CASE REPORT

Sclerosing lipogranuloma: an unusual scrotal mass

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Sclerosing lipogranuloma of the male genitalia without a history of injection of exogenous material is extremely rare. This is the first case reported from a Canadian center. This 33 year old man developed sclerosing lipogranuloma of his scrotum 3 months after being diagnosed with infectious mononucleosis. There was no history of injection of exogenous substances or trauma. His lesion was painless, sudden in onset, "Y-shaped", associated with eosinophila and spontaneously regressed after partial resection.

A review of the available English literature on sclerosing lipogranuloma from 1966 to 2001 was completed to compare our case report to previously available reports. The results show definite differences in the presentation of primary versus secondary sclerosing lipogranuloma. Sixty-eight per cent of the cases of primary sclerosing lipogranuloma involved the scrotum only while 63% of secondary sclerosing lipogranuloma involved the penis only. Seven per cent of lesions attributed to primary sclerosing lipogranuloma were painful compared to 69% of secondary sclerosing lipogranulomas. Cases of primary sclerosing lipogranuloma were often described as "Y-shaped" and were unlikely to recur.

Understanding the typical presentation of this condition will allow future cases to be recognized more easily and managed appropriately. Primary sclerosing lipogranuloma may be diagnosed by fine needle aspiration or excisional biopsy and then managed conservatively avoiding more complex and invasive surgery.

Key Words: sclerosing lipogranuloma, paraffinoma

Introduction

Sclerosing lipogranuloma is an inflammatory response to endogenous lipid degeneration which usually occurs in the subcutaneous tissue of the scrotum or on the dorsal side of the penis[1] but the etiology is controversial. It is often referred to as a "paraffinoma" when it is secondary to injection of exogenous substances such as paraffin, silicone and mineral oil in an effort to enhance erection, penile size or sexual potency.[2] Without a history of injection of exogenous material, it is referred to as primary sclerosing lipogranuloma caused, hypothetically, by trauma, inflammation, infection (e.g. tuberculous or fungal) or allergy.[1-3]

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TABLE 1. **Review of 71 cases of sclerosing lipogranuloma**

Source	Age	Site	Shape	Painful or painless
Hirokawa[1]	62	scrotum, dorsal penis	Y	-
Sahin[16]	17	ventral, dorsal penis	5 firm nodules	painful
Matsushima[6]	54	inguinal area	1 mass	painless
	36	scrotal septum	1 mass	painless
Golomb[17]	37	midline scrotum	1 mass	painless
Carlson[18]	78	scrotum, penis	many large nodules	-
Watanabe[5]	29	scrotum	round	painless
	36	scrotum	Y	painless
	40	scrotum	Y	painless
	29	scrotum	circular	painless
	39	scrotum	Y	painless
Matsuda[4]	44	scrotum	Y	painless
	45	scrotum	Y	painless
	29	scrotum	Y	painless
	37	scrotum	Y	painless
Takihara[3]	37	scrotum	Y	painless
	39	scrotum	Y	painless
Claudy[19]	52	pubic area, scrotum, penis	-	-
Rollins[20]	48	scrotum, lungs	many masses	-
Lee[2] 26 cases	mean of 39.6	penis	-	73% painful
Nakamura[9]	47	penis	1 lobulated mass	painful
Baladas[10]	31	scrotum	1 swelling	-
Arthaud[8]	51	suprapubic, penis	1 lobulated mass	painful
Oertel[7] 23 cases	mean of 34	scrotum, penis perineum, breast	-	-

Duration	Evidence of exogenous substances	Treatment	Follow up
2 weeks	none	needle aspiration, partial resection	no progression after 9 months
6 months	none	partial resection x 3	rapid recurrence after 2 resections
11 months	none	right orchidectomy	no recurrence x 56 months
-	none	complete resection	no recurrence x 45 months
-	none	complete resection	no recurrence x 3 months
-	none	partial resection, prostatectomy	-
7 days	none	-	-
10 days	none	-	-
14 days	none	-	-
21 days	none	-	-
23 days	none	-	-
2 weeks	none	partial resection	spontaneous regression 2/12
2 weeks	none	partial resection	spontaneous regression 2/12
3 days	positive	partial resection	spontaneous regression 2/12
1 month	none	partial resection	spontaneous regression 2/12
1 month	none	partial resection	no recurrence x 2.5 years
-	none	partial resection	no recurrence x 1 year
-	none	-	-
-	positive	death resulted	-
mean of 18.5 months	positive	various operations	-
2 years	positive	complete resection	-
6 months	positive	complete resection	
-	positive	partial resection x 2	remaining tissue did not regress
-	positive in 91%	78% excised, 26% orchiectomy	marked recurrences

Case report

A 33 year old male presented with a 2 week history of a painless, growing scrotal mass. He had been diagnosed with infectious mononucleosis 3 months earlier. There was no history of infection, trauma or injection of exogenous materials. Past medical history was unremarkable. He had no known allergies and his only medication was occasional celebrex for jaw pain.

On examination, the penis, penile urethra, urethral meatus, testes and epididymes were normal. There was a hard, irregular and mobile 3.5 cm x 2.5 cm mass arising within the midline of the scrotum and extending to almost encircle the base of the penis. It was deep but appeared to be separate from the bulbous urethra and other scrotal structures.

A scrotal ultrasound reported that both testes were normal in size, shape and echogenicity; there was a 4 cm left epididymal cyst. Doppler examination was unremarkable. A complete blood count reported WBC 5800 (27% segmented neutrophils, 2% bands, 4% eosinophils, 1% basophils and 2% atypical lymphocytes), hemoglobin 146 and platelets 246 000.

Flexible urethroscopy confirmed no urethral involvement. Exploration of the scrotum revealed that the exact origin of the mass was not discernable and that total excision would have involved a very extensive dissection up to and around the penis; several frozen sections showed

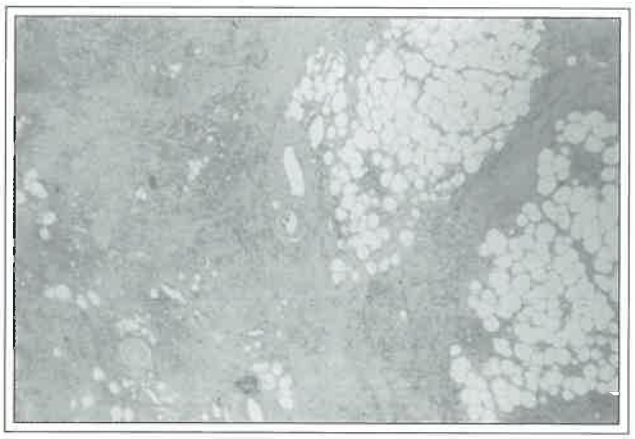

Figure 1. H & E (magnification x 40): Fibrovascular and fatty tissue with inflammatory cell reaction.

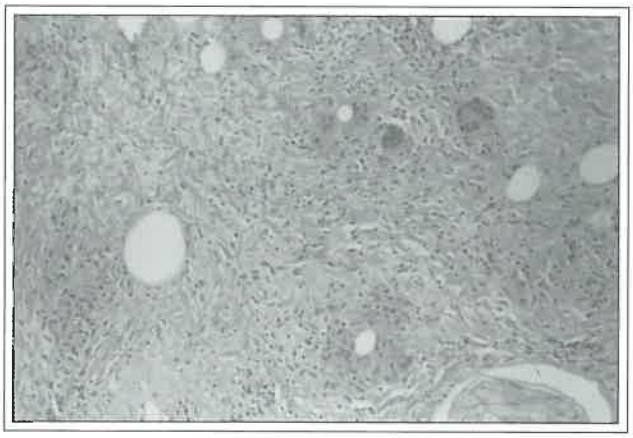

Figure 2. H & E (magnification x 160): Fibrosis with granulomatous reaction.

fig. 3

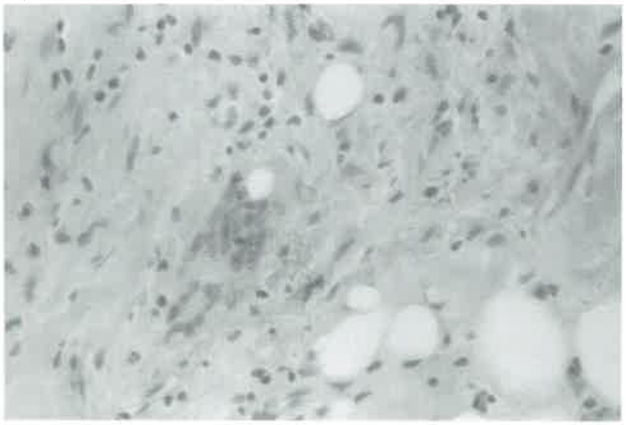

fig. 4

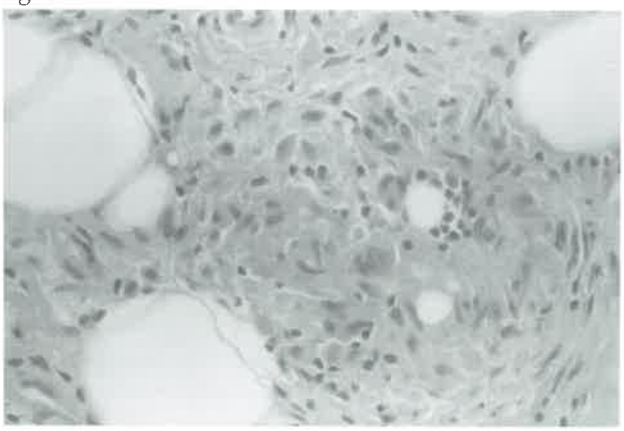

Figure 3 and 4. H & E (magnification x 400): Granulomatous reaction with nodular aggregates of epitheloid histiocytes and multinucleated giant cells. Note: multinucleated cell with cytoplasmic vacuoles.

TABLE 2. **Summary of primary compared with exogenously induced-lipogranuloma**

	Primary sclerosing lipogranuloma	Number of cases available to review	Secondary sclerosing lipogranuloma	Number of cases available to review
% with scrotum only involved	68.4	19 of 19	28.8	52 of 52
% with penis only involved	31.6	19 of 19	63.5	52 of 52
% painful	6.7	15 of 19	68.7	32 of 52

no evidence of malignancy but the exact histological diagnosis was unclear. Partial excision of the mass was performed.

Post-operative course was uneventful and the last follow-up visit at 8 months confirmed that the remainder of the mass had slowly, spontaneously disappeared.

Pathology report

The specimen was a tan to yellow, soft tissue mass measuring 3.8 cm x 2.5 cm x 2 cm. Histological sections revealed fibrosis, fatty tissue and granulomatous reaction composed of epitheloid histocytes and multinucleated giant cells (Figures 1-4). Intracytoplasmic vacuoles were present in some giant cells. Foamy histocytes, lymphocytes and many eosinophils were also noted. On polarization, no foreign material was recognized. Zeihl Nelson and periodic acid Schiff histochemical stains were negative for microorganisms. Electron microscopy showed chronic granulomatous inflammation with eosinophilia. There was no evidence of malignancy. A section of the scrotal mass subjected to DNA analysis showed no evidence of clonal proliferation of B or T lymphocytes. A month after initial excision, a final diagnosis of sclerosing lipogranuloma was made.

Discussion

This report confirms findings previously shown to be suggestive of primary sclerosing lipogranuloma and summarized in Table 1. Our patient had a "Y-shaped" painless lesion of recent onset that was growing rapidly but regressed completely after partial excision. The "Y-shaped" lesion and sudden onset are typical[1,3-5] and the lack of pain and tenderness are common.[3-6] Complete regression of the remaining mass following partial resection has been previously reported.[3,4] On the other hand, sclerosing lipogranuloma secondary to injection of exogenous material tends to recur if not completely excised[7] and tends to be associated with a longer history and pain.[8,9] Details of the comparisons between the primary sclerosing lipogranuloma cases and those associated with exogenous agents are presented in Table 2.

An association between sclerosing lipogranuloma and eosinophilia has been noted. One report showed that 3 of 4 patients had elevated eosinophils in their blood (7%, 11% and 15%) while another showed that both of their cases had eosinophilia (5% and 11%) as well as eosinophilic infiltration histologically.[3] Our patient had 4% eosinophilia and eosinophilic infiltration. The recent recognition of the association between eosinophils and primary sclerosing lipogranuloma suggests that an allergic mechanism might play a role in its etiology.[3]

There have been a few unusual case reports of sclerosing lipogranuloma. One occurred within 48 hours of a herniorraphy and varicocelectomy and was postulated to be the result of the paraffin used to lubricate the laparoscopic instruments.[10] Another paper included a description of sudden death from a self-administered, probably inadvertent intravascular, injection of mineral oil.[2] Although there are no previous reports of sclerosing lipogranuloma secondary to infectious mononucleosis, it is possible that these two entities are related. For example, the Epstein Barr virus is associated with genital lesions.[11-15]

In summary, primary sclerosing lipogranuloma is most likely to present as a painless mass involving only the median raphe of the scrotum, be "Y-shaped", be associated with eosinophilia, spontaneously regress following partial resection and not recur. If the history

and physical examination are suggestive of it, the diagnosis could be confirmed by fine needle aspiration to avoid more invasive surgery such as orchiectomy or extensive resections for suspected malignancy.[1] □

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