

IPSILATERAL SYNCHRONOUS MANIFESTATION OF AN HIV-INFECTION ASSOCIATED PLUNGING RANULA AND SUBLINGUAL SALIVARY GLAND SIALOCOELE: A REVIEW AND CASE REPORT

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SUMMARY

Plunging ranula is a rare lesion and even more in HIV-infected patients. There has been only one case documented in a 15-year old that had the vertical form HIV-infection. We report a plunging ranula occurring simultaneously with a sublingual salivary gland sialocoele as two separate lesions in an HIV-infected female patient.

Key Words: Ranula, salivary glands

INTRODUCTION

Salivary gland diseases (SGD) constitute some of the manifestations associated with the human immunodeficiency syndrome (HIV) infection and acquired immunodeficiency syndrome (AIDS). It is known that HIV infection associated SGD is most common in children with a high prevalence in Africa and Latin America (Schiodt, 1992; Schiodt et al., 1992; Ranganathan and Hemalata, 2006; Chidzonga, 2007). The diseases include a vast range including lymphomas, lymphadenopathy within the parotid gland with the diffuse infiltrative lymphocytosis syndrome (DILS), parotid cysts, parotitis, Sjogren's like syndrome and the Sicca complex (Schiodt, 1992; Kazi et al., 1996; Schiodt, 1997; McArthur et al., 2003; Rivera et al., 2003). More recently, studies in Zimbabwe, South Africa, Kenya and Uganda reported ranulae as a potential oral lesion in HIV infection (Chidzonga, 2007; Syebele and Butow, 2010; Butt et al., 2010; Kamulega and Okello, 2012). Apparently there has been only one case of a plunging ranula in an African –American male with HIV infection documented in the literature (Hershkin and Miller 2007). We report an additional case of an HIV-infected African woman who presented with ipsilateral synchronous plunging and sublingual ranulae.

CASE REPORT

A 33-year-old woman presented with a chief complaint of a painless swelling below the tongue on the left side for the past 8 months while, the swelling on the same side of the neck had started three weeks ago. The intraoral swelling had gradually increased in size causing difficulty in speech and mastication. The medical history revealed a diagnosis of HIV infection and anti-retroviral therapy. Extra-oral examination showed a soft fluctuant swelling in the left submandibular area measuring 6cm by 6cm (Fig. 1) and intra-orally there was another bluish cystic swelling in the left floor of the mouth measuring 3cm by 5cm (Fig. 2). Both the swellings were painless on palpation. Based on the history and clinical examination a provisional diagnosis of a plunging ranula was made. Hematological investigations showed a marked eosinophilia, a reversed differential white blood cell count and a high ESR. The absolute CD4 cell count was 257cells/mm³ and a CD4:CD8 ratio of 0.25 indicative of a depressed immunity. Under nasotracheal intubation, the lesions were approached using cervical (Fig. 3) and intra-oral incisions. Both the left submandibular and sublingual salivary glands including the cysts were excised. The cysts were not in communication,

appearing as separate cavities within the parenchyma of their respective salivary glands (Fig. 4). Intra-orally blunt dissection was used and both the lingual nerve and the Wharton's duct were identified and preserved. No scar tissue was present around the parenchyma of the lesions. The hypoglossal nerve was identified and spared during the excision of the submandibular gland. Histopathological examination confirmed

the diagnosis of mucous extravasation cysts. This was characterised by chronically inflamed compressed fibrous connective tissue walls surrounding empty spaces and supported by haemorrhagic fibrous connective tissue with lobules of salivary serous and mucous acinii, and lymph nodes depicted lymphoid hyperplasia (Fig.5).The patient recovered uneventfully and has not shown any signs of recurrence after a year's follow up.



Fig 1. Extra-oral presentation of the plunging ranula. Fig 2 Intra-oral presentation of the ranula in the floor of the mouth



Fig 3. Intra-operative presentation: white arrow points to submandibular gland and the black arrow shows the cyst within the gland. Fig 4. Photograph showing both the sublingual and submandibular glands with the cysts. Fig 5. Histopathology of the specimen using H&E stain (X100). Glandular architecture is completely destroyed with extravasation of mucin and remnants of the ducts.

DISCUSSION

In Cameroon HIV infection associated SGD was shown to have been more severe in HIV- positive black patients compared with HIV-positive American patients, a marked fibrosis was also noted in the minor salivary glands of the African patients (Kazi et al., 1996). McArthur noted DILs to have been more common among the West Africans, demonstrating a severe salivary gland atypia (96%) - a feature strongly associated with HIV

infection and AIDS (McArthur et al., 2003). In addition increased inflammatory cell infiltration and fibrosis was also observed in the glandular parenchyma (McArthur et al., 2003; Kazi et al., 1996). It is, therefore, not surprising to note an increasing number of reports of ranulae occurring in HIV-infected patients from the African continent (Chokunonga, 1999; Chidzonga, 2007; Syebele and Butow, 2010; Butt et al., 2010; Kamulega and Okello, 2012). Although, both Chidzonga and Kamulega recorded a relatively small

number of patients with plunging ranulae in comparison to oral ranulae in their populations, they did not comment about the immune status of the cases in their population (Chidzonga, 2007; Kamulega and Okello 2010). Plunging ranula is a rare lesion and even more so in those who are HIV-infected with only one documented in a 15-year old who had the vertical form of HIV-infection. The patient had been on highly active antiretroviral treatment (HAART) when the lesion developed. The present case had the acquired form of HIV infection and was also on HAART. Clinically, it appeared as one lesion whence intra-operatively two

separate cysts originating from the sublingual and submandibular glands were encountered. Our histopathologic findings were consistent with the features reported by Kazi et al. (1996) and McArthur et al. (2003).

Whether these lesions may be more frequent in the less developed countries of the East and Southern Africa region needs further investigation through more studies. As more of HIV-infected patients present with extravasation cysts it would be interesting to note the histopathological features of the affected glands and the lesions since the etiology and pathogenesis still remains a mystery.

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