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Case report

Lane's type pseudosarcoma of glans penis

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Key words

Pseudosarcoma • Immunohistochemistry

Summary

We herein present a rare case of polypoid "pseudosarcomatous lesion" of the glans penis, associated with in situ or mini-invasive squamous carcinoma. These lesions, described by Lane, in the upper aerodigestive tract, can rarely occur elsewhere. Immunohistochemistry is crucial for a correct diagnostic interpretation, confirming that the aytpical cells are components (fibroblasts, myofibroblasts, endothelial) of granulation tissue.

Introduction

Aim of this report is to present a peculiar and controversial lesion in a location so far never described, and on the basis of our data and those of the literature make some interpretative considerations.

Case report

A seventy nine-year old man, has had for about 3 months an ulcerated, pedunculated polypoid mass on the ventral aspect of the glans penis in correspondence of the frenulum, the size of 2.2 cm x 1.5 x 1. Resistant to local therapies. The lesion is excised in toto.

Materials and methods

At macroscopic examination the surgical specimen was oval shaped, measured 2,2 x 1,5 centimetres x 1. The cut surface of it appeared pink, moist, granular, the outer surface smooth, covered by a layer of fibrin- necrotic material.
The entire sample, formalin-fixed and paraffin-embedded, was stained with hematoxylin-eosin and tested immunohistochemically with the following antibodies: vimentin (monoclonal 1:50 DAKO), CK AE1-AE3 (monoclonal 1:50 DAKO), EMA (monoclonal 1:50 DAKO), alpha-smooth muscle actin (monoclonal 1:50 DAKO), calponin (monoclonal 1:50 DAKO), CD68 (ready to use DAKO), S100 (polyclonal 1:400 DAKO), KI67 (monoclonal 1:75 DAKO), CD34 (monoclonal 1:20 DAKO), CD31 (monoclonal 1:20 DAKO),P63 (monoclomal 1:50 Dako).

Histology

The surface of the neoformation appears widely ulcerated. The residual squamous epithelium is strongly atypical with the characters of in situ carcinoma (Figg. 1, 2). The bulk of is formed by a connective tissue rich in vessels, some thrombosed with promi-

Tab. I.

CK Ae1- ae3	Ema	Vim	Smact	Calp	Cd68	Cd34	Cd31	S100	Ki67	P63
Surf + Fig.7	Surf +	SS +	SS + Figg. 8-9	SS + Fig. 10	SS + Fig. 11	SS + Fig. 12	SS+	-	Surf+ Fig. 13	Surf+ Fig.14

Vim=Vimentin,Smact= Smooth muscle actin, Calp= Calponin, Surf = Surface, SS = Sarcomatoid Stroma

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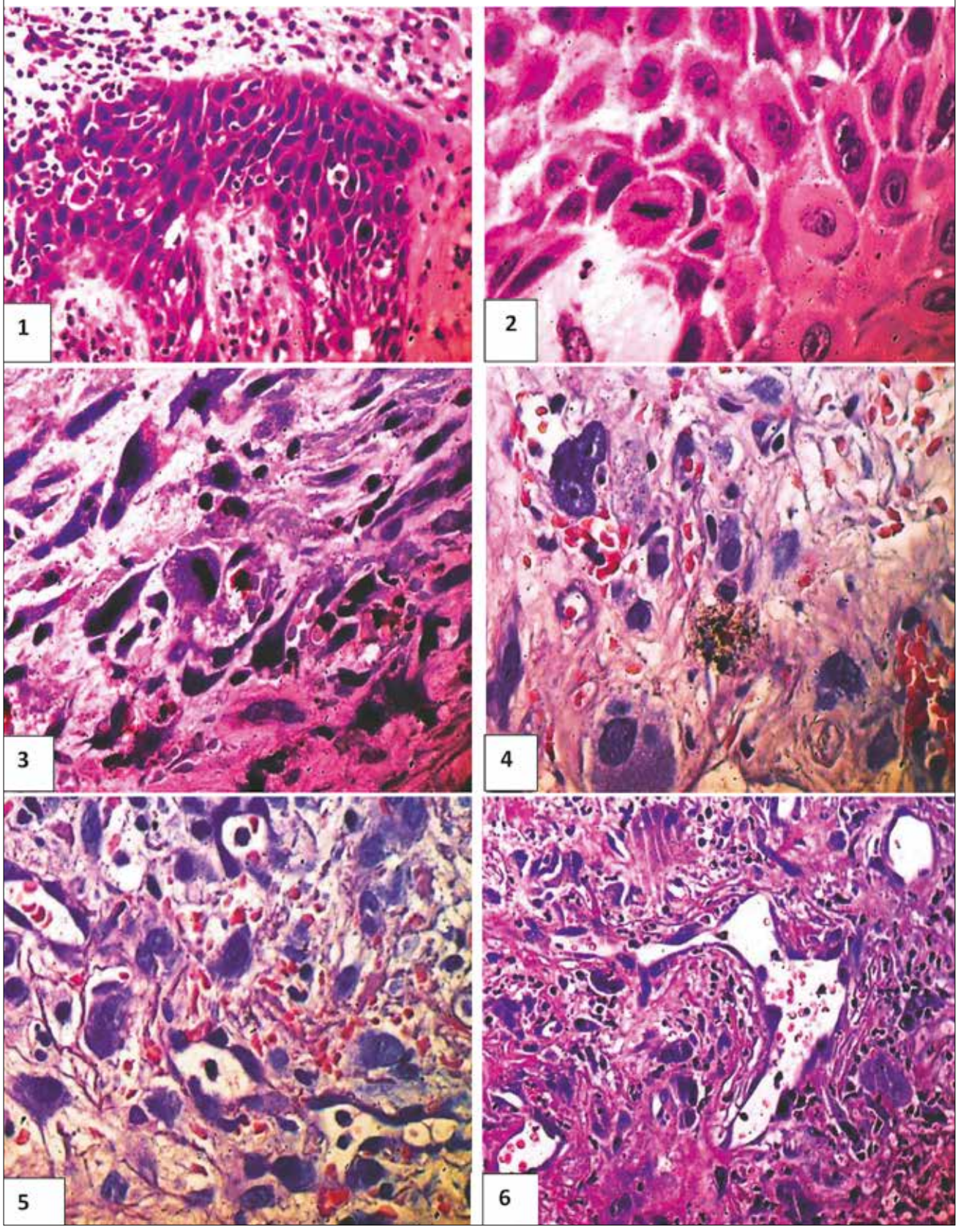
Figs. 1, 2. Strongly atypical superificial squamous epithelium, with the characters of in situ carcinoma. **Figs. 3, 4.** Stromal atypical elements: spindled, ganglion-like, multinucleated Touton-type. **Figs. 5, 6.** Thrombosed vessels with prominent endothelium.



nent endothelium (Figg. 5, 6, 13, 14) and. by a brisk proliferation of pleomorphic, atypical elements: spindled, ganglion-like, multinucleated Touton- type (Figg. 3, 4). The mitotic activity was sporadic, sometimes atypical. The immunohistochemical analysis shows immunoreactivity for epithelial markers (cytokeratins; EMA) restricted to the overlying squamous epithelium, while the atypical, pleomorphic stromal elements showed diffuse positivity for vimentin, alpha-smooth muscle actin, calponin CD68 CD34, and CD31 P63 and Ki67, staining was limited to the squamous epithelium.

Discussion

In 1957 N. Lane reported a series of 10 cases of polypoid lesions (Sarcoma-like) associated with carcinoma of the mouth, fauces and larynx. "These had in common the presence of an intramucosal or invasive squamous carcinoma, which was often incospicuous and easly overlooked, associated with an anaplastic stroma of sarcomatous appearance, which was often bulky and preponderant… In the author's cases the "sarcomatous" stroma is regarded as a secondary reactive phenomenon and per se is interpreted as probably non-neoplastic" [1].

Since that publication, Pubmed lists 62 reports concerning this lesion. Of these 34 had esophageal location, 28 pharyngo-laryngeal, 2 lingual [2 11], 2 anal [12 13], 1 oral [5] and 1 Cervical uterine [9].

To date no reports of this type of lesion on the external male genitalia are present in the literature.

The distinctive features of these lesions are: the polypoid appearance the surface epithelium of non- keratinizing squamous type, the ulceration, the coexistence of a squamous cell carcinoma, usually in situ or minimally invasive, the predominant localization at the level of the upper aerodigestive tract.

It should be emphasized, however, that the other locations, although rarely, may be mucosal membranes with a structure similar to those of the upper aero-digestive tract.

Over the years there has been an active debate about the meaning of the "sarcomatous" component.

Some authors, following the opinion of Lane, considered it as a reactive process, while others as as neoplastic in nature (sarcomatoid carcinoma). With regard to the lesions of the pharyngo-laryngeal district, a deeper analysis of the abstracts points out that about 50% of the authors are oriented towards the diagnosis of sarcomatoid carcinoma or carcinosarcoma, while the remaining 50% are inclined to believe that the "sarcomatous" component is of reactive nature of [1 3-5 7 9 11 22].

As far as the esophageal lesions are concerned, 60% of the authors are oriented towards a diagnosis of sarcomatoid carcinoma or carcinosarcoma, 32% tend to consider it as a reactive lesion [8 15-20 23].

Actually a careful evaluation of the morphological data allows us to differentiate the lesion described by Lane from the true carcinosarcomas and sarcomatoid carcinomas. The former has always polypoid morphology, it occurs in areas covered by non keratinising stratified squamous epithelium, and it is not associated with a non- or mini-infiltrating epithelial, proliferative lesion. The sarcomatoid carcinoma is usually widely infiltrating. These concepts were clearly expressed, several years ago, in a previous publication by one of the authors of this report [3].

The debate lasted until the advent of the immunohistochemical survey making it possible to separate the true carcinosarcomas from sarcomatoid carcinomas, from reactive lesions, although, in truth, in the most recent literature the problems do not seem completely solved. This happens, in our opinion,when those morphological and histogenetic differences, between different pathological entities, which we have specified above, are not clearly borne in mind.

An exhaustive description and immunohistochemical evaluation of the sarcomatoid carcinoma of the penis is found in a report [21] on the study of 15 cases. The macroscopic description is that of a vegetating (fungantig) tumor, often warty and largely infiltrating. In this tumor the sarcomatoid elements coexpress epithelial (cytokeratin, EMA) and mesenchymal (vimentin) markers, in the absence of myogenic and angiogenic markers. In our case the profile is quite different as the so-called "pseudosarcomatous" component is entirely negative to the epithelial markers, while it is polimmunophenotypic in nature, being composed of fibroblasts, myofibroblasts, endothelial cells This cellular composition perfectly replicates that of a granulation tissue.

In the final analysis the immunohistochemistry fully confirms what was suggested by Lane in his seminal report.

Conclusions

We herein report a unique case of "pseudosarcomatous" lesion on the external male genitalia.

In our opinion we think that the process arises as a result of a pathological process involving squamous epithelium, including (dysplasia/ca in situ) or minimally invasive carcinoma. This condition is responsible of epithelial erosion/ulceration which induces, in the subepithelial stroma, the formation of a granulation tissue containing atypical fibroblasts, myofibroblasts and endothelial cells.

This confirms Lane's opinion that this is a reactive process that, could be better defined as an atypical granulation tissue in the variegated world of pseudosarcomas, worthy of the eponymous of Lane's Tumor.

Fig. 7. Cytokeratin. **Figs. 8,9.** SMactin. **Fig. 10.** Calponin. **Fig. 11.** CD68. **Fig. 12.** CD34.

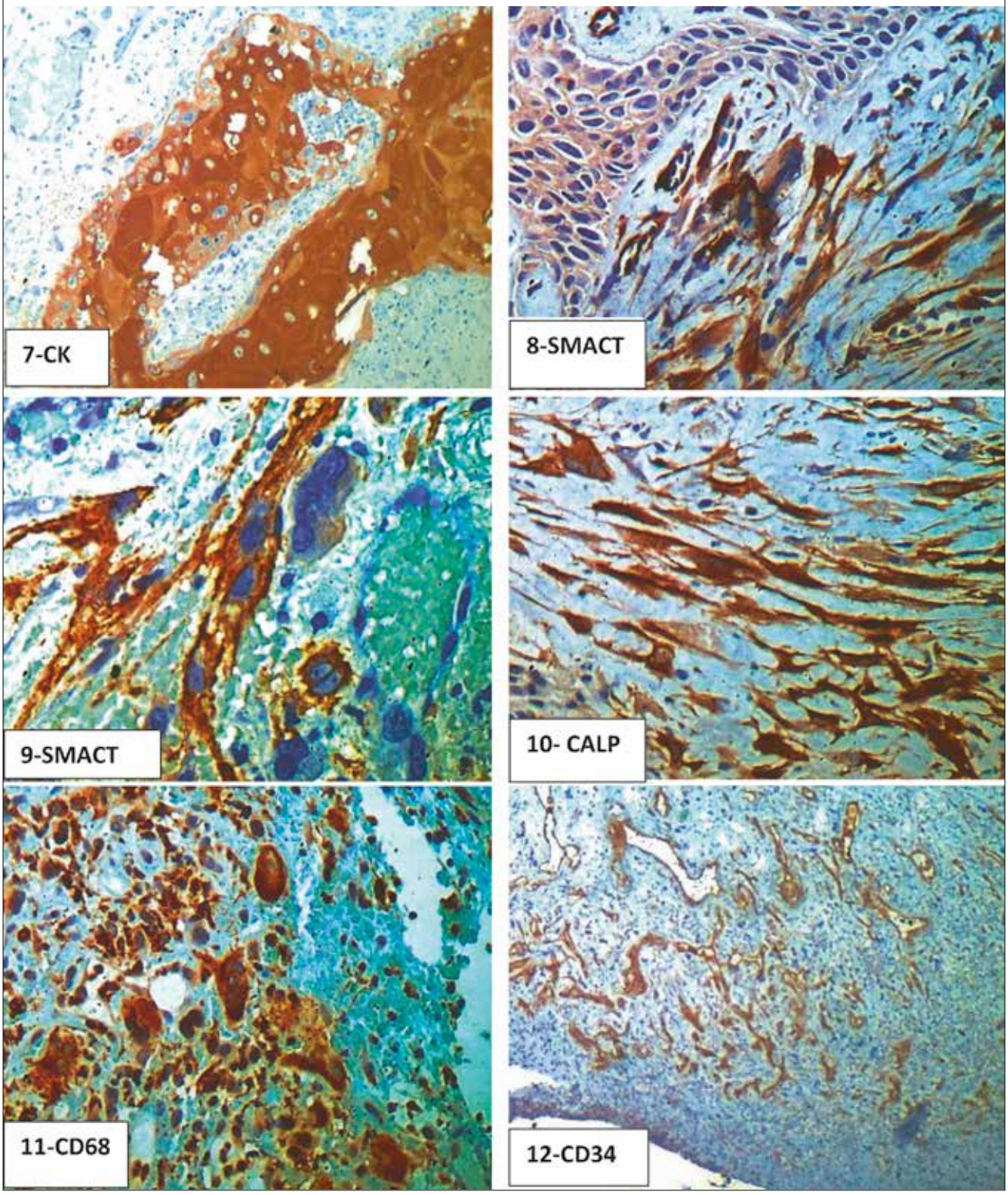
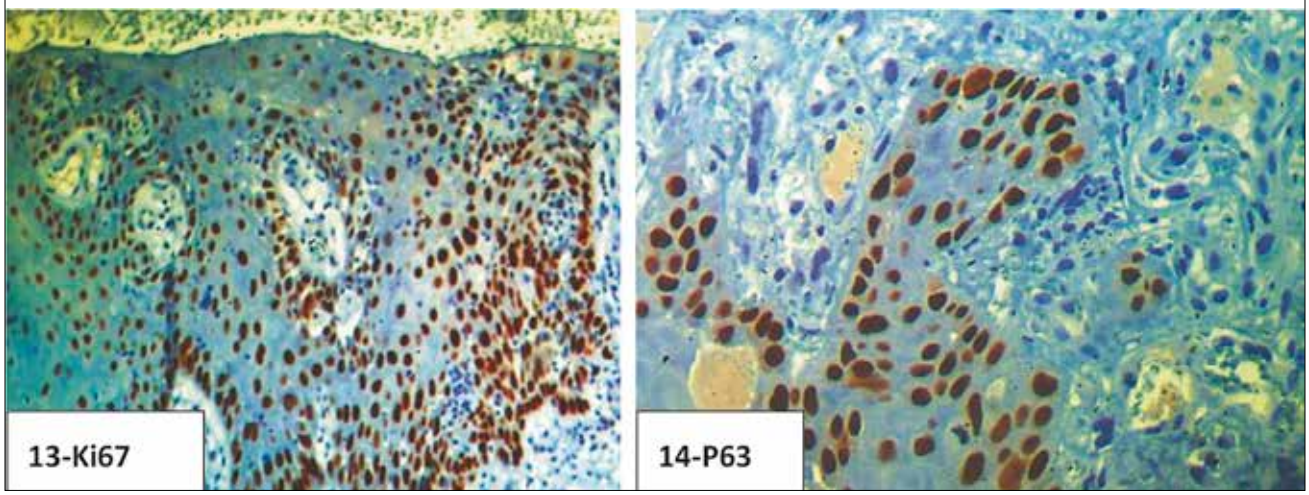
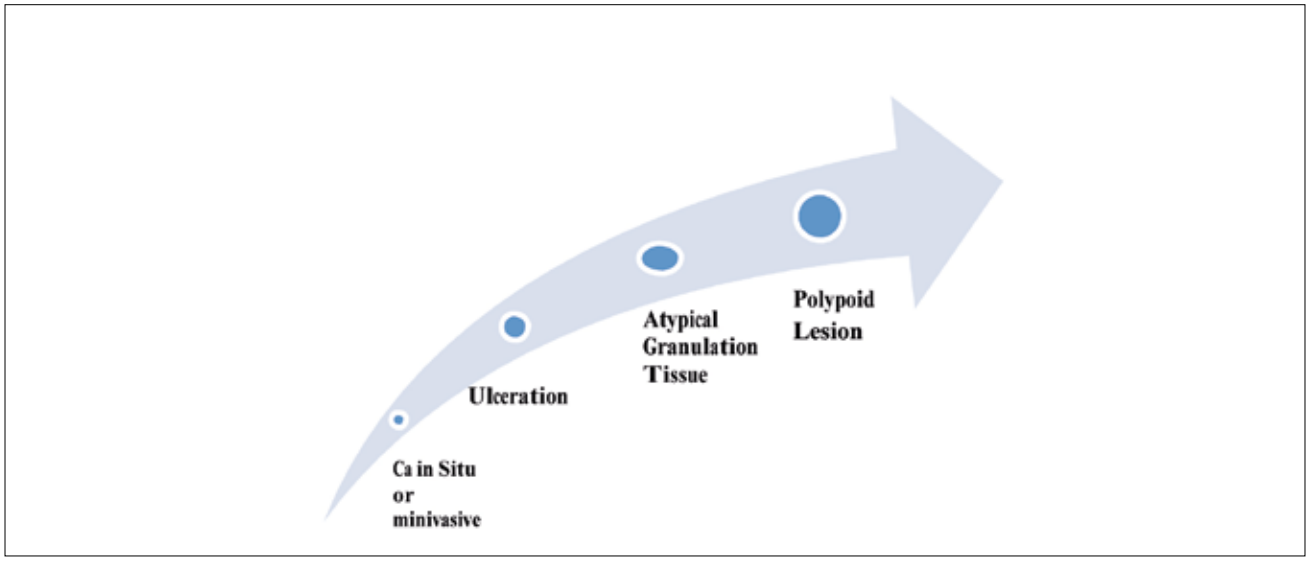


Fig. 13. KI67. **Fig.14.** P63.





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