Letter to the Editor

Role of multimodality treatment in Kimura disease of head and neck region

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Dear Editor,

We read with great interest an article published in the Bangladesh Journal of Medical Science entitled 'Kimura Disease: A case report and review of literature'[1]. The author described the successful treatment of the lesion which to date has no consensus in the definitive treatment. The case was treated with combination of oral antihistamine (Cetirizine) and corticosteroid (Prednisolone). Patient was remained symptom-free for 6 months duration. Although Kimura disease (KD) is a benign condition, the general recurrence rate was reported as high as 60% to 80%[2]. In addition, the local recurrence after stop corticosteroid is not uncommon, thus we think patient need long term follow up and the corticosteroid should be tapering down and stop cautiously. Serum eosinophil probably a reasonable marker as a guide of the treatment because its level was found to be related to the size of mass and therapeutic response[3].

The pathogenesis of KD remains unknown, but allergy, atopy, autoimmunity, and parasite infestation are considered possible risk factors[4]. Surgeons around the world have tried with medical therapy, surgery or even the combination of both in order to combat the lesion. Medical therapy alone probably sufficient for small lesion, however, for huge lesion that cause facial disfigurement or obstructing the upper airway, combination of medical and surgical therapies may be more beneficial. Oral corticosteroid is the most frequent used medication due to the good and fast response; however systemic complications is the drawback in long term treatment. Successful treatment of head and neck KD with combination of 2 medications is rarely reported. One reported case of KD involving bilateral upper eyelids and parotid regions was successfully treated with combination of oral corticosteroid and intravenous methotrexate with disease free upon 2 years follow-up[4]. Combination of oral corticosteroid with other group of medication with lesser side effect profile like antihistamine is consider more favorable strategy in order to minimized possible complication of corticosteroid.

Although head and neck region, specifically parotid gland and cervical lymph node are the most common sites of KD, there are no large series or comparison study on treatment particular to that region. Most of the studies are case report or limited sample size retrospective study[5-15] (Table 1). The main treatment is superficial parotidectomy, surgical excision and oral prednisolone, either as single or combination therapies. The corticosteroid treatment used with different intentions, either to cure the disease or as adjunct to surgery. It may use before or after surgery with intention to reduce size and vascularity of the lesion in order to ease excision and reduce complication or prevent recurrence respectively.

Consistent with the published article, we think combination of at least two therapies either

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combination of two different group medications or surgery with perioperative corticosteroid or with adjuvant radiotherapy appeared promising result with lower recurrence rate compared to single modality[16-19] (Table 2). Gastrointestinal problem and steroid-induced dermatitis are between side effect of steroid that were reported and the treatment need to withdraw[17]. Moderate dose of radiotherapy (20–45 Gy within 3–5 weeks) was reported as optimal dose for local control without complication or secondary malignancy observed[17,18]. This could be explained by the advancement of radiotherapy technology recently. However, the controversial still going on and debatable, as nature of KD is considered indolent and no risk of malignant transformation.

We think the approach to head and neck KD is highly individualized and combination of multimodality treatments should be considered. Patient should be selected and explained properly regarding each treatment and need to be informed regarding high recurrence rate.

Table 1. Summary of literatures on treatment of KD in head and neck region

Authors (year)	Study, Number of patients	Duration of symptoms/ region involved	Treatment	Outcome	Follow up period
Ghafar M et al. (2019)	Case report, 1 patient	13 years/ left parotid and cervical lymph node	Left superficial parotidectomy Pre-operative oral prednisolone 60mg daily for 2 weeks then tapering dose for 4 weeks Post-operative oral prednisolone 60mg daily, duration NM	NM	NM
Sharma R (2017)	Case report, 1 patient	6 years/ right parotid gland and cervical lymph node	Superficial parotidectomy Pre-operative oral prednisolone 20mg OD for 1 year Post-operative oral prednisolone 40mg daily for 6 months, then tapering down for 6 months and finally stopped	No recurrence	1 year
Arshad AR (2003)	Retrospective study, 8 patients	1 to 20 years/ parotid gland (side NM), and cervical lymph node	Superficial parotidectomy followed by initial high-dose steroid therapy and low-dose maintenance for a period of 1 year in all cases (dosage of steroid NM)	No recurrence	NM
Woo SH et al. (2017)	Case report, 1 patient	10 years/ bilateral parotid glands and cervical lymph node	Surgical excision via transverse incision bilateral cheeks	No recurrence	4 years
Kapoor NS et al. (2012)	Case report, 2 patients	NM/ 1 patient involved left parotid gland; the other patient involved buccal region (not discuss here) and cervical lymph node	Surgical excision twice at other center (procedure NM) 3rd operation – superficial parotidectomy	No recurrence	30 years
Yazici D et al. (2007)	Case report, 1 patient	10 years/ left parotid gland and retro auricular region	Oral prednisolone, 1mg/kg per day and tapering off 1/4 of the dose every 4 days until the dose reached 2 mg/day	No recurrence (still on oral prednisolone 2mg/day)	6 months
Muniraju M et al. (2019)	Case report, 1 patient	6 years/ left parotid gland, temporal, bilateral post auricular regions and cervical lymph node	Superficial parotidectomy and excision of subcutaneous mass	NM	NM
Glibbery Net al. (2018)	Case report, 1 patient	1 year/ left parotid gland and cervical lymph node	Superficial parotidectomy	No recurrence	20 months
Singh LS et al. (2014)	Case report, 1 patient	NM/ left parotid gland and cervical lymph node	Superficial parotidectomy followed by oral cetrizine 10 mg daily	No recurrence (still on oral cetrizine 10 mg daily)	NM

Authors (year)	Study, Number of patients	Duration of symptoms/ region involved	Treatment	Outcome	Follow up period
Dik VK et al. (2010)	Case report, 1 patient	6 years/ bilateral parotid glands and cervical lymph node	Oral prednisolone 30 mg daily (duration NM)	Swelling diminished	NM
Ragu R et al. (2014)	Case report, 1 patient	6 years/ right parotid gland and cervical lymph node	Superficial parotidectomy	No recurrence	1 year

NM, not mention; ND, not discuss

Table 2. Summary of 4 studies comparing single versus combined therapies

Authors (year)	Region involved	Study, Number of patients	Treatment	Outcome	Follow up period
Ye P et al. (2016)	Armpit, bone, eyelid, BR, FR, groin, IOR, lip, LN, NC, PAR, parotid gland, SCR, SMR, SR, TR, TW, wrist, ZR	Meta-analysis of 22 reports, 570 patients	SE alone, RA alone and SRRA	-No significant difference in recurrence in SE and RA alone -RA alone has 1.718 times higher risk of recurrence compared to SRRA -SE alone has 4.722 times higher risk of recurrence compared to SRRA	2 months to 28 years
Chen QL et al. (2015)	Breast, elbow, eyelid, inguinal, NC, parietal region, PAR, parotid gland, pre-auricular SMR, SR,	Retrospective study, 32 patients			1 month to 9 years
		10 patients	SE alone	80% recurrence	
		5 patients	SES	60% recurrence	
		17 patients	SRRA	41% recurrence	
Chang AR et al. (2006)	Arm, axilla, back, cheek, forearm, groin, leg, LN, PAR parotid gland, SMG	Retrospective study, 23 patients			6 months to 10 years
		3 patients	Steroid alone	2 SD 1 PD	
		6 patients	SE alone	2 recurrence 1 Par 2 NED 1 loss follow up	
		8 patients	RA with steroid	3 recurrence 4 NED 1 SD	
		4 patients	SRRA	1 recurrence 3 NED	
		2 patients	SRRA and steroid	1 SD 1 NED	
Zhang G et al. (2020)	Parotid gland and other sites (not mention in detail)	24 patients			6 months to 9 years
		13 patients	SE alone	46% recurrence	
		7 patients	SES	71.4% recurrence	
		4 patients	SRRA	No recurrence	

BR, buccal region; FR, frontal region; IOR, infraorbital region; LN, lymph node; NC, nasal cavity; NED, no evidence of disease; Par, partial response, PAR, postauricular region; PD, progressive disease: RA, radiotherapy; SCR, supraclavicular region; SD, stable disease; SE, surgical excision; SES, surgical excision combined with steroid; SMR, submental region; SR, submandibular region; SRRA, surgical excision combined with radiotherapy; TR, temporal region; TW, thoracic wall; ZR, zygomatic region

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