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Abstract

Much of the bone, cartilage and smooth muscle of the vertebrate face is derived from neural crest (NC) cells. During craniofacial development, the anterior neural ridge (ANR) and olfactory pit (OP) signaling centers are responsible for driving the outgrowth, survival, and differentiation of NC populated facial prominences, primarily via FGF. While much is known about the functional importance of signaling centers, relatively little is understood of how these signaling centers are made and maintained. In this report we describe a dramatic craniofacial malformation in mice mutant for the zinc finger transcription factor gene *Sp8*. At E14.5 they show facial prominences that are reduced in size and underdeveloped, giving an almost faceless phenotype. At later times they show severe midline defects, excencephaly, hyperterlorism, cleft palate, and a striking loss of many NC and paraxial mesoderm derived cranial bones. *Sp8* expression was primarily restricted to the ANR and OP regions during craniofacial development. Analysis of an extensive series of conditional *Sp8* mutants confirmed the critical role of *Sp8* in signaling centers, and not directly in the NC and paraxial mesoderm cells. The NC cells of the *Sp8* mutants showed increased levels of