A 49-year-old man was admitted for medical examination because of epigastric discomfort. Esophagogastroscopy revealed a small, elevated, verrucoid pink-yellowish mucosal lesion, 3 mm in diameter, in the upper third of the esophagus (Fig. 1).

Low power histological examination revealed an exophytic lesion resembling a squamous papilloma, with the typical papillomatosis, acanthosis, and hyperparakeratosis of the esophageal squamous epithelium (Fig. 2a). At higher magnification, neutrophilic intraepithelial exocytosis was observed, and the subepithelial connective tissue appeared infiltrated by clear cells with foamy cytoplasm and small nuclei with no atypia (Fig. 2b, c). At immunohistochemistry, the foamy cells were negative for cytokeratins, s-100 protein, and CD1a, while CD68 was strongly positive (Fig. 2d), indicating the histiocytic nature of the cells. The histological and immunohistochemical features allowed making a diagnosis of verruciform xanthoma of the esophagus.

Verruciform xanthoma is a lesion characteristically described in the oral cavity and genital skin [1]. It is usually solitary, but cases of multifocal lesions have been reported [1]. The main histological feature is the presence of foamy histiocytes in the subepithelial stroma of a squamous epithelium displaying papillomatosis, acanthosis, and hyperkeratosis, as observed in papillomatous/verrucous lesions. Intraepithelial neutrophilic infiltration is another hallmark. The etiology is still unknown, most cases being unrelated to a viral infection. The presence of human papilloma virus in the epithelial cells has been demonstrated in only two reported lesions in the oral mucosa [2] and scrotum [3]. To the best of our knowledge, our case represents the second description in the English literature of verruciform xanthoma in the esophagus [4].