcutaneous bruising over the neck and anterior chest wall. Our case report did not have nor develop this latter feature.

The spread of haematoma in our case followed the boundaries of the retropharyngeal space. This allowed communication with the superior mediastinum and the production of superior vena cava compression. The extensive sinus track that developed allowed resolution of the haematoma as it drained in a cranial direction through the posterior hypopharyngeal wall defect.

All reported cases of retropharyngeal haematoma were self limiting and none life-threatening therefore surgical intervention was not required (Al-Fallouji *et al.*, 1993; Dingle *et al.*, 1993). Each case has to be considered on its own merits as to whether surgical intervention to protect the airway or drain the haematoma is warranted. We chose a conservative approach for several reasons: at no point did the patient experience any life-threatening airway compromise; radiologically there was evidence that the haematoma was diffusely spread throughout the local tissues and in addition the hypopharyngeal sinus was functioning as a 'physiological' drain.

Haemorrhage from dilated superficial tonsillar vessels may occur in patients with acute tonsillitis. The haemorrhage in our case did not stem from the tonsil but from an erosion of vessels in the hypopharyngeal wall. It is difficult to explain why such an erosion and subsequent haemorrage occurred at the site it did, particularly in the absence of any trauma to the pharynx. In this case thrombocytopenia was a direct result of bone marrow suppression by the virus as has been previously noted by Iishi *et al.* (1991) and Levy *et al.* (1993). The altered clotting profile occurred as a consequence of deranged hepatic function, again a direct result of viral infection (Markin, 1994). It is reasonable to assume that these effects, together with the widespread tonsillar and hypopharyngeal mucosal necrosis, from the Epstein-Barr virus infection, combined to provide the unique conditions for this complication to occur.

This case provides yet further evidence that Epstein-Barr virus infection is not always as benign as it was once assumed to be.

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