CASE REPORT

A 49-year-old female noticed a deep-seated nodule with rubbery consistency on right upper arm for 2 months. Erythematous change on surface was seen (Fig. 1). She was suffered from mild itching sensation without pain or tenderness. The lesion progressed in size gradually. She visited our hospital for help and excisional biopsy was performed. Histologically, a well-defined nodule circumscribed by eosinophilic cuticle was found in subcutaneous tissue. The wall of nodule consisted of three layers, including eosinophilic integument, smooth muscle fibers, and columnar tegumental cells. Numerous basophilic calcareous bodies, excretory ducts, and smooth muscle bundles were noted in loose stroma of parenchyma (Fig. 2). Besides, bothrium, a suction groove, was discovered in the cephalic direction after serial sections (Fig. 3). There was no pro-toscolex, suckers, or hooklets. A dense inflammatory cell infiltrate, containing lymphohistiocytes, eosinophils, plasma cells and multinucleate giant cells, was present around the nodule. The laboratory tests revealed no leukocytosis or eosinophilia.

Fig. 1
A well-defined, 3 x 2 cm in size, deep-seated nodule with erythematous change on surface.

Fig. 2
Basophilic calcareous body (long arrow), excretory duct with eosinophilic wall (short arrow), and smooth muscle bundles in loose stroma, circumscribed by three-layered body wall. (H&E, original magnification x400)

Fig. 3
Bothrium (arrow) at the cephalic end of serial sections. (H&E, original magnification x400)
DIAGNOSIS: Cutaneous Sparganosis

DISCUSSION

Human sparganosis is an incidental infection caused by the plerocercoid larva of a cestode belonging to the genus of *Spirometra*. The first case was described by Patrick Manson in 1882. There are two forms of human sparganosis, i.e. nonproliferative and proliferative. The majority was nonproliferative sparganosis, caused by *S. mansoni* or *S. mansonoides* in most cases. Dogs and cats are final hosts of spargana. Human can be infected by drinking water contaminated with infected *Cyclops* species (the first intermediate host), eating partially cooked meat of fish, frog or snake (the second intermediate host), or applying raw flesh of infected animals on wounds or eyes as a poultice. Our patient denied drinking unboiled water or using raw flesh as a poultice. However, she is a super-lover of “sashimi” and takes raw fish almost every day. As a result, eating raw fish was inferred as the infectious source in our case. It is possible that, this is the first case of sparganosis transmitted by eating raw fish.

Spargana most commonly invade subcutaneous tissue, especially in the abdominal wall, chest and limbs. Ocular and visceral sparganosis were reported less frequently. Twenty-five cases have been reported in Taiwan since 1922 with 13 of them presenting with skin involvement. The typical feature is a slowly migrating subcutaneous nodule. Erythema, pruritus, and local tenderness may develop as well.

Histologically, calcareous body, excretory canals, and smooth muscle fibers can be seen in loose parenchyma circumscribed by the three-layer body wall, which consists of eosinophilic integument, smooth muscle fibers, and columnar tegumental cells. Calcareous body is the characteristic finding of cestode. Other possible cutaneous cestode infection presenting as a subcutaneous nodule, including coenurosis, cysticercosis, and echinococcosis, should be taken into differential diagnoses of cutaneous sparganosis. However, cystic cavity and protoscolex seen in coenurosis and cysticercosis were absent in our patient. Echinococcosis can be differentiated from the sparganosis by its laminated thick cuticular membrane and inner germinal layer, which is not the situation in our case. Besides, bothrium is not present in other situations except sparganosis.

Total excision of the nodule where the parasite lives is considered curative. If removing incompletely, residual segments of the sparganum can continue to survive. Antiparasitic therapy is not proved effective.

In conclusion, cutaneous sparganosis is an incidental infection of human. The clinical presentation as a subcutaneous nodule is frequently misdiagnosed as a lipoma. However, the definite diagnosis can be made by histological examination. Unlike other reported cases of human sparganosis which were infected by drinking contaminated water, our patient denied drinking unclean water or using raw meat as a poultice. The history makes us pay attention to the possibility of disease transmission by sashimi. This case reminds us sparganosis should be considered when encountering a common subcutaneous nodule, which is usually thought as a lipoma clinically, and detailed history including dietary habits and animal contact must be inquired.

REFERENCES